

Cotard's Syndrome: Two Cases of Self-Starvation

Síndrome de Cotard: Dois Casos de Recusa Alimentar

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RESUMO

Introdução: A Síndrome de Cotard é uma condição clínica relativamente rara que se caracteriza por vários graus de delírios nihilistas, quase sempre na forma de auto-negação.

Objectivos: Descrever dois casos de Síndrome de Cotard associados a recusa alimentar e realizar uma revisão do conceito e das características clínicas desta síndrome.

Métodos: Realizou-se a recolha de informação de dois casos clínicos associados a recusa alimentar. Procedeu-se a uma revisão do conceito e das características clínicas que lhe estão associadas.

Resultados e Conclusões: O primeiro caso versa sobre uma mulher que acreditava que o seu esófago e estômago estariam colados. Foi medicada com sertralina, mirtazapina e risperidona, com bons resultados. O segundo caso descreve um homem que acreditava que a sua garganta fora queimada e que não possuía nenhum órgão interno. Foi medicado com clomipramina e risperidona, mostrando grande melhoria. Esta síndrome é uma entidade nosológica e clínica que não deve ser esquecida. É essencial fornecer uma abordagem terapêutica urgente e adequada em pacientes com esta síndrome.

Palavras-Chave: Síndrome de Cotard; Depressão; Delírio Nihilista.

ABSTRACT

Background: *Cotard's syndrome is a relatively rare condition characterized by various degrees of nihilist delusions, often in the form of self-negation.*

Aims: *To report two cases of Cotard's syndrome associated with self-starvation and to review the concept and clinical features of the condition.*

Methods: *Two clinical cases of the syndrome were obtained and a literature review of the theme was shortly surveyed.*

Results and Conclusions: *The first case is about a woman who believed that her esophagus and stomach were glued. She was treated with sertraline, mirtazapine and risperidone with good results. The second case describes a man who believed his throat was burnt and he had no internal organs. He was treated with clomipramine and risperidone showing great improvement. This syndrome is a nosological and clinical entity that should not be forgotten. It is es-*

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sential to provide an urgent and adequate therapeutic approach to these patients.

Key-Words: *Cotard Syndrome; Depression; Nihilistic Delusion.*

INTRODUCTION

Few pathologies of the self are as profound and striking as those reported in cases of Cotard's syndrome, which can involve the belief that one is dead. The assertions of some patients with this delusion come close to violating the famous Cartesian dictum "cogito ergo sum". Descartes explored the limits of radical skepticism and concluded that whereas one could certainly doubt the evidence of one's senses, it was not possible to doubt one's existence. Yet some Cotard patients maintain that they are dead or that they do not exist^{1,2}.

The classic reports of this condition were published by the neurologist Jules Cotard³, who described a clinical state that he termed *délire des négations*. The French eponym *délire de Cotard* was later adopted and translated into English as Cotard's syndrome⁴. Although this later designation is often identified with the belief that one is dead, it must be underlined that the condition is a syndrome and not a symptom as it is often incorrectly conveyed in literature. Cotard himself did not regard that belief as an essential defining feature of the condition he described^{1,5}. Young and Leafhead's analysis of Cotard's cases revealed a series of commonly occurring features and symptoms, including self-deprecatory delusions, suicidal ideation, feelings of guilt, and denial of body parts. Young and Leafhead's subsequent comparison of three patients with

the belief that they had in fact died revealed a consistent combination of additional symptoms including depressed mood, abnormal feelings, depersonalization and derealization, and evidence of face-processing impairments. More exotic concurrent symptoms have been reported elsewhere, including hydrophobia⁶ and lycanthropy^{2,7}.

During the 20th century, it continued to attract the attention of diagnostic phenomenologists. Currently, many investigators of the condition conceptualize it as at least a component of a mental disorder, frequently a major depressive disorder⁴. However, the issue of whether the Cotard's phenomenon constitutes a unique mental disorder, a discrete syndrome (associated with a range of conditions such as depression, psychosis, organic conditions and so on), or merely a psychiatric symptom remains unanswered^{1,2}.

Berrios and Luque, in an extensive review of the conceptual history of Cotard's syndrome, concluded that Jules Cotard probably viewed it as a subtype of melancholia (anxious melancholia)⁵. In an attempt to further understand the phenomenology of the condition, Berrios and Luque, using an exploratory factor analysis of 100 cases of Cotard's syndrome reported in literature, extracted three factors: psychotic depression, Cotard type I and Cotard type II⁴. The psychotic depression patients mostly had depression and few nihilistic delusions. Cotard type I patients on the other hand, had only the nihilistic delusions (pure Cotard's syndrome) and few depressive symptoms, whereas Cotard type II patients were a mixed group with depression, anxiety and auditory hallucinations¹⁰.

In the same study, Berrios and Luque diagnosed depression in 89% of the 100 cases. The most frequent nihilistic delusions were related to the body (86%) and with existence (69%). Anxiety (65%) and guilt (63%) were common, followed by hypochondriacal delusions (58%) and immortality delusions (55%)¹¹.

There are several proposed mechanisms for Cotard's syndrome. It may begin with anomalous perceptual experiences or their disconnection from emotional or limbic processes, and there may be a failure in belief evaluation or a tendency to negative self-attribution^{2,8,9,12}.

Many patients have psychiatric disease with psychotic depression, an internalized attributional style, or associated depersonalization².

It is worth emphasizing that the syndrome is best conceptualized as being on a spectrum (complete/incomplete): the complete form in which nihilistic delusions are clearly present and the incomplete forms which are often combinations of depressed mood, delusions of guilt and hypochondriasis, and hallucinations. Also the nihilistic delusion itself could vary in its degree of severity – from severe (patient denies his own and the world's existence) to mild (patient feels that he is losing his reasoning and feelings). Yamada et al.¹³ attempted to trace the onset and longitudinal progression of the condition from a phenomenological perspective and identified three distinct stages: the germination stage (prodromal period associated with depression and hypochondriacal symptoms), the blooming stage (full blown development of the syndrome with delusions of negation) and the chronic stage (chronic depressive type or chronic delusional type). They equated the above three stages to

the earlier classification of the syndrome by Berrios and Luque as follows: the germination stage corresponds to psychotic depression, the blooming stage to Cotard type II and the chronic stage to Cotard type I¹⁰.

The current diagnostic classifications (DSM-5 and CID-10) exclude Cotard's syndrome, confirming the trend to reject it as a nosological and clinical entity¹⁴.

The aims of this article are to describe the clinical presentation and the management of two cases of Cotard's syndrome and to review the concept and clinical features of the condition.

CASE STUDY 1

The first case we report is about a 66-year-old woman that will be addressed as Mrs. H.

Mrs. H. had no psychiatric background up until April 2012, when she was admitted to our department after developing a clinical picture of depressed mood and progressive onset of delusions of ruin, ill health, guilt and self-starvation. The patient started experiencing these symptoms after her husband's unexpected death, approximately six months prior to admission.

Mrs. H. had a medical history of hysterectomy, cholecystectomy and appendectomy. There was no history of substance misuse and she was taking no medication when admitted to our inpatient ward.

Mrs. H. lived in Gaia for years in a symbiotic relationship with her husband. Their sons were living in far distant cities, so they would only be present occasionally supporting their parents the way they could. After her husband's sudden death, Mrs. H. couldn't face his absence and the solitude. She started feeling

depressed, describing insomnia, anorexia and intense anguish. A month later she developed delusions of ruin, believing her pension wasn't enough to ensure her subsistence and alleging her refrigerator wasn't working well because the food had a bad taste and was constantly becoming deteriorated, which she continued stating even after a technician confirmed there was no problem with it. She also described that she had no decent clothes because they were dirty and shabby. At the same time, she developed olfactory hallucinations, describing herself as malodorous and refusing to leave the house. She considered that this was all her fault revealing a very prominent feeling of guilt.

A month later, the clinical picture worsened significantly as Mrs. H. started believing that her esophagus and stomach were glued together, so that it was not possible for her to ingest any food. She was perfectly convinced that she had been through some kind of organic disease that was in the basis of the collage and that this condition made her incapable of ingesting any food. In this context, she started a process of self-starvation that led to a loss of 33 pounds in one month. She would only ingest liquids occasionally describing pain while eating. By the time this happened, her daughter travelled to Gaia and found her mother a lot thinner. She took Mrs. H. to her general doctor and batteries of exams were carried out, not revealing any physical pathology. As Mrs. H. maintained her ruin and guilt beliefs and con-

tinued refusing to ingest any solid food, her daughter took her to a psychiatric emergency where she was transferred to our unit.

As she was admitted to our department, she looked very thin (she had lost 42 pounds in the meanwhile and her BMI was 14 by the time). She showed no *insight* regarding her clinical condition.

She was submitted to a rigorous investigation, including cranial computerized tomography, hematologic and biochemical routine tests and a neuropsychological evaluation. Nothing was detected in CT scan and the analytic study showed an irrelevant change of the lipid profile with no use of drugs detected. The MMSE and the clock drawing test didn't reveal any deficit (she punctuated 23/30* in the MMSE and she had studied for 6 years).

On transfer to our unit (day 0), her initial therapeutic regimen included sertraline 50 mg (once a day), risperidone 2 mg (once a day) and bromazepam 1,5 mg (trice a day). As there was no improvement to her clinical condition, after day 10 we increased sertraline to 100 mg (once a day) and risperidone to 4 mg (once a day). Mrs. H. was already eating by the time, though in small intakes. Still, there was poor improvement concerning her mood and psychotic beliefs, so at day 20 we added mirtazapine 30 mg (once a day) and increased risperidone to 6 mg (twice a day). In the following days, Mrs. H. progressively improved, diluting the

* The MMSE scale adapted for the Portuguese population considers the existence of cognitive impairment when the patient punctuates 15 or less and is illiterate, 22 or less and has between 1 and 11 years of study or even if the patient punctuates 27 or less and studied for more than 11 years.

delusions of ruin and guilt and the Cotard's syndrome. She had great improvements concerning her mood and was discharged at day 30 with no symptoms at all and being medicated with sertraline 100 mg (once a day), risperidone 6 mg (once a day), mirtazapine 30 mg (once a day) and bromazepam 1,5 mg (trice a day). She went to live with her daughter and was referenced to our outpatient unit.

CASE STUDY 2

The second case we report is about a 59-year-old married man from Vila Nova de Gaia that will be addressed as Mr. A. and who has been retired since 2008, having worked his entire life as a teacher.

Mr. A. had a medical background of alcoholism dependence, being abstinent for 7 years. There was no history of other substance misuse. He had also been through two major depressive episodes in 2005 and 2006 (without evidence of psychotic symptoms) and was treated with antidepressants during hospitalization. After 2006, the patient discontinued the follow up as well as the medication.

In December 2011, five months prior to his admission to our department, the patient began to describe depressed mood, something he defined as "melancholy", having multiple somatic complaints related to the digestive system. There was no life-event associated with the beginning of the symptoms.

In early 2012, Mr. A. attended the general emergency room several times and was treated with fluvoxamine 100 mg (once a day), which he took irregularly and without any considerable improvement. Several tests were carried out in the primary care, including hematolog-

ic and biochemical routine tests, showing no relevant changes. Progressively, he developed a clinical picture of social isolation, neglect of personal hygiene and lack of pleasure in daily activities.

In March 2012, the clinical picture worsened considerably, as Mr. A. started denying himself and believing he had no healthy organs. Accordingly, he completely refused to eat, being in self-starvation until he was admitted to our department, one month later. He lost a significant amount of weight (approximately 22 pounds, having a BMI of 18) in a one-month period. Mr. A. explained his refusal to eat claiming his throat was completely burnt and had no function at all, also stating he couldn't breathe, describing shortness of breath, nor swallow any aliments because his larynx was rotten. He believed all his organs were destroyed except for his brain and heart. His speech was centered in this nihilistic delusion of being clinically dead.

One week prior to admission, Mr. A. developed total insomnia and intense anguish. Two days before he was admitted to our department he ran away from home completely adrift.

In April 2012, when his wife took him to the psychiatry emergency room for the last time, he presented himself restless, *distressed* and tearful while describing his nihilistic beliefs. He was uncooperative and rejected hospitalization, showing impaired judgment, arguing he was already dead and had no psychiatric problem. Considering there was no *insight* and that the patient's condition was life threatening, he was involuntarily hospitalized in our department.

On transfer to our unit he was submitted to a detailed investigation, including CT scan, hematologic and biochemical routine tests and a neuropsychological evaluation. Nothing was detected in CT and the analytic study showed no relevant changes. No substance misuse was detected. The MMSE and the clock drawing test didn't reveal any deficit (he punctuated 28/30* in the MMSE and had a university degree).

His initial therapeutic regimen included a very slow intravenous infusion of clomipramine that lasted for 9 days and was progressively increased to a maximum dose of 150 mg. This infusion was coupled with an oral dose of clomipramine 75 mg (three intakes of 25 mg at the main meals) and risperidone 2 mg (once a day). We also added flurazepam 30 mg (once a day) because of the persistent insomnia. On day 10, Mr. A. was already eating and in the following weeks he showed great improvement of his psychotic and mood symptoms.

He was discharged on the 24th day to outpatient follow-up in voluntary regimen, being medicated with clomipramine 150 mg (two daily intakes of 75 mg), risperidone 2 mg (once a day) and flurazepam 30 mg (once a day).

DISCUSSION AND CONCLUSIONS

Clinical states of the Cotard's syndrome are rarely encountered today, most probably due to the swift treatment of the psychotic disorder with the present medical treatment and/or the decrease in the number of institutionalized patients¹⁵. However, the two case studies presented report patients followed in 2012 at our department, which must be a warning that the

syndrome is a nosological and clinical entity that should not be forgotten.

No quality data are available on the prevalence and incidence of the condition. The only prevalence study was performed in a selected psychogeriatric setting in Hong Kong¹⁴.

Most of the cases have been reported in elderly patients⁴, being the mean age of patients 52 years, but is also occasionally described in children and adolescents¹⁴. Diagnosis of Cotard's syndrome in people less than 25 years of age is associated with bipolar disorder²⁰. The disorder is more common in women²¹.

Most cases are associated with major depressive disorder. However, Cotard's syndrome has also been noted to co-occur with other mental disorders, including schizophrenia, mental retardation, bipolar disorder, and disorders secondary to general medical conditions such as brain injury, seizure disorder, and typhoid fever¹⁶.

The two cases described are associated with major depressive disorder and both reflect what Cotard, in 1882, meant to say with the French term "*délire des négations*"^{17,18,19}.

Considering Berrios and Luque⁴ classification, it's hard for us to match the reported cases in any of their categories as both patients developed prominent depressive symptoms and nihilistic delusions, so they couldn't match the psychotic depression and Cotard type I categories. Also, it wasn't reported auditory hallucinations in any of the cases, which excludes the match with Cotard type II category.

However, we consider that both cases suit the incomplete form, as there is a combination of depressed mood, delusions of guilt and hypochondriasis. In this form of the syndrome the

hypochondriac complaints are often attributed to malfunction and occlusion of the organ, mainly digestive tract and abdominal viscera, and less frequently with the organs of respiration and circulation¹¹. At this point, the second case seems to be uncommon, as the patient was convinced he couldn't breathe because of the rotten larynx.

The fact that both cases came to psychiatric attention due to their refusal to eat is common. This refusal was noted by Cotard himself³.

The reported cases highlight the importance of an urgent therapeutic approach in patients with Cotard's syndrome who present with somatic and nihilistic delusions associated with self-starvation²².

We believe that in both cases the nihilistic delusions should be considered severe, according to the nihilistic delusion severity scale¹⁰.

Featuring the Yamada et al stages¹³, the reported cases went through the germination stage, as there was a prodromal period associated with depression and hypochondriac symptoms, and a blooming stage, because of the occurring of a full blown development of the syndrome with delusions of negation. Neither of them moved to the chronic stage.

Considering the second case, other issues may be discussed, besides the nihilistic delusions. Mr. A. had no *insight* concerning his condition. A person suffering from the syndrome can exhibit markedly impaired judgment, with important psychiatric forensic implications. In believing themselves they require no food and subjecting themselves to significant, sometimes life threatening weight loss, coupled with the lack of *insight* inherent in the Cotard delusion, individuals such as Mr. A. can

often meet the criteria for involuntary hospitalization¹⁶.

There are several reports of successful pharmacologic treatment of Cotard's syndrome.

Various authors recommend electroconvulsive therapy as the first choice for psychotic depression¹¹ and some reports have indicated the advantages of ECT associated with psychotic symptoms^{19,22,24}.

Madani and Sabbe suggested that ECT is an important option for patients with psychotic depression, while antipsychotics should produce better effects in Cotard's syndrome type I. Especially with an underlying mood disorder, ECT was reported as a very useful technique in many case reports. In most reports, ECT was followed by a pharmacologic maintenance treatment, usually combination strategies with antidepressant and antipsychotics¹⁴.

The second choice would be antidepressants²³, specifically the tricyclic, and antipsychotics¹². Monotherapy with agents such as amitriptyline, duloxetine, fluoxetine, paroxetine, olanzapine, sulpiride, or lithium has been reported to be effective. However, combination strategies often are used (clomipramine and amitriptyline; pimozide and amitriptyline; haloperidol and clomipramine; cyamemazine and paroxetine; risperidone and fluoxetine; haloperidol and mirtazapine; risperidone and sertraline; risperidone and citalopram; clozapine and fluvoxamine or imipramine)¹⁴.

In the reported cases, antidepressants and antipsychotics were used as a first choice with great improvements as the doses were increased to high levels.

There is little information suggesting a specific neuroanatomical basis underlying Cotard's

syndrome. However, a potentially relevant clue may be the fact that the affected person usually questions the authenticity of the self and/or the environment by insisting that what appears to exist does not in fact exist. This stance is reminiscent of cases of delusional misidentification phenomena where the authenticity of the self, others, or the environment is questioned¹⁶. Young and colleagues have proposed that the syndrome results from a similar anomalous perceptual experience to that putatively involved in the Capgras delusion. The available phenomenological and neuropsychological information for delusional misidentification and Cotard's syndromes suggests that non-dominant cerebral deficits may be a significant etiologic factor¹⁶.

The first imaging study on Cotard's syndrome suggested that was associated with multifocal brain atrophy and interhemispheric fissure enlargement²⁵. In general, there seems to be an important role for the frontotemporoparietal circuitry in the pathophysiology of it. Nevertheless, in most cases, gross structural changes on structural brain imaging were absent¹⁴, as seen in both cases presented. There are studies about SPECT in Cotard patients showing that asymmetric striatal D2 receptor binding favoring the left side (123 I-iodobenzamide SPECT). Nevertheless, absence of abnormalities in cerebral perfusion or metabolism patterns was also shown²⁶.

For the most part, there are no electroencephalogram abnormalities revealed, although nonspecific abnormalities and abnormalities suggestive of the underlying organic condition have been mentioned in some cases¹⁰.

Only a few reports mention detailed neuropsychological examination and, in these, normal recognition of emotional facial expressions is typically observed¹⁰.

Attempting to understand individuals with the condition from both a phenomenological as well as biological viewpoint may eventually pave the way for clearer connections between psychological and biological causation, as well as behavioral disability recognized by the law, in persons who meet criteria for involuntary civil commitment¹⁶.

Complete recovery may occur as spontaneously and suddenly as onset of Cotard's syndrome, even in the most severe cases²¹.

The overall prognosis seems to be determined mostly by the treatment options and prognosis of the underlying disorders.

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