Cotard syndrome and intellectual disability: a case report

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Historically, Cotard's syndrome has been described as various types of delirium. The main one is nihilistic delusion or delusion of negation. It has been reported in various neuropsychiatric disorders; however, there is little literature referring to it in a context of mental retardation. The objective of this Study is to review the features of Cotard's syndrome, based on a case study. This is a 19-year-old patient meeting the criteria for mental retardation and Cotard's syndrome. Literature has been reviewed, finding poor data depicting similar cases. It would be very advisable to further evaluate the correlation between mental retardation and Cotard´s syndrome and investigate response to treatment, as there are no protocols in our Region.

Key words: *Mental disorders, Psychosis, Delirium, Syndrome, Mental retardation*

Introduction

Cotard´s syndrome is a condition described as non frequent. The main features of this syndrome is presence of Nihilist delusions or delusions of negation. The patient thinks some parts of his/her body are missing or they work in an anomalous manner. However, Cotard´s syndrome does not have this type of delusions only, as there are other symptoms, such as immortality, guilt, hypochondriacal, delusions. Apart from other type of affection conditions and hallucinations[1].

It was first described by Jules Cotard in his monography, "Du délire hypochondriaque dans une forme grave de la mélancolie anxieuse", in 1880. He described the case of a 43-year-old woman who reported not to have a brain, nerves, stomach; she said she did not have a soul; she also reported God or Devil did not exist; and she reported she did not need to eat for living, she could live for ever, unless she is burned[2,3].

Various mental disorders have been reported, such as affective disorders, catatonia, schizophrenic spectrum, etc. and neuorological illnesses, such as subdural hemorrhage, Parkinson's disease, multiple sclerosis, epilepsy, semantic dementia, insular cortex atrophy, ischemic cerebrovascular disease, etc. where Cotard´s syndrome has been reported. There is little literature about reported Cotard´s syndrome cases within mental retardation context. This is proved by means

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of epidemiological studies. For instance a study made in Mexico, 479 patients were found with some type of primary psychiatric disorders, within which 150 patients with schizophrenia were included. Only 3 of them had Cotard´s syndrome, (0,62%), all of them had a psychotic depression[8] base. In another study, after reviewing 346 schizophrenic patients, Cotard´s syndrome was found in 3 patients only (0.87%)[9]. In order to briefly review some features of Cotard´s syndrome, we shall present the case of a young mentally retarded patient whom during his evolution had this syndrome.

Clinic Case

He is a 19-year-old male patient, from a Province in Peru. He is under his aunt´s care (he was abandoned by his parents as a baby). He was born at home from an unplanned pregnancy. He had no prenatal pre-birth controls. He had no apparent psychomotor development conditions. Incomplete schooling at a non regular school (till second grade). His academic/discipline performance was low from from primary school. No apparent mental disorders in his family. No early childhood background data

Among his personal psychiatric record, when he was a child he was absent minded, he did not obey orders, he ran away from home. When he was 12 purgative behavior and restricted food ingestion, apart from stealing and frequently running away from home to play on the internet. That year he had a sprain in his right lower limb when he was playing in the river, so he needed grafting. When he was 17, he had a careless personal care; he lost 6 kilos and was diagnosed anorexia nervosa and compulsive gambling at a Hospital in a Province of Peru. He did not follow neither his treatment with Carbamazepine, Clonazepam y Fluoxetine nor his control.

4 months before he he was hospitalized. When he was 18 his aunt noticed he was highly irritated, he did not pay attention to his personal care, he was absent minded had no energy to perform his daily activities, he was insomnia, and lost appetite. 1 month later, one night he suddenly woke up and said he had a severe migraine, he even "hit his head against the wall" trying to relief his pain. sleep onset got worse and started to say: "I am dead, I need you to buy my coffin". After that his aunt noticed the patient had to frequently to the rest room and introduced his fingers in his anus. The patient said he did this, because he had a stomachache. The patient ran away from his home and he was found around the Province by the Police. He was taken to the Hospital, as he had his left foot broken and his head wounded. The patient said he "was run over by a car"; however, he had a self aggressive behavior. When he was at the Hospital he tried to choke his aunt for no reason. He insisted on trying to run away from his home. He was diagnosed with schizophrenia. Under treatment he was medicated with Risperidone 2 mg/per day, sodium valproate 500 mg/per day and Clonazepam 1 mg/per day. Despite the treatment, no amelioration was noticed. The patient started to say: "I am going to die, hold my hands, because I am going to kill myself". Because of all the aforementioned, he was referred to the National Mental Health Institute Honorio Delgado-Hideyo Noguchi (INSM HD-HN).

When the patient was first examined he was skinny and had a splint in his left foot. He had a weakened muscle strength. He had a 5-centimeter scar on his forefront and another one on at the lower third of his right foot The rest of his examination was within normal parameters.

When he was mentally examined he was found awake, time/space/personal oriented, he did not feel conscious of a mental disorder, easily distracted, damage delirium, delusion of grandeur, cenesthopathic delusions, imperative auditory hallucinations, poor abstraction ability, preserved memory, sleep onset.

Blood analysis (hemogram, liver functions test, kidney functions test, thyroid functions test, glucose, urea, creatinine) was normal. Some neuropsychological tests were performed. The examination concluded he had a a very low IQ; therefore mental retardation was considered and organic psychosis. A psychopharmacological treatment treatment was recommended with Risperidone 4 mg/per day.

While he was at the Hospital, several times he reported nihilist delirium: "I am dead, I do not feel my body, I do not feel my muscles,

I only know I am made of bones", "my eyes and my intestine are dead, that is why I insert my finger in my anus", "I have no heart, I am dead", "I am dead, because my heart does not beat since long ago", "I am made of bones, I have neither intestine nor bones", "I do not have internal organs, I do not feel my body".

He was neurologically examined. He was subject to a electroencephalogram depicting abnormal status, as it showed persistent and slow activity and a nuclear magnetic resonance reporting no contributory findings.

He also had several symptoms of psychomotor agitation. Sometimes he even tried to choke himself with his own hands, he ran and hit against the wall, he insulted and hit the clinical personnel. Because of all the aforementioned, his antipsychotic treatment was changed by Olanzapine. He evidenced no amelioration. He was medicated with Clozapine and sodium valproate, but his self-destructive/psychotic behavior did not decrease. He was subject to 20 de electroconvulsive therapy sessions (TEC); however, no amelioration was evidenced. After 18 months at the Hospital, he still had agitation symptoms y self-aggression episodes. He was medicated with lithium carbonate, reaching up to 1,200 mg/per day, Olanzapine 30 mg/per day, Levomepromazine 25 mg/per day, because of his persistent insomnia. He was also subject to behavioral therapy. After one month under this treatment, he reported: "I am alive, my heart is beating, yes, I am alive, I am all bones". After a few days he reported "my organs are working". His pyschomotor agitation episodes and his psychotic behavior decreased. He evidenced significant clinical improvement regarding his conduct; however, he started with generalized tremors. Currently the patient is medicated with lithium carbonate 900 mg/per day, Olanzapine 30 mg/per day, Propranolol 1200 mg/per day, Levomepromazine 25 mg/according to his insomnia, his tremors decreased (this condition was a medication side effect) and behavioral management. He is still at the Hospital, because of social problems.

Discussion

Cotard´s syndrome is described as a rare psychiatric condition. It is not described in any Classification System (CIE -10, DSM – 5). However, literature mentions several reports of cases related to psychotic/Affective/organic cases, etc.

Berrios & Luque, in 1995, performed a retrospective research in 100 cases, and this condition was classified, according to its symptoms, in three types: 1) psychotic depression: anxiety, guilt delirium, depression and auditory hallucinations; 2) Cotard type I: hypochondrial delusions, nihilist delusions of the body, concept and existence; 3) Cotard type II: anxiety, delusions of immortality, auditory hallucinations, nihilist delusions of the existence and suicidal behavior[10].

Our case depicts a implausible pathology that has been described in literature and is one of the few described for mental retardation, despite it has been already correlated with this condition[11]. Besides, it has been reported in various medical conditions, such as cerebral infarctions, frontotemporal atrophy, epilepsy, encephalitis, brain tumours y cranio-encephalic traumas[12].

Our Report calls attention, as it meets what Yamada et al stated as stages in Cotard´s syndrome: 1) Germination: finding hypochondria, cenesthopathy and depressive mood; 2) Outcropping: nihilist delusions and/or immortality delirium, anxiety and negativism, y 3) Chronic: with two results: a) persistent emotional symptoms, b) systematization of paranoid type) (as the patient´s case)[13].

On the other hand, various investigations described successful management of this disorder, among them pharmacological treatments: a) anti depressive monotherapy, antipsychotic or lithium y b) combined treatment with anti depressive and antipsychotic medication. However, the most effective described strategy for Cotard´s syndrome, so far, is the electroconvulsive therapy (TEC)[12,14,15].

As stated in our Report, various medications schemes and TEC sessions were used with no

further results. That is why the choice was to use Lithium with Olanzapine, as medication in order to stop self aggression behavior, and levomepromazine for persisting insomiac, despite medications, with a favorable evolution. Unfortunately, there are no Reports describing amelioration by using Lithium, but we suppose that, because its eutimizante effect, such conducts were self limited.

It is important to highlight that the patient started to have tremors, despite the last dosage of lithium was 0.53 mmol/L, so adding Propranolol made them slightly decrease. This makes us to suppose the patient is sensitive to side effects, but assessing the risk-benefit the rightest thing was to continue the dosage, along with behavioral therapy, aimed to reinforce stability.

In our work we did not have his prenatal/ early childhood record available, because of all his aforementioned family problems. Besides, due to his constant agitation, it was impossible to follow a routine treatment with behavioral therapy.

It would be very advisable to further evaluate the correlation between mental retardation and Cotard´s syndrome and investigate response to treatment, that even though it is relatively known, it does not have an associated protocol in our Region.

Ethical Responsibility:
The patient´s family described in this paper granted their consent for publishing his medical record.

Bibliography

1. Huarcaya-Victoria J, Bojórquez-De la Torre J, De la Cruz-Oré J. Estructura factorial del Sïndrome de Cotard: revisión sistemática de reportes de caso. Rev Colomb Psiquiat 2018; [En prensa].

2. Cotard J. On hypochondriacal delusions in a severe form of anxious melancholia. Hist Psychiatry. 1999;10(38):274-8.

3. Cotard J. Del delirio hipocondriaco en una forma grave de melancolía ansiosa. In: Álvarez J, Colina F, Esteban R, editors. Delirios melancólicos: Negación y Enormidad. Madrid: La Biblioteca de los Alienistas del Pisuerga; 2009. p. 3-9.

4. Perez D, Fuchs B, Epstein J, M. A Case of Cotard Syndrome in a Woman With a Right Subdural Hemorrhage. J Neuropsychiatry Clin Neurosci 26:1, Winter 2014

5. Mendez M, Ramírez-Bermúdez J. Cotard syndrome in semantic dementia. Psychosomatics. 2011 November ; 52(6): 571–574. doi:10.1016/j.psym.2011.06.004

6. Nishio Y, Mori E. Delusions of Death in a Patient with Right Hemisphere Infarction. Cogn Behav Neurol Volume 25, Number 4, December 2012

7. Mendhekar D, Gupta N. Recurrent Postictal Depression with Cotard Delusion Indian Journal of Pediatrics, Volume 72—June, 2005

8. 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.

9. Stompe T, Schanda H. Das Cotard-Syndrom bei schizophrenen Erkrankungen. Neuropsychiatr 2013;27(1):38-46.

10. Berrios G, Luque R. Cotard's síndrome: analysis of 100 cases. Acta Psychiatr Scand. 1995; 91: 185-188.

11. Sottile F, et al. Cotard's Syndrome Triggered by Fear in a Patient with Intellectual Disability: Causal or Casual Link?, Issues in Mental Health Nursing. 2018. DOI: 10.1080/01612840.2017.1417520

12. Moschopoulos, N. P., Kaprinis, S., & Nimatoudis, J. (2016). [Cotard's syndrome: Case report and a brief review of literature]. Psychiatriki, 27, 296–302. doi:10.22365/jpsych.2016.274.296

13. Yamada K, Katsuragi S, Fujii I. A case study of Cotard's syndrome: stages and diagnosis. Acta Psychiatr Scand. 1999; 100: 396-399.

14. Weiss C, Santander J, Torres R. Catatonia, neuroleptic malignant syndrome, and Cotard syndrome in a 22-year-old woman: A case report. Case Rep Psychiatry. 2013; 2013: 452646.

15. Grover S, Aneja J, Mahajan S, Varma S. Cotard's syndrome: Two case reports and a brief review of literature. 2014; 5(1): 59-62.

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