

Cotard's syndrome with schizophreniform disorder can be successfully treated with electroconvulsive therapy: case report

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We report a case of Cotard's syndrome associated with psychotic symptoms. A 27-year-old man was admitted to hospital with the diagnosis of schizophreniform disorder. His presenting symptoms, which had started 1 month before hospital admission, were somatic delusions of gastrointestinal and cardiovascular malfunction and the absence of a stomach, which resulted in a decrease in weight from 75 kg to 63 kg in 1 month. Cranial computed tomographic images showed dilatation of the lateral and third ventricles, whereas magnetic resonance imaging revealed central atrophy and lateral ventricle dilatation. Single-photon emission computed tomography demonstrated left temporal, left frontal and left parietal hypoperfusion. The patient did not respond to antipsychotic therapies, but he was successfully treated with electroconvulsive therapy. This report emphasizes that Cotard's syndrome may be accompanied by lesions of the left hemisphere and that electroconvulsive therapy could be the first-line therapy in such patients with psychotic disorder.

Nous signalons un cas de syndrome de Cotard associé à des symptômes psychotiques. Un homme de 27 ans est admis à l'hôpital avec diagnostic de troubles schizophréniformes. Ses symptômes à l'admission (délires somatiques d'un dérèglement gastro-intestinal et cardiovasculaire et d'une absence d'estomac) avaient commencé un mois avant son hospitalisation et avaient causé une perte de poids (de 75 kg à 63 kg) durant cet intervalle d'un mois. Une tomographie par émission de positrons a révélé une dilatation du ventricule latéral et du troisième ventricule, tandis qu'une imagerie par résonance magnétique indiquait une atrophie centrale et une dilatation du ventricule latéral. Une tomographie monophotonique d'émission a fait voir une hypoperfusion temporale gauche, frontale gauche et pariétale gauche. Le patient n'a pas réagi à des thérapies antipsychotiques, mais a été traité avec succès par électrochocs. Le présent rapport indique que le syndrome de Cotard peut s'accompagner de lésions de l'hémisphère gauche et que des électrochocs pourraient être la thérapie de première ligne chez de tels patients affectés d'un trouble psychotique.

Introduction

Cotard's syndrome was originally described in 1880 by the French psychiatrist Jules Cotard, who called it the *délire des négations*.¹ The characteristic symptom of the

syndrome is nihilistic delusion. Typically, patients believe they have lost organs, blood or body parts, or even that they are dead.^{2,3} This relatively rare syndrome exists in patients with depression, schizophrenia and psychotic disorder caused by a general medical

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condition, and it is often associated with dementia.

Delusions are the principal manifestation of psychosis in neurologic disorders. Cotard's syndrome and other content-specific delusions may be observed in neurologic illnesses, organic lesions of the brain and traumatic brain injury. In patients with Cotard's syndrome, brain atrophy has been reported to occur more frequently when compared with controls.⁴

We present the case of a patient with Cotard's syndrome who was treated successfully with electroconvulsive therapy (ECT), in whom neuroimaging studies had revealed reduced regional cerebral blood flow and dilatation of the lateral and third ventricles.

Case

Mr. G, a 27-year-old single man, was referred to our outpatient clinic by his gastroenterologist for evaluation and treatment of his psychiatric symptoms. He presented with a 1-month history of symptoms that included shortness of breath, heartburn and refusal to eat solid food, leading to a weight loss of 12 kg (from 75 kg to 63 kg). He had constipation, lack of appetite, abdominal distension, ideas of gastrointestinal system malfunction, shortness of breath and headache. According to his mother, he was complaining of constipation, anorexia, weight loss, stomach and heart problems, insomnia and self-induced vomiting.

The patient reported that his headaches had started 2 years previously and that he had visited a neurologist who prescribed various analgesics, which he had taken until recently. His parents had observed during the last 2 years the patient's low mood, social withdrawal and failure in his job. Because of the cardiac and gastrointestinal complaints that had started 1 month before, cardiologic and gastrointestinal examinations were performed; they revealed no organic pathology that might result in such symptoms. Although all the patient's test results were normal, he was given an appointment for endoscopic examination by his gastroenterologist because of his persistent ideas of stomach and intestine malfunction. From the history taken both from him and from his parents, the characteristic symptoms of schizophrenia were understood to have started within the preceding month.

Mr. G underwent a psychiatric outpatient examination and was diagnosed with schizophreniform disorder according to the criteria of the *Diagnostic and Statistical Manual of Mental Disorders*, fourth edition

(DSM-IV),⁵ and thereafter he was admitted to hospital. He was prescribed haloperidol, 10 mg/d. He escaped from the inpatient unit 3–4 hours after admission, while the nurse was closing the secure door. He was seized by police officers at a shopping centre some 3–4 miles away from the hospital. He declared that the reason for his escape was the lack of oxygen in the clinic. The next morning, he attempted to escape through the ventilation area of the lavatories, but he was not successful because of the secured doors of the ventilation system. The haloperidol dosage was increased to 20 mg/d, and zuclopenthixol acetate, 50 mg, was administered by intramuscular injection.

In his first week in the hospital, the patient refused solid food and preferred to drink only water and fruit juices. His nihilistic delusional ideations were that he had no stomach and a nonfunctional gastrointestinal system, that his stomach had connected with his heart, which had no beat, and that he could not excrete feces. He used these ideas to explain his rejection of solid food. With the antipsychotic therapy, he showed some improvement and started to eat some solid food.

No neurologic or psychiatric disease was present in his family history. His past medical history revealed that when he was 6 years old, he had meningitis after mumps, from which he fully recovered after 25 days of in-hospital treatment. He had done well at school. There was no history of habitual cigarette or alcohol use.

The results of routine hematologic and biochemical tests, including thyroid hormone tests, were all normal. Two cranial computed tomographic (CT) images acquired 1 month and 2 years before the admission to hospital both demonstrated dilatation of the lateral and third ventricles. Magnetic resonance imaging (MRI) showed central atrophy and bilateral atrophic dilatation at the temporal horns of the lateral ventricle. The findings of an electroencephalogram were normal. Single-photon emission computed tomography (SPECT) was performed, and the brain image (technetium 99m hexamethylpropylamine oxime SPECT) revealed left temporal, left inferior frontal and left parietal hypoperfusion.

Because of the extrapyramidal side effects of haloperidol, biperiden was added, 4 mg/d, and the dosage was increased to 6 mg/d in the second week. At the end of the sixth week, the severity of the extrapyramidal side effects led to the decision to change the antipsychotic medication; quetiapine was started

and haloperidol was tapered over 1 week. Quetiapine was increased up to 800 mg/d, then the patient started to gain weight and show improvement. However, despite improvement in social function, he continued to show somatic delusions after 2 weeks of quetiapine treatment.

Because of the severity of the presentation and the lack of response to medication, Mr. G was referred for a course of ECT. He received a total of 12 bilateral ECT treatments (Thymatron DGx, Somatics, Lake Bluff, Ill) at a rate of 3 per week. ECT dosage was selected according to the standard settings of device percent energy (percent energy 30, frequency 50 Hz, stimulus duration 1.68 s, number of pulses 168, charge delivered 151.2 mC). All treatments were performed under general anesthesia with propofol and succinylcholine chloride. At the end of the first week of ECT treatment, the patient showed improvement, and after 12 treatments the patient was back to his premorbid status. Thereafter, ECT was stopped and olanzapine, 10 mg/d, started. Posttreatment SPECT performed the following week showed total improvement of left inferior frontal and left parietal hypoperfusion and revealed minimal hypoperfusion at the left temporal lobe.

After discharge from the hospital, no somatic or psychotic symptoms were reported by both the patient and his relatives, and he was still in remission and had good occupational and social functioning at 4 months' follow-up.

Discussion

Schizophreniform disorder was diagnosed in the initial examination of the patient described here, and he was admitted to hospital because of his bizarre delusions. His escape attempts were interpreted as disorganized behaviour, and an antipsychotic medication was started. His nihilistic delusions were suggestive of Cotard's syndrome; CT, MRI and SPECT images showed brain abnormalities consistently.

Content-specific delusions are frequently associated with neurologic disorders. In the literature, content-specific delusions have been reported to have a specific theme or topic,³ but there is no delusional content that distinguishes neurologic illnesses from idiopathic psychotic processes such as schizophrenia.

The findings of our patient's neurologic examination were normal, but the underlying brain morphology and blood flow alterations were consistent with Co-

tard's syndrome. Delusions are most common in diseases affecting both hemispheres, such as degenerative disorders and vascular dementia. When delusions follow unilateral lesions, the laterality of the damage may influence the content of the delusions.⁶ Contrary to the previously reported cases of Cotard's syndrome that mostly featured right temporal or frontal abnormalities,^{4,7,8} left inferior frontal and left parietal hypoperfusion was detected in our case.⁹

The patient's rejection of solid food in the acute phase was life threatening and could have led to the psychiatric-legal issues associated with the self-starvation that may occur in patients with Cotard's syndrome.¹⁰ Our case highlights the importance of an urgent therapeutic approach in patients with Cotard's syndrome who present with somatic and nihilistic delusions associated with self-starvation.¹⁰

It is common for the onset of delusions to be delayed for a considerable period after the occurrence of an insult to the brain. In the present case, prodromal symptoms started 2 years before the patient's admission to hospital. Headaches and social isolation were the initial complaints, and CT performed 2 years before the admission showed brain atrophy.

Previous reports have indicated the advantages of ECT in Cotard's syndrome associated with psychotic symptoms.^{8,11,12} In the present case, various antipsychotic drugs failed to provide improvement, and the patient was successfully treated with ECT.

In conclusion, this case provides further evidence that blood flow changes in the left hemisphere and brain atrophy in general could be associated with Cotard's syndrome and that ECT could be the first-line therapy in Cotard's syndrome associated with psychotic symptoms.

Competing interests: None declared.

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