International Neuropsychiatric Disease Journal
2(6): 348-355, 2014

SCIENCEDOMAIN *international*
www.sciencedomain.org

Cotard's Syndrome: Clinical Case Presentation and Literature Review

Alessandro Carano[1,2], Domenico De Berardis[2,3*], Marilde Cavuto[4], Carla Ortolani[4], Giampaolo Perna[5], Alessandro Valchera[6], Monica Mazza[7], Michele Fornaro[8], Felice Iasevoli[9], Giovanni Martinotti[2] and Massimo Di Giannantonio[2]

[1]Department of Mental Health, Psychiatric Service of Diagnosis and Treatment, Hospital "C. G. Mazzoni", NHS, 63100 Ascoli Piceno, Italy.
[2]Department of Neurosciences and Imaging, University "G. D'Annunzio", 66100 Chieti, Italy.
[3]Psychiatric Service of Diagnosis and Treatment, Department of Mental Health, "G. Mazzini" Hospital, NHS, ASL 4, 64100 Teramo, Italy.
[4]IASM, 67100 L'Aquila, Italy.
[5]Hermanas Hospitalarias, FoRiPsi, Department of Clinical Neurosciences, Villa San BenedettoMenni, Albese con Cassano, 22302 Como, Italy.
[6]Hermanas Hospitalarias, FoRiPsi, Villa S. Giuseppe Hospital, 63100 Ascoli Piceno, Italy.
[7]Department of Health Science, University of L'Aquila; 67100 L'Aquila, Italy
[8]Department of Formative Sciences, University of Catania, 95100 Catania, Italy.
[9]Laboratory of Molecular Psychiatry and Psychopharmacotherapeutics, Section of Psychiatry, Department of Neuroscience, University School of Medicine "Federico II," 80131 Naples, Italy.

Authors' contributions

All authors have contributed to the present paper with equal efforts. All authors read and approved the final manuscript.

Case Study

Received 28th September 2013
Accepted 2nd June 2014
Published 23rd July 2014

ABSTRACT

In 1880 French neurologist Jules Cotard described a condition characterized by delusion of negation (nihilistic delusion) in a melancholia context. Recently, there has been a resurgence of interest in Cotard's syndrome. The most prominent symptoms of Cotard's Syndrome are depressive mood, nihilistic delusions concerning one's own body and one's own existence, delusions of guilt, immortality and hypochondria. The aim of the present

**Corresponding author: Email: dodebera@aliceposta.it;*

paper is to review literature evidences concerning Cotard's syndrome and to describe a clinical case keeping in the background the recent trends on its psychopathological implications. In the clinical study, the following sequence of stages emerged: the dissociative side, expressed as a loss of body-mind cohesion; the 'mixed' mood disorder, with depressive-manic episodes, and a persecutory background, all coexisting in the anguish of the idea of a body falling apart, the anguish of a descent towards the abyss of melancholia and/or an ascent to unlimited euphoria, characteristic of an "uncommon alarm" for loss of Self cohesion.

Keywords: Cotard; nihilistic delusion; melancholia; negation; depression.

1. INTRODUCTION

In 1880 French neurologist Jules Cotard described a condition characterized by a delusion of negation with corporeal themes in a melancholic context [1]. At first, he formulated it as a new type of depression characterized by anxious melancholia, the idea of damnation or rejection, the insensitivity to pain, the delusions of nonexistence concerning one's own body, and the delusions of immortality [2]. Cotard categorized it as *Lypémanie*, a kind of psychotic depression described by Esquirol [3]. In 1882, he introduced the term *délire des négations* [4]: the first author who used the term Cotard's Syndrome was Séglas in his book «Le délire de Négation» [5].

Since then, the identification of this clinical entity has received multiple corroborations: however, the object of debate was whether Cotard's Syndrome may be a distinct disorder or a symptom of other disorders [6]. Cotard's purpose was to compare the delusion of negation to the delusion of persecution as stated by his mentor Laségue and he depicted its antinomic manifestation, the 'delusion of grandeur' [2] under the same perspective in 1888.

After some acknowledgments by Séglas [7], Regis [8] and Toulouse [9], several findings, although dissonant, have succeeded especially by French clinicians who, even with critical acumen, preferred to keep the traditional image [10-14]. Most recent studies about this "uncommon syndrome", have instead considerably drifted away from them, giving the lead to new psychopathological interpretations [15-20].

So the nosographical figure of Cotard's Syndrome remains unclear [21]. Cotard's Syndrome is currently not classified as a separate disorder in ICD-10 and DSM-IV. It is not determined if it pertains to the delusional themes area or it is related to the sense of immanent ruin in some depressive episodes: patients that belong to both psychotic areas may express experiences of somatic loss associated with psychomotor arrest, a kind of deep melancholic state [22]. For these reasons Cotard's Syndrome has recently been supposed to be an intermediate form [23]. Moreover, the majority of reports were based on single cases and few cohort studies were published [19-22].

In the present report, we describe a case of Cotard's Syndrome, discussing it from a psychopathological point of view.

2. CLINICAL CASE PRESENTATION

2.1 Case Histor

U. is a 45 years old woman who lives in a small hill town in Marche (Italy). She came to our observation at the Psychiatric Service of Diagnosis and Treatment of Civitanova Marche (Marche, Italy) in January 2009, with her husband, for the recurrence of dissociative symptoms with delusions of negation and guilt, ideation focused on the body with experience of corporeal disruption and affective flattening on a depressive background. Insight was present. At the admission, the anamnesis was collected with the help of both patient and her husband. The patient's father died of an acute myocardial infarction in 1989, at the age of 68. Her mother is 74: she suffers from a bilateral glaucoma and is described as a rigid, non affective person, particularly irascible «…she's depressed too, but has never wanted to be treated; she's always complaining and criticizing everything I do…».

The patient is an only child. At the present she's a housewife. In 1987, at the age of 23, after a 5 years long engagement she married. Her same-aged husband is a bricklayer, and he's apparently in good health. They have two children, apparently both in good health. After delivering her second child she had a uterine prolapse. Three years ago she underwent hysterectomy and vaginal reconstruction.

Her first pregnancy, strongly desired, was remembered as a pleasant experience. During the second pregnancy she refers to have had gestational problems since the 3rd month (threat of miscarriage), which attenuated during the following trimesters, but then she had an induced labor due to premature rupture of membranes. Through post-partum the patient reported a persisting melancholy with marked affective instability and somatic symptoms («I had high blood pressure, headache, I was depressed»). These notes weren't confirmed by U.'s husband, who, on his own, underlined the continuity with her personality traits, yet present before childbirth, with the exception of a pervasive state of tension for the baby's health.

However, despite these premises, the first psychopathologic episode appeared about a year and a half after the second child's birth, and was characterized by thought disorder, thymic and behavioral instability, hyperesthesia and delusions. The first contact with our local Psychiatric Service happened on April 1996, for an urgent consultancy. The psychiatrist on duty described the following psychopathological situation: "The ideation is focused on persecutory themes. Since the last 3-4 days, the patient has been presenting anxiety, irritability and she has been having difficulties in normal daily activities. Recently she has had the sensation of someone trying to poison her. She suffered from mixed insomnia." The psychiatrist prescribed her an essentially sedative psychopharmacologic therapy (delorazepam 1mg tid and levomepromazine 25mg bid). Considering the overstaying of a florid symptomatology, a few days later she was hospitalized on Obligatory Sanitary Treatment (TSO) regimen in our Psychiatric Service of Diagnosis and Treatment, where she remained from 05/02/1996 to 05/21/1996. Then, she was discharged with an ICD-10 diagnosis of "Paranoid schizophrenia" and a pharmacologic therapy (haloperidol 6mg/day and delorazepam 3mg/day).

After discharge, the patient was followed by our local Psychiatric Service, but she alternated periods of attendance, during which she respected appointment schedules and therapy, and periods of autarchic organization of therapies and care with private specialists. In July '99, a

psychiatrist she consulted privately, noticed an episode of dysphoria with interpretative features, and assigned her a therapy with an atypical antipsychotic (olanzapine 10mg/day). Over the years, up until now, she has continued the antipsychotic therapy (even if, during intercritical periods of relative wellness, she tends to abandon the drug), associated to phases of marked depression, with an antidepressant therapy (interchanging in different moments citalopram, paroxetine and fluoxetine). In 2005 she underwent a brain CT scan without contrast agent, which highlighted a "moderate increase of ventricular and cerebrospinal fluid spaces with no focal lesions of the parenchyma" and this is in agreement with that reported in the neuroimaging studies of Cotard's syndrome [18,19,24]. A constant in our patient's reminiscence is the presence of alternating depressive-dysphoric symptoms, anticipated by fleeting excitement around Christmas, Easter and summer holidays (mostly during August). During last Christmas holidays she showed signs of marked thymic instability associated with somatic delusions, anxiety of decay and insomnia, leading her to the present hospitalization.

2.2 Psychopathological Evidences

The day after hospitalization, U. appeared to be perplexed, rigid and blocked in her posture; she could perform few movements and looked as "petrified". Her face was amimic, her glance moved slowly and fixed on those who tried to communicate with her. Affectivity was constrained and inhibited, the moments of "communication" with the others were short and fleeting, the patient withdrew from any physical contact, refusing any form of relation with the outside world. Only after several requests by doctors during a medical examination she exclaimed: «…I have lost everything…I don't have anything now…I lost my spine… I don't have the mind, I don't have it anymore…there's nothing to do…no one can do anything…». It seemed as if a little window opened for an emotional interchange, but it became a desperate narration of the patient's nihilistic delusions: «…You don't believe me, no one believes me… I can't cry because I don't have tears, I never had them…I don't have lungs, I don't have an intestine, I'm empty! I can't walk…I don't have legs, I can't eat…all my teeth fell off, I can't go to the bathroom…because everything is connected… …My whole body has become empty and petrified… I've lost everything…Kill me, I'm useless, I don't want to suffer anymore. Even God cannot do anything for me…».

2.3 Treatment

The burdensome presence of such destructive anguish as well as a pervasive opposition to accept treatment and any kind of alimentation, made us decide for a parenteral therapy, in order to restore her electrolytic-metabolic balance as well as to start an antipsychotic and sedative therapy (rotation of diazepam drips and a last generation antipsychotic drug, aripiprazole, through intramuscular vials). A few hours after infusion, with her characteristic emotive detachment, she repeated coughing, trying, as she explained it as «…to expel stones from throat…». In the following hours, the patient's temperature rose to 39.6ºC, which persisted for several days, without any hematological finding of infections. Therefore, the antipsychotic therapy was temporarily suspended while still maintaining IV doses of diazepam.

A few days later, when the temperature decreased, the antipsychotic therapy with aripiprazole was restored, an antidepressant drug (sertraline 100mg/day) was introduced and the case history was modified: a gradual reduction of body-related "losing" impressions and emptiness sensations was observed. Contemporarily themes of "external alarm" took

shape, along with themes of expulsion as a solution of the crisis: “the bed is burning…the room smells like methane…I have to eliminate the plug obstructing my intestine”. A new emotional reactivity showed (although the emotions, not well controlled, related to past experiences) and sensations of sadness and guilt were expressed; particularly, it prevailed that is was no more her body but her future life that was going to pieces («…I’m destroyed, depressed, I think about my family, my home…»). While the hospitalization period went by, the previously deep depressive state was permeated by some anti-polar aspects, essentially characterized by soliloquies, vacuity, light euphoria an thought derailment, even if these echoed back to the usual themes of somatic malaise. Finally, the patient achieved a full and abrupt recovery with aripiprazole and sertraline treatments within two weeks.

The last observation was done in September 2013: the patient was taking aripiprazole and sertraline with a good compliance and complete recovery.

3. DISCUSSION

As Jules Cotard noticed, the “corp désorganisé” takes completely up the space of lived experience, nourishes the unbearable “pain” and the terrible emptiness that not even U. could perceive as a “mental” production («…I feel like a doll wandering with no body nor head or mind…») and integrate in a significance plan. Considering the observed and the referred previous episodes, intense somatic sensations, mostly restrained to “mouth”, “spine” and “brain”, the first body parts to register discomfort, emerge. The clinical case started as a germination stage, with purely psychic aspects, interpretative experiences and general fears, but it soon moved towards somatic references, poisoning themes and polarization on specific body parts. The social fears set up as pollution impressions.

Continuing with the analysis of the case history, we observed that the altered sensations developed into corporeal destruction themes [25,26]. The description offered by U. about her mood when the psychopathologic breakdown started and during her first days of hospitalization, is that of an unstructured anguish, more exactly a sense of a broken, disarticulated, lost, destroyed and petrified body. In Cotard’s Syndrome something more radical and archaic happens, i.e. a visceral involvement [26]. Jules Cotard referred about an observed patient: “She asserts she doesn’t have a brain anymore, neither nerves, nor internal organs: she has nothing but skin and bones of a disorganized body” [1]. This appears to be a lacerating feeling, rare to find, even if it occasionally shows in some “catastrophic” experiences, typical of numerous psychotic onsets [27].

The difficulty in understanding the Cotardian experiences lies in the usual temptation of offering a metaphoric reading of such experiences: the patient talks “as” if the body were crumbling, alludes to a sense of bewilderment, thus revealing a loss of cohesion of the Self. In other words, the description a Cotardian patient offers about his/her experience would express a difficulty of identity [28]; this is what Eugenio Borgna claims: “The identity of the self and the identity of the body give in to the radical leveling out of the significances, and they dissolve. This can get to the laceration of the body and its partition into separate, non-belonging fragments” [29].

Taking the psychopathologic dissertation literally, we can see the recurring of an archaic phase of Self organization; and it is to this archaic phase that the description offered by the patient seems to bring back. Enumerating the various body parts like disjointed and dysfunctional and having an altered vision of own body, brings on the terribly present phase which Gaddini calls “basic mental organization”, namely the evolutionary moment preceding

the constitution and definition of the Self [30]. The Cotardian symptoms appear, for their own intensity, short-lasting, while the lived experiences result intolerable. The hospitalization, perceived as necessary and protective, the administration of antipsychotic and antidepressant treatment and the establishment of a defensive strategy, calmed down U.'s fears and changed the clinical course into delusional aspects mixed with episodes of thymic fluctuation in both depressive and manic direction. On the cognitive side there is an alternation of thought derailment, reversal of annihilation feelings (sometimes with opposite sensations), megalomaniac ideas, persecution ideas.

The presence of those symptoms confirms the construction of a defense able to compensate the body fragmentation experience, but it also maintains a *continuum* in psychopathological expressions. It's surprising, then, to see these situations active in synchronization: this creates a simultaneity case, with nosographic and management and pharmacological problems [19,31,32].

4. CONCLUSION

In summary, in the present case a sequence of stages happened one after the other, activating a precise script in the tumultuous succession of the *Stimmung* [22]. Three connected levels emerged: the dissociative side, expressed by a loss of body-mind cohesion; the 'mixed' mood disorder, with depressive-manic episodes, and a persecutory background. All of these levels coexisted with the anguish of the idea of a body falling apart, the anguish of a descent towards the abyss of melancholia and/or an ascent to unlimited euphoria, characteristic of an "uncommon alarm" for loss of Self cohesion [33].

Concerning U.'s sudden disappearance of a Cotardian clinical picture, it has been reported that complete recovery may occur as spontaneously and suddenly as an onset of Cotard's Syndrome, even in the most severe cases [34,35].

CONSENT

All authors declare that written informed consent was obtained from the patient for publication of this case report.

ETHICAL APPROVAL

Not applicable.

ACKNOWLEDGMENTS

The authors thank Mrs. Roberta Polimanti for checking and editing the english language.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Cotard J. Hypochondriacal delusions in a severe form of anxious melancholia. Ann Med Psychol. 1880;4:168–174.

2. Pearn J, Gardner-Thorpe C. Jules Cotard (1840-1889): His life and the unique syndrome which bears his name. Neurology. 2002;58(9):1400-3.
3. Esquirol JED. Mental illness used only under medical reports, hygienic and forensic. Paris: Bailliere; 1838.
4. Cotard J. Delirium negations. Arch Neurol. 1882;4:152–170.
5. Séglas J. Delirium negations. Paris: Masson; 1897.
6. Huber CG, Agorastos A. We are all zombies anyway: Aggression in Cotard's syndrome. J Neuropsychiatry Clin Neurosci. 2012;24(3):E21.
7. Séglas J. Clinical Lectures on Mental and Nervous Diseases. Paris: Asselin and Houzeau; 1895.
8. Régis E. Historical note on the delusion of negation. Gazette Med Paris 1893:2(6):61–4.
9. Toulouse E. Note on a case of delusion of negation. Ann Med Psychol. 1893;51:259–70.
10. Got JA. Contribution to the study of Cotard syndrome, its prognostic value. Bordeaux thesis medicine; 1912.
11. Capgras J, Daumezon G. Syndrome de Cotard atypique. Ann Med Psychol. 1936;94:806–12.
12. Ey H: Délire de négations. In: Ey H, editor. Psychiatric Studies, Paris 16: Désclées Brouwer et Cie; 1950.
13. Bourgeois M. Le Cotard syndrome today. Additional note on the delusion of negation. Ann Med Psychol. 1969;127:534–44.
14. Leroux A. Clinical update of global negation delirium. Ann Med Psychol. 1986;144(9):971–86.
15. Bourgeois M. Jules Cotard syndrome and a hundred years later. Ann Med Psychol. 1980;138(10):1165–1180.
16. Trémine T. Centenary of Cotard syndrome. Evol Psychiat. 1982;47:1033–44.
17. Enoch D, Ball H. Cotard's syndrome. In: Enoch D, Ball H, editors. Uncommon Pyschiatric Syndromes, (4th edition). London: Arnold Publishers; 2001.
18. De Risio S, De Rossi G, Sarchiapone M, et al. A case of Cotard syndrome: (123)I-IBZM SPECT imaging of striatal D(2) receptor binding. Psychiatry Res. 2004;130(1):109-12.
19. Debruyne H, Portzky M, Van den Eynde F, Audenaert K. Cotard's syndrome: A review. Curr Psychiatry Rep. 2009;11(3):197-202.
20. McKay R, Cipolotti L. Attributional style in a case of Cotard delusion. Conscious Cogn. 2007;16(2):349-59.
21. Machado L, Peregrino A, Azoubel S, Cerqueira H, Lima Filho LE. Cotard's syndrome and major depression with psychotic symptoms. Rev Bras Psiquiatr. 2013;35(2):212
22. Berrios GE, Luque R. Cotard's syndrome: Analysis of 100 cases. Acta Psychiatr Scand. 1995;91(3):185-8.
23. Young AW, Leafhead KM, Szulecka TK. The Capgras and Cotard delusions. Psychopathology. 1994;27(3-5):226-31.
24. Charland-Verville V, Bruno MA, Bahri MA, et al. Brain dead yet mind alive: A positron emission tomography case study of brain metabolism in Cotard's syndrome. Cortex. 2013;49(7):1997-9.
25. Stompe T, Schanda H. The Cotard syndrome in schizophrenic disorders. Neuropsychiatr. 2013;27(1):38-46.
26. Nagy A, Vörös V, Tényi T. About the Cotard's syndrome. Neuropsychopharmacol Hung. 2008;10(4):213-24.
27. Ghaffari-Nejad A, Kerdegari M, Reihani-Kermani H. Self-mutilation of the nose in a schizophrenic patient with Cotard syndrome. Arch Iran Med. 2007;10(4):540-2.

28. De Berardis D, Serroni N, Campanella D, Marasco V, Moschetta FS, Di Giannantonio M. A case of Cotard's Syndrome successfully treated with aripiprazole monotherapy. Prog Neuropsychopharmacol Biol Psychiatry. 2010;34(7):1347-8.
29. Borgna E. The metamorphosis of the body. In Galimberti U, editor. Il Corpo (the Body). Milano: Feltrinelli; 1983.
30. Gaddini E. Notes on the mind-body problem. Riv Psicoanal. 1981;27(1):3–29.
31. Soultanian C, Perisse D, Révah-Levy A, Luque R, Mazet P, Cohen D. Cotard's syndrome in adolescents and young adults: a possible onset of bipolar disorder requiring a mood stabilizer? J Child Adolesc Psychopharmacol. 2005;15(4):706-11.
32. Consoli A, Soultanian C, Tanguy ML, et al. Cotard's syndrome in adolescents and young adults is associated with an increased risk of bipolar disorder. Bipolar Disord. 2007;9(6):665-8.
33. Ramirez-Bermudez J, Aguilar-Venegas LC, Crail-Melendez D, Espinola-Nadurille M, Nente F, Mendez MF. Cotard syndrome in neurological and psychiatric patients. J Neuropsychiatry Clin Neurosci. 2010;22(4):409-16
34. Nagy A, Vörös V, Tényi T. About the Cotard's syndrome. Neuropsychopharmacol Hung. 2008;10(4):213-24.
35. Chou PH, Lin BT, Lan TH, Chan CH. Chronic Cotard's syndrome: Recovery from 2 years' bed-ridden status. Psychiatry Clin Neurosci. 2011;65(3):301.

Peer-review history:
The peer review history for this paper can be accessed here:
http://www.sciencedomain.org/review-history.php?iid=501&id=29&aid=5434