Journal of Psychiatry and Psychology Research
JPPR, 3(2): 152-155
www.scitcentral.com

ISSN: 2640-6136

Case Report: Open Access

Temporal Seizure Focus-Induced Cotard Syndrome: A Case Study and Literature Review

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Received August 14, 2019; Accepted September 27, 2019; Published March 27, 2020

INTRODUCTION

Approximately half of all patients with seizure disorders have co-morbid psychiatric symptoms including anxiety, fear, a sense of unreality, psychosis and depression [1]. Ictal psychosis is commonly associated with temporal lobe foci [2] and is the most common peri-ictal neuropsychiatric symptom occurring in approximately 7.8% of patients with epilepsy [3]. Though there are many presentations, syndromes are usually episodic and can be manifested as depersonalization and other dissociative phenomena, and themes of death are also common [4]. Cotard syndrome is a rare psychiatric disorder that is characterized by nihilistic delusions regarding one's own body and frequently, concerning the delusional belief of being dead. Although it was first described in the late 1800's, the literature is almost completely dominated by case reports due to the rarity of the condition.

First described in 1880, Cotard believed that he had identified a new type of melancholic, psychotic depression characterized by "anxious melancholia, ideas of damnation or possession, suicidal behavior, insensitivity to pain, delusions of nonexistence and of immortality". Though the initial clinical picture described by Cotard has been debated, a 1995 analysis of 100 cases of Cotard syndrome identified the most prominent symptoms as depression (89%), nihilistic delusions about one's own body (86%) and one's own existence (69%), anxiety (65%), delusions of guilt (63%), hypochondriacal delusions (58%) and delusions of immortality (55%). In terms of its clinical profile, Berrios and Luque revealed no significant differences between men and women in manifestations of the condition or underlying diagnostic categories, however Cotard syndrome seems to be more prevalent in women [5]. In addition, the syndrome occurred across all ages though with an increased likelihood of developing the syndrome with age, with the authors finding a mean age of 52 years [6]. A prevalence rate of 3.2% has been determined in severely depressed older adults though other prevalence estimates have been lower at 0.57% [7].

This analysis of 100 cases of Cotard syndrome provided the first evidence-based classification of 3 types of Cotard Syndrome, defined in groupings as psychotic depression, Cotard Type 1 and Cotard Type 2. They identified a syndrome consistent with psychotic depression with prominent melancholia but few nihilistic delusions, Cotard Type 1 patients developed a prominent delusional component over depression, and Cotard Type 2 was characterized by a combination of anxiety, depression and auditory hallucinations [6]. A staging system has been further proposed by Yamada et al. in 1999 [8], with three stages: germination, blooming stage, and chronic stage, which further divides into two forms, though this has not become generally accepted for clinical use.

We offer a case report of ictal onset Cotard Syndrome and the patient's preoccupation with being dead serving as a clue to the diagnosis of recurrent complex partial seizures, which were not readily apparent and required significant support and advocacy for repeat EEG by the psychiatric team. This represents a rare presentation of Cotard Syndrome that has been rarely described in the literature and highlights the importance of maintaining a wide differential and ensuring that medical workup is not terminated by premature diagnosis.

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Citation: Ignaszewski MJ & Kilpatrick W. (2020) Temporal Seizure Focus-Induced Cotard Syndrome: A Case Study and Literature Review. J Psychiatry Psychol Res, 3(2): 152-155.

CASE REPORT

Our patient is a 78 year old Caucasian man with no formal past psychiatric history and stable right frontal meningioma. He had been admitted with acute onset dyspnea and chest pressure, underwent restorative cardiac catheterization, however after two events of seizure-like activity and a passive suicidal comment to a treatment team member, psychiatry was consulted. It was later revealed that he had called his girlfriend prior to the cardiac catheterization to say goodbye to her, believing that he would die during the procedure. The seizure like events were felt to be psychogenic and non-epileptic in nature by the medical team as one of the episodes occurred while the cardiologist was reviewing risks and benefits of cardiac catheterization, and were described as an increase in anxiety with reduced verbal responsiveness and twitching movements of his hand, waxing movement of his arm and resisted attempts at opening his eyes. Neurology agreed that this was an anxiety reaction.

The initial psychiatric assessment revealed a well-groomed elderly gentleman with a resting tremor bilaterally in the hands and a continuous oral tremor, as well as cog wheeling. There was some thought blocking and intermittent latency to speech. He was oriented and not found to be confused, however was noted to have significant cognitive impairment on formal cognitive testing scoring only 13 out of 30 on MOCA with difficulty in visuospatial and executive domains, delayed recall, impaired abstraction, some limitation sustaining attention and trouble with language as well. Collateral contact with his longtime partner revealed that there was no previous psychiatric history and no overt evidence of cognitive decline, having been independent with all ADL's prior to admission. The statements and telephone call were understood to have occurred secondary to confusion about the risks of cardiac catheterization while getting consented, believing that the doctor told him that he was going to die rather than the fact that this was a risk of the procedure. He was not felt to be at risk of harm to self.

Over the course of hospitalization, he made statements to nursing staff that "the lord says I am dying and I can only drink water". He expressed a "strange" experience to have heard God "talk to me" and was found to be disorganized, with significant latency to speech and notable confusion on subsequent psychiatric evaluations. He refused to eat because God had told him death was imminent. He appeared to be able to sustain attention and nursing staff had not noted him to be delirious or overtly hallucinating and there were no reports of behavioral concerns noted besides his apparent refusal to eat. He would become transiently unresponsive for periods of time, which was felt to be a behavioral response as he was oriented and not confused outside of these episodes. Following this, he started making statements that "I am dead" and refused phlebotomy for a blood draw expressing a sentiment of "why bother, you won't get any blood because you need to be alive for that." He would remove his telemetry leads asking "why do I need these things if I'm dead? My heart isn't beating" and "haven't you heard? I'm dead". He made nihilistic statements that "you don't care about me" and that "you won't come back". He would become upset and agitated when this was challenged. Despite minimal oral intake, he experienced emesis and fecal incontinence. Physical exam revealed extreme lower extremity rigidity with inability to manipulate his legs.

Discussion about the etiology of his symptoms showed a broad differential, including psychotic depression, however further workup was recommended to exclude a delirious state. He continued to have poor attention to hygiene, poor eye contact and increasing latency of response and a suspicion of catatonia was raised. A benzodiazepine challenge was postponed to allow for an expanded medical workup and EEG. CT Head revealed patchy regions of diminished attenuation of white matter, mild prominence of ventricles and chronic thalamic infarcts, which might explain the cognitive deficits and Parkinsonian symptoms. MRI ruled out an acute CNS insult such as embolus from cardiac catheterization, showing generalized brain volume loss and non-specific white matter signal changes most likely representing chronic small vessel disease and multiple scattered punctate foci in the brain parenchyma, reflecting hypertensive micro hemorrhages or amyloid angiopathy and unchanged right frontal meningioma. Initial EEG was performed and was negative for seizure or epileptic foci however results were limited as our patient only tolerated the leads for several hours prior to removing them. Ultimately, the benzodiazepine challenge showed no immediate improvement in symptoms, mirtazapine was ineffective for improving appetite or regulating sleep and he was started on low dose Seroquel. There was no appreciable symptomatic improvement and he continued to have periods of stereotyped eye blinking and unresponsiveness with fluctuating cognition and attention. He was treated with antibiotics for a CAUTI.

Given the limited duration of the initial EEG, his high vulnerability given multi-morbidity including frontal meningioma, a repeat VEEG was performed which revealed seizures of right temporal onset in the setting of generalized background slowing. Seroquel was discontinued and he was started on gabapentin 300 mg three times daily to treat the seizures and with the idea that this could also target and reduce his tremor, per neurology recommendations. With treatment, he experienced reversal of nihilistic delusions and was no longer making statements about being dead. He was able to recall his belief that he had been dead and when asked how he made sense of this, responded that "I guess it was just easier believing that at the time". He was discharged from hospital in his pre-morbid mental state and followed up with neurology as an outpatient with no known symptom recurrence.

DISCUSSION

Cotard syndrome is a fascinating, rare syndrome that can be seen in psychiatric and neurological patients, and traverses this nebulous boundary; when present, it should prompt consideration of an underlying disorder [9]. Neurological diseases that have shown clinical expression of Cotard have included Parkinson's disease, migraines, neoplasms, traumatic brain injury, viral encephalidities, arteriovenous malformations and multiple sclerosis [10], with organic lesions frequently noted in the non-dominant temporoparietal region. Postulated hypotheses include dysfunction in parietal areas associated with facial and bodily recognition and processing cortical subsystems within a unique 'center of body-image perception' for familiarity detection [11]. These deficits may contribute to an absence of familiarity and corporeal denial [12]; however this has been inconsistently supported by functional imaging studies [13]. Nevertheless, an increasing number of case reports have localized Cotard syndrome to the non-dominant temporoparietal region, such as Cotard development in a woman with right subdural hematoma [14], which highlights the importance of maintaining organic cerebral etiology on the differential list, and these findings may provide clues to underlying etiology.

Complete recovery may occur and prognosis is linked to identification of the underlying cause [5]. Therefore, a wide differential is imperative to inform a comprehensive diagnostic workup with treatment based around the underlying condition [8]. The literature reveals reports of several successful psychopharmacological treatments for Cotard syndrome, including antidepressant agents, atypical antipsychotics, and conventional mood stabilizers. Combination management with an antipsychotic and antidepressant has been effective through a series of case reports, with dual and triple therapy reported, as well. Anticonvulsants have been used infrequently in the literature, though benzodiazepines have been used in several cases that have been associated with treatment of concurrent catatonia and Cotard, as well as in an agitated patient [15]. Our patient did not show any symptomatic improvement following treatment with mirtazapine, quetiapine, an Ativan challenge for questioned catatonia, or antibiotic treatment for suspected UTI. Symptom improvement came with treatment of the underlying disorder.

Electroconvulsive therapy is an important treatment that has been beneficial in Cotard syndrome, particularly with psychotic depression in patients with limited oral intake and poor participation in pharmacologic treatment. Unsurprisingly, given its documented clinical effectiveness and evidence base for the management of treatment refractory disorders and conditions requiring rapid symptom reversal as a result of life-limiting nature, ECT has been the ultimate treatment for a number of cases, used in isolation, as sequential therapy and combination treatment [16].

Rarely in the literature has Cotard Syndrome been associated with seizures, described exclusively in a case series by Drake in 1988 [17]. Drake identified three patients who developed Cotard syndrome secondary to seizures with right frontotemporal structural lesions; these symptoms are consistent with sensation of being dead experienced by individuals with complex partial seizures [4]. These patients were treated with carbamazepine and valproic acid with resolution of symptoms, represent the first management of Cotard syndrome with anticonvulsant agents and we have identified a fourth patient who developed Cotard delusion relating to an underlying seizure disorder with clinical improvement with the use of Gabapentin.

Though we identify our patient as falling within the Type 1 Cotard classification (psychotic symptoms predominant over depressive symptoms), this provides information regarding symptomatic breakdown rather than providing a treatment algorithm. Successful treatment was reliant on correct identification of the underlying etiology, which was challenging in this case. His periodic unresponsiveness and numerous symptoms lead to limited diagnostic clarity, including considerations of delirium and catatonia. Distinctive features in our case that lead to appropriate diagnosis included the episodic nature of symptoms, occurrence of stereotypic movements that are generally consistent with seizures (rhythmic twitching, eye blinking), periodic unresponsiveness and intermittent confusion. As his Cotard symptoms resolved with treatment of seizures, this was felt to be the most likely cause of his fluctuating mental status; this was further supported by EEG that did not show general slowing or delta waves, which does not rule out delirium though may increase likelihood of other etiologies. An additional factor that introduced further confusion was the presence of Parkinson-like signs; these could not be entirely explained, due to absence of premorbid symptoms, with differential including co-occurring symptoms of catatonia that has been described in the literature as occurring with Cotard. Rigidity preceded administration of Seroquel, and dosage used would not be anticipated to cause dystonic reaction, EPS or NMS.

The etiology of new onset complex partial seizures in our elderly patient could not be definitively concluded; suspicion about contributing effect from his right meningioma was considered, particularly in the context of a vulnerable brain with atrophic and other micro vascular changes on imaging. These factors, alongside noted cognitive deficits at the time of initial assessment that had not been evident per long-term partner, may have predisposed him to development of Cotard Syndrome.

CONCLUSION

Cotard syndrome is characterized by nihilistic delusions regarding one's own body and frequently, concerning the delusional belief of being dead, occurring in a variety of psychiatric and neurologic conditions. To date, there has

been paucity of data regarding Cotard Delusion occurring in a peri-ictal state, though seizures may accompany Cotard syndrome and need to be considered during the diagnostic workup. Treatment of Cotard syndrome relies on accurately diagnosing the underlying cause, with several putative mechanisms for development however no consensus has been reached regarding etiology. We present a case of a patient who developed Cotard delusion secondary to temporal lobe seizures with symptomatic remission following administration of gabapentin. This is the first use of the anticonvulsant gabapentin to reverse Cotard delusion; however other anticonvulsants have been beneficial in three identified cases of Cotard syndrome secondary to temporal lobe seizures. The characteristic delusions of Cotard syndrome may represent a form of unreality associated with temporal lobe epilepsy and provide insight into the pathogenesis of this rare condition.

DISCLOSURE OF CONFLICT OF INTEREST

There are no conflicts of interest to disclose.

ACKNOWLEDGEMENT

No funding was accepted for the manuscript. There were no statistical experts involved. We would like to acknowledge Dr. Bruce Waslick for assistance with article composition.

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