



CASO CLÍNICO/CASE REPORT

Síndrome de Cotard: Um Caso Clínico Raro Cotard's Syndrome: A Rare Clinical Case

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Abstract

Cotard's syndrome is a rare neuropsychiatric condition. It is a syndrome characterized by the presence of mood-congruent nihilistic delusions. The aim of the present paper is to review literature evidence concerning Cotard's syndrome and to describe a clinical case. Data were obtained from clinical records and bibliographic research on PubMed.

We present the case of a 68-year-old woman with Cotard's syndrome, in the context of a depressive disorder, to illustrate both how impairing the condition can be and how a course of effective, individualized therapy can improve outcome. Regarding treatment, the combination of antipsychotics and antidepressants is often used, but if this shows no improvement, electroconvulsive therapy is suggested.

There is a need for greater awareness of this syndrome, to ensure better and faster identification and treatment of patients.

Resumo

A síndrome de Cotard é uma condição neuropsiquiátrica rara. É uma síndrome que se caracteriza pela presença de delírios niilistas congruentes com o humor. O objetivo do presente trabalho é rever as evidências da literatura sobre a síndrome de Cotard e descrever um caso clínico identificado no nosso serviço. Os dados foram obtidos a partir do processo clínico de um paciente e através de pesquisa bibliográfica no PubMed.

Apresenta-se o caso de uma mulher de 68 anos, com síndrome de Cotard, no contexto de um transtorno depressivo, para ilustrar como este quadro pode ser prejudicial e desafiante e, de que modo a terapia individualizada e eficaz pode melhorar o quadro. Em relação ao tratamento, a combinação de antipsicóticos e antidepressivos é frequentemente utilizada, mas se não houver melhoria, sugere-se a eletroconvulsivoterapia.

É necessária uma maior consciencialização desta síndrome, que garanta uma melhor e mais rápida identificação e tratamento dos pacientes.

Keywords: Delusions; Electroconvulsive Therapy; Psychotic Disorders/drug therapy; Psychotic Disorders/therapy

Palavras-Chave: Delírios; Eletroconvulsivoterapia; Perturbações Psicóticas/tratamento; Perturbações Psicóticas/tratamento farmacológico

INTRODUCTION

Cotard's syndrome is a rare neuropsychiatric condition. It was first described, in 1880, by Jules Cotard, a French psychiatrist, as "negation delirium" and the term "Cotard's

syndrome" was proposed, in 1893, by Emil Regis.¹ Described as an autonomous systematized negative delusion, characterized by hypochondriacal and nihilistic ideas of denial of the functioning or existence of body organs,

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sometimes accompanied by ideas of immortality. This author describes delusion as symptomatic of a particular form of depressive illness that would include ideas of guilt, insensitivity to pain and suicidality.

These patients habitually rejected food due to the denial of the different digestive organs. In the forms most observed in clinical practice, an incomplete Cotard's picture appears, reduced to hypochondriac complaints attributed to the malfunction and occlusion of the organs, which fundamentally have to do with the digestive tract and abdominal viscera, and less frequently with the organs of respiration and circulation.² The author described a particular type of delusional formation, which reported in a considerable number of melancholic patients, especially in those who showed morbid anxiety. According to the author, these patients showed a systematic attitude of denial, accompanied by ideas of guilt, ruin and condemnation. They accused themselves of being useless, a source of unhappiness for the family, a source of infectious contamination or bad luck for those who met them. The attitude of denial could manifest itself from lighter forms, such as denying the existence or functioning of certain organs, to the belief of being dead, denying its existence, or even reaching the extreme of denying the existence of the external world.³

The syndrome is usually encountered in middle-aged or older people and is more common in females.^{3,4} Advanced age has been found to increase the likelihood of developing Cotard's syndrome.⁵

Cases have been reported in patients with mood disorders, psychotic disorders, and medical conditions. Though most found in unipolar depression, bipolar depression and schizophrenia, it has also been found in organic conditions such as neurosyphilis, epilepsy, parietal lobe tumors, Parkinson's disease and multiple sclerosis.⁶ Severe depression is the condition most often associated with this syndrome.⁷ There is growing consensus to consider Cotard's syndrome and its typical nihilistic delusions as a symptom of an underlying disorder. Nihilistic delusions are reported as an example of mood congruent delusions of a depressive episode with psychotic features.⁸

The duration of syndrome can vary from weeks to years depending on the underlying disorder.⁷ One of the consequences of this syndrome is self-starvation because of negation of existence of self.⁴ Patients are mostly found in advanced state, in severe psychomotor retardation with impairment in biological functions, requiring electroconvulsive therapy. Interestingly, contrary to their presentation, prognosis is good.⁶

Although it is not a diagnostic entity in our current psychiatry practice, knowledge on Cotard's syndrome and specific approach to treating patients is valuable. Larger-scale research is needed to clarify the pathophysiologic mechanisms that underpin this condition.

CASE REPORT

A 68-year-old, female, married. Lives with her husband and son. Studied until the 4th year of school and is retired. Medium-low socioeconomic level. Personal history of

arterial hypertension and type 2 diabetes mellitus. She had a previous medical history of recurrent depressive disorder. No relevant family history. Medicated with sertraline, diazepam, losartan, and metformin.

She was brought to the urgency, by her son, with complaints of insomnia, anhedonia, food refusal, anxiety, anorexia, and lack of strength, with two months of evolution. A month before, she started to believe that her body was putrid, the organs did not work anymore, could not eat, or do anything else because "the muscles did not work".

Upon clinical examination, the patient was awake, oriented in person, partially oriented in time and space, with deeply depressed mood, psychomotor retardation, bradypsychic thinking, apathy, decreased socialization, loss of appetite and subsequent weight loss of 8 kg, poor self-care, ruminative thoughts of death and nihilistic delusions concerning her own body. She had delusions of ruin and needed help to perform basic daily tasks, such as eating and personal hygiene. Physical and neurological examination without major changes. Organicity screening was carried out with analytical routines (blood count, renal function, ionogram, hepatic and thyroid function, viral serology, folic acid and vitamin B12 dosage), as well as chest x-ray and electrocardiogram, without alterations. A head computed tomography and electroencephalogram were done and showed no pathological findings. There was no personal history of substance abuse. The patient refused care and had no insight regarding her clinical condition. In accordance with the Portuguese Mental Health Law, she was admitted involuntarily to the inpatient Psychiatry unit, for clinical stabilization.

The patient presented a Cotard's syndrome inserted in a depressive condition. The current diagnostic classifications (DSM-5 and CID-11) omit this syndrome, confirming the tendency to deny its nosological entity. According to CID-11, the patient was diagnosed with recurrent depressive disorder, with psychotic symptoms.

Initial treatment consisted of fluoxetine 20 mg/day, mexazolam 1 mg/day and olanzapine 10 mg/day. After 7 days, fluoxetine was increased to 40 mg/day. After 2 weeks there was no therapeutic response, and her depressive symptoms were aggravated. Due to poor response, she started venlafaxine 150 mg/day, plus mirtazapine 15 mg/day and risperidone 2 mg/day. Fluoxetine and olanzapine were stopped.

Few alterations were noted (improved appetite and resolution of insomnia) and so, after 2 weeks, clomipramine 25 mg/day was added and then incremented to 50 mg/day a few days after. Risperidone was reduced to 1 mg/day and mirtazapine was stopped.

At this time, the patient showed improvement in her clinical status and began to feed properly and take care of her personal hygiene. She maintained treatment with clomipramine 50 mg/day, venlafaxine 150 mg/day and risperidone 1 mg/day. She also started cognitive behavioral psychotherapy and weekly activities at the Day Hospital. After a week, there was a progressive and substantial improvement of the clinical status. At discharge, she was oriented, cooperative, with euthymic mood, resolution of anhedonia, anxiety, and lack of strength. The delusional

ideas of ruin and other psychotic symptoms were in remission. Recovery of appetite, sleep, and ability to perform basic daily tasks. She developed partial insight regarding the episode. The patient was discharged 7 weeks after admission and referred to psychiatric outpatient treatment.

DISCUSSION

Cotard's syndrome is characterized by anxious melancholia, negation delusion (individuals feel major changes in their bodies and deny the existence or function of one or several parts of their organs or bodies) and nihilistic delusion (individuals believe that they or all people are dead).^{4,6,9} The most prominent symptoms are depressive mood, nihilistic delusions concerning one's own body and one's own existence, delusions of guilt, immortality and hypochondria.^{2,10}

Studies found that the most prominent symptoms are depressive mood (89%), nihilistic delusions concerning one's own existence (69%), anxiety (65%), delusions of guilt (63%), delusions of immortality (55%) and hypochondriac delusions (58%).⁸

No quality data are available on the prevalence and incidence of Cotard's syndrome. Studies suggest a prevalence of 0.57%-3.2%.⁸

The first evidence-based classification was made by Berrios and Luque in 1995.⁸ They described three types of Cotard's syndrome: Psychotic depression (patients with melancholia and nihilistic delusions), Cotard type 1 included the pure forms of nihilistic delusions without affective symptoms and Cotard type 2 with a mixed group of symptoms of anxiety, depression and auditory hallucinations.^{4,11} It is also generally said that symptoms of Cotard's syndrome develop over the period. Three stages of development of Cotard's syndrome have been proposed: germination stage, blooming stage and chronic stage.⁴

The germination or prodromal stage is characterized by hypochondriasis and cenesthopathy. The blooming stage involves development of full symptoms of nihilistic delusions and chronic stage involves chronic changes in mood and systematization of delusions.^{4,12}

Very little evidence has been described regarding the biological basis of the typical delusions of this syndrome. Structural, metabolic and toxic changes have often been associated, as well as changes in regional cerebral blood flow, but a causal relationship has never been perfectly established.

Critchley *et al* presented an overview of bizarre psychiatric syndromes that were related to parietal brain disfunction. In this work, the role of premorbid personality characteristics in these bizarre syndromes was crucial. On one hand, for Cotard's syndrome, patients with a more internal attribution style, which often co-occurs with depression, are more likely to develop the syndrome. On the other hand, patients with a more external attribution style, which more often co-occurs with paranoia, should have a better chance of developing Capgras' syndrome⁸ (Capgras' syndrome is an identification delusion characterized by the belief that someone close has been replaced by an "imposter"). Therefore, the modern "two-stage" psychopathological model explaining both

syndromes would rely on a first stage constituted by the abnormal perception – most likely due to lesions or dysfunctions in particular brain areas deputed to process emotions recognition – and a second stage that is essentially based on one own's typical attributional style signature, which tends to project externally in Capgras' syndrome (paranoid style) and internally in Cotard's syndrome (depressed style).⁸

It is thought that the origins of both syndromes are related to a dysfunction in an information-processing subsystem in which face and body recognition are associated with an effect of familiarity. When the normal felling of familiarity is absent, patients may experience an unusual felling of derealization and depersonalization. Empiric research on this disturbance in information processing is lacking for Cotard's syndrome but it exists for Capgras' syndrome.⁸ However, this model does not explain why some patients only deny the existence of parts of the body and not the Self or deny the existence of objects or people in the extra personal space. It also does not allow to explain the simultaneous manifestation of typical Cotard's syndrome delusions and Capgras syndrome observed in some cases.³ In terms of neurobiological evidence, many studies suggest lack of gross abnormalities in the neuroimaging. However, studies which have found neurobiological changes suggest involvement of fronto-temporo-parietal circuitry in the pathophysiology of this syndrome.⁴ There seems to be also a correlation between prefrontal dysfunction and nihilistic beliefs in neurological patients.¹³ Studies seem to delineate a neurobiological framework of CS in which the associative areas deputed to control the complex processing of psychic and somatic self, as well as the consciousness, appear as hypofunctioning with a consequent hyperactivation of basal ganglia and thalamic areas, where emotional and somatosensory signals are elaborated.

In line with this vision, some authors have proposed that CS might derive from a substantial disconnection of the sensorimotor-elaborating areas from limbic regions, therefore, determining a complete lack of emotional processing of the environment and then the sensation of being dead.¹⁴ Since 1880, similar cases have been reported worldwide and many authors have written about its possible etiology, some seeing Cotard's syndrome as a syndrome, others as an independent entity, many including it in the psychotic disorders, but with the majority still insisting that it belongs to the affective spectrum.⁷

The nosographic figure of Cotard's syndrome remains unclear and is not classified as an isolated disorder in DSM-V neither in ICD-11; at DSM-V nihilist delusion is classified as delusion congruent with the humor inserted in a serious depressive episode with psychotic symptoms.^{8,11,14} This reflects the current status of Cotard's syndrome, which is conceptualized as part of an underlying disorder, mostly a depressive episode.⁸

Although not a separate diagnostic entity, the phenomenologic study of rare monothematic delusional disorders remains important to have a better understanding of the pathophysiology of delusions.¹³

Treatment of Cotard's syndrome should focus on the underlying condition.¹ It is not clear which treatment should

be considered as the first choice.¹¹ Treatment of Cotard's syndrome secondary to an underlying mood disorder with ECT has been observed to be extremely effective.⁹ Even though ECT has been the treatment most frequently indicated in the literature, some reports of the combined use of antipsychotic and antidepressant drugs can also be found when psychotic depression is the underlying illness.¹ It is described the combined use of imipramine and risperidone with effective results in remitting psychotic depression symptoms.¹ The use of drug therapies with ECT is thought to be most effective in most of the cases who did not respond well to drug therapy alone.⁷ The prognosis seems to

be determined mostly by the treatment options and prognosis for the underlying disorder.

Our case here described was compatible with the different descriptions available in the literature for this syndrome, with features such as a depressed mood, nihilistic delusion and delusions of guilt.

Although rare, Cotard's syndrome is still a present phenomenon in the psychiatric clinics all over the world and it involves big suicide risk, because of that, it is recommended to institute treatment as early as possible.¹⁵ It is important that the clinicians be aware to the many possibilities of manifestation of this syndrome.

Prêmios e Apresentações Prévias

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