“Extant of the living dead”: a case report on Cotard’s syndrome and its treatment aspects from a tertiary care hospital in India

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Abstract
Cotard’s syndrome has always borne a historical construct, being linked to pure psychosis, affective disorders as well as organic disorders like Parkinson’s, temporal lobe epilepsy, migraine, and cerebral infarction. Emphasis has remained on the psychopathology, involving beliefs of negation, damnation, guilt, and denouncing of body organs. Treatment aspects has been focused on less frequently; moreover, sparsely from India. We present a case of bipolar affective disorder, current episode severe depression with psychotic symptoms (Cotard’s syndrome) with special focus on treatment.

Keywords: Suicide. ECT. Antipsychotics.

INTRODUCTION
History documents that the entity Cotard’s syndrome is bequeathed by the Gallo Neuro-psychiatrist Julius Cotard (1840-1889) who in his 1880 lecture at a Societé Medico-Psychologique meeting presented the case of a lady aged 43 years who denied existence of her organs, claiming she was only skin and bones, and also, denied the existence of her own soul, the god or the devil. She further held the belief that her body could not die a natural death and the only way to free herself from mortal existence was to immolate herself.[1] At the outset, Cotard ascribed the case as “délire hypochondiaque” (hypochondriac delirium), believing it as “lypemanie”, a type of psychotic depression.[2] Two years hence, Esquirol termed the same as “délire des négations” (delirium arnison). [3] In 1893, Emil Régis coined the term “délire de Cotard” (delirium of Cotard).[4] Following the demise of Cotard, to describe presentations with anxious melancholia, delusions of absence of organs, negation, damnation, and immortality although the eponym Cotard’s syndrome was originally coined by Seglas in 1887.[1] We present a case of Cotard’s syndrome with bipolar affective disorder, and highlight the treatment aspects as per historical evolution along with specific treatment aspects in our case.

CASE HISTORY
A 40-year-old widow, a coolie worker currently unemployed since past year with poor social support, hailing from low socioeconomic status with moderately well-adjusted personality with no significant psychiatric family history presented to the psychiatry outpatient department (OPD) with history of feeling persistently sad, associated crying spells, death wish, poor sleep and appetite, and inability to work over the last one year. Worsening of symptoms for six months with complaints that her uterus was rotten and leaking was reported. According to her, bones were malformed, she did not have a backbone, associated severe back pain and hence, she could not sit or walk. She reported of having shrunken and shrivelled body as that of the very old and expressed guilt over sexual activities, stating that her lower body was now destroyed as a result of such acts. Suicidal ideas and one suicide attempt by self-immolation was discovered three months back, which had resulted in minor burns. Past history revealed similar episode of crying spells, death wish, sense of worthlessness, hopelessness, and auditory hallucinations, three years back for which patient had been admitted in psychiatric care and received three electroconvulsive therapies (ECTs) and pharmacotherapy, with remission of symptoms. Further, three episodes of excessive cheerfulness, overfamiliarity, hypersexuality in the form of soliciting multiple partners. Frequent alterations with family over this was reported by the informants, having occurred 13 years, eight years, and two years back, respectively.

For her present symptoms, several religious therapies were sought and patient visited multiple gynaecologists for her uterine ailments, documents stating reassurance and no organicity, and referral to psychiatry on two occasions. On examination, patient was dishevelled, had poor hygiene, made eye contact fleetingly, was elaborative only with female psychiatrists, wailing throughout the interview, expressing repeatedly that her lower body was rotten and purulent, particularly her uterus, and these ailments had befallen her as a direct consequence of her sexual behaviour with strange
men, that it was hopeless and she was better off dead than bringing further disgrace to her family. Her speech, though relevant and coherent, was reduced in quantity and tone. Mood was depressed with delusion of guilt and damnation, nihilistic delusions, anhedonia, worthlessness, hopelessness, helplessness, and active suicidal ideations were recorded. Insight and judgement were impaired. She was admitted and underwent blood investigations that included complete blood count, blood glucose level, liver function test, renal function test, human immunodeficiency virus (HIV), HBsAg, and thyroid function test which were within normal limits. Her computed tomography (CT) scan of brain, chest X-ray, and ultrasound abdomen were unremarkable. The X-ray of lumbar spine was done to evaluate her back pain which suggested lumbar spondylosis. A final diagnosis of bipolar affective disorder, current episode severe depression with psychotic symptoms (Cotard’s syndrome) was made as per the tenth revision of the International Statistical Classification of Diseases and Related Health Problems (ICD-10).[5] Treatment was initiated and improvements were noted with

| Table 1: Timeline of treatment and improvement |
|-----------------------------------------------|
| **Timeline** | **Treatment** | **Improvement** | **Scales’ score** |
| Day 1 - day 2 | Tab risperidone 4 mg, tab chlordiazepoxide 20 mg in divided doses | Nil | BDI: 54 |
| | | | SSI: 35 |
| | | | BABS: 22 |
| Day 3 - day 7 | Tab amitriptyline 100 mg, tab risperidone 4 mg, tab chlordiazepoxide 10 mg HS | Ill-kempt, wailing during interview reduced, mood depressed, socially withdrawn, suicidal idea ++++, delusion of guilt ++++, nihilistic delusion ++++ | BDI: 54 |
| | | | SSI: 35 |
| | | | BABS: 22 |
| Day 8 | 1st ECT | Suicidal idea ++++, delusion of guilt ++++, nihilistic delusion ++++ | BDI: 48 |
| | | | SSI: 22 |
| | | | BABS: 22 |
| Day 9 - day 11 | Tab amitriptyline 150 mg, tab risperidone 4 mg | Mood depressed but reactive and no wailing, personal care and hygiene maintained | BDI: 38 |
| Day 12 | 2nd ECT | Suicidal idea ++, delusion of guilt ++, nihilistic delusion ++++ | SSI: 18 |
| | | | BABS: 22 |
| Day 13 - day 14 | Tab amitriptyline 150 mg, tab risperidone 4 mg | Mood depressed but reactive, social interaction improved | BDI: 32 |
| Day 15 | 3rd ECT | Death wish +, delusion of guilt +/-, nihilistic delusion ++ | SSI: 8 |
| | | | BABS: 13 |
| Day 16 - day 17 | Tab amitriptyline 150 mg, tab risperidone 4 mg | Mood depressed but reactive, social interaction improved | BDI: 28 |
| Day 18 | 4th ECT | No suicidal ideation, death wish +/-, no delusion of guilt, nihilistic delusions + | SSI: 0 |
| | | | BABS: 6 |
| Day 19 - day 20 | Tab amitriptyline 150 mg, tab risperidone 2 mg | Mood anxious and reactive, social interaction improved | BDI: 22 |
| Day 21 | 5th ECT | No suicidal ideation, no delusion of guilt, no nihilistic delusion | SSI: 0 |
| | | | BABS: 0 |
| Day 22 - day 24 | Tab amitriptyline 150 mg, tab risperidone 2 mg | Mood euthymic with social interaction | BDI: 11 |
| Day 25 | 6th ECT | No suicidal ideation, no delusion of guilt, no nihilistic delusion, mood euthymic with social interaction | SSI: 0 |
| | | | BABS: 0 |
| Day 26 - day 34 | Tab sodium valproate 1000 mg, tab amitriptyline 150 mg | No suicidal ideation, no delusion of guilt, no nihilistic delusion, mood euthymic with social interaction | BDI: 5 |
| | | | SSI: 0 |
| | | | BABS: 0 |

BDI=Beck’s Depression Inventory, SSI=the Scale for Suicide Ideation, BABS=Brown’s Assessment of Beliefs Scale, ECT=Electroconvulsive therapy
use of Beck's Depression Inventory (BDI),[6] the Scale for Suicide Ideation (SSI),[7] and Brown's Assessment of Beliefs Scale (BABS)[8] (Table 1).

DISCUSSION

Time and evolution have garnered fierce debate as to whether Cotard's syndrome be considered as a separate nosological entity or as a subsyndrome of the depressive, schizophrenic, or psycho-organic disorder. Lacking consensus, Cotard syndrome has been denied a diagnostic autonomy within the fourth or fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV or DSM-5)[9,10] and ICD-10,[5] and continues being described as subsyndrome in schizophrenia[11] and organic disorders,[12] but especially in patients suffering from severe mood disorder.[13] In DSM-IV-TR,[14] nihilistic delusions are categorised as mood congruent delusions within a depressive episode with psychotic features.

In 1995, based on a retrospective factor analysis of 100 cases in literature, Berrios and Luque[3] subdivided Cotard's syndrome into three types. First, classical Cotard's syndrome included a form of psychotic depression, characterised by anxiety, melancholic delusions of guilt, and auditory hallucinations. Our case appeared to fall under this type. Secondly, Cotard's syndrome type I that was associated with hypochondriac and nihilistic delusions, and absence of depressive episodes. Thirdly, Cotard's syndrome type II, with anxiety, depression, auditory hallucinations, delusion of immortality, nihilistic delusions, and suicidal behaviour as characteristic features.[15]

Sound epidemiological data regarding Cotard's syndrome is lacking, with no reports from India. A Hong Kong-based prevalence study in a selected psychogeriatric population reported Cotard's syndrome in two out of 349 patients.[16] Inclusion of only severely depressed elderly recorded a prevalence of 3.2%.[16] In a Mexican study of psychiatric patients, 0.62% (n=three) revealed Cotard's syndrome.[17] The chances of Cotard's delusion emerging appear to increase with age.[18] An analysis of 100 cases reported mean age of 56 years,[3] and more recently, a mean age of 47.7 years was found in an analysis of 138 case reports.[19] Females appear to be more at risk than males. The syndrome is found across different ethnic groups.[18] Individuals below 25 years exhibiting Cotard's was construed to be associated with bipolar disorder.[19] Our case had similar findings with regard to gender being female,

Figure 1: Treatment options for Cotard's syndrome [21]
but was against the previous findings in terms of younger age association in bipolar affective disorder with Cotard’s.

**Treatment aspects**

Several reports have been published, but no randomised studies have been performed for Cotard’s syndrome. The most commonly reported strategy is ECT. On the classification of Berrios and Luque,[3] a suggestion was made that ECT is indicated in patients with Cotard’s syndrome and psychotic depression, while antipsychotics exert better effects in classical Cotard’s.[13] Successful pharmacotherapeutic approaches have also been published, mostly with antidepressants, antipsychotics, or a combination of both.[15] Bipolar disorder should be considered in patients under the age of 25 years.[19] Adding bromocriptine to clomipramine and lithium had a beneficial effect in a patient with bipolar disorder type I.[19] Special measures may be needed due to an important risk of suicide.[20] Our case report highlights the success of a combination therapy with ECT in conjunction with antipsychotics and antidepressants in the acute phase, and sodium valproate in the maintenance phase of bipolar type I with Cotard’s syndrome (Figure 1).[21]

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