Number of words: 1497

Number of tables: None

Number of references: 28

Delusional misidentification syndromes in obsessive-compulsive disorder

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Conflict of interest: All authors, with exception of Dr. Serra-Pinheiro, declare that they have no conflict of interest. Dr. Serra-Pinheiro has served as consultant for SIRE and received academic support for medical meetings from SIRE, Eli-lilly, Novartis and Janssen.

Acknowledgements: Prof. Leonardo Fontenelle was supported by grants from Fundação de Apoio a Pesquisa do Estado do Rio de Janeiro (FAPERJ) and Conselho Nacional de Desenvolvimento Científico e Tecnológico (CNPq). Prof. Christos Pantelis was supported by a National Health and Medical Research Council (NHMRC) Senior Principal Research Fellowship and NHMRC Program Grants.

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Key words: Obsessive-Compulsive Disorder; Delusions; Capgras Syndrome; Psychopathology

Abstract

Delusional misidentification syndromes (DMS) have been rarely reported in patients with conditions other than schizophrenia-related disorders, diffuse brain disease (dementia) and focal neurological illness. In this report, we describe DMS (i.e. Capgras and Fregoli syndromes) in two patients with severe and treatment resistant obsessive-compulsive disorder (OCD), one with paranoid personality disorder (PPD) and the other with a pervasive developmental disorder (PDD) not otherwise specified. While our findings highlight an interesting phenomenon (the occurrence of DMS in OCD), it is presently unclear whether this association is rare or relatively common but underreported. Misidentification syndromes might be the ultimate result of a combination of obsessive fears and preexisting cognitive bias/deficits, such as mistrustfulness (in PPD) or poor theory of mind (in PDD).

INTRODUCTION

Patients with obsessive-compulsive disorder (OCD) have been classically described as displaying a high level of insight in relation to their own obsessions or compulsions. However, there is a growing recognition that a significant proportion of patients with OCD may be partially or totally unable to recognize their irrationality [1]. In such cases, the symptoms have been best classified as overvalued ideas or delusions, respectively [2]. However, since there is almost no information on the overlap between the content of OCD-related cognitions (e.g. aggressive, sexual, religious, and contamination themes) and psychotic disorders-related delusions (e.g. persecutory, jealous, erotomaniac, somatic and grandiose themes), the existence of a continuum between OCD and delusional or psychotic disorders remains elusive.

Delusional misidentification syndromes (DMS) are conditions in which a patient repeatedly misidentifies persons, places, objects, or events [3]. They are relatively rare psychopathologic phenomena, occurring in about 4% of patients with functional psychosis [4]. The most common form of DMS is Capgras syndrome, i.e. the “hypoidentification” (non-recognition) of familiar persons, who the patient believes have been replaced by “doubles” or imposters [3, 5]. There are, however, other less common DMS, e.g. the “hyperidentification” (recognition) of well known persons (usually a persecutor) among people in the environment (i.e. Fregoli syndrome), who might also interchange with each other (i.e. Intermetamorphosis), or transform into the patient’s self (i.e. syndrome of subjective doubles) [5]. The diagnosis of DMS is more than a mere

exercise of academic interest, given its association with violent behavior [6, 7] or even homicide [8].

Misidentification syndromes have been rarely reported in patients with primary non-psychotic conditions [9, 10]. We are only aware of the report of a 35 year-old married woman with lifelong OCD who had incapacitating doubts about whether her husband, her parents, her cat, and even the city that she lived in had been replaced by identical-appearing duplicates [11]. Of note, this patient's OCD was resistant to "antipsychotics, antidepressants, and multiple courses of electroconvulsive therapy", and recurred even after two neurosurgical procedures (i.e. cingulotomies). In the present paper, we aimed at contributing to the literature by describing two additional patients who developed DMS (Capgras and Fregoli syndromes) during the course of severe and treatment resistant OCD.

CASE REPORTS

Mrs. A, a 20 year-old married secretary with OCD and paranoid personality disorder (PPD), began exhibiting obsessions with aggressive content and repeating rituals at age 10. At that time, she was concerned about her parents' safety and had to touch objects three to seven times to "undo evil". Concurrently, Mrs. A also exhibited motor tics (including echopraxia), which disappeared during adolescence. While a current diagnosis of OCD was confirmed using the Structured Clinical Interview for DSM-IV Axis I disorder (SCID), paranoid personality disorder (PPD) was diagnosed on clinical grounds (e.g. Mrs. A described that people always

want to deceive her). At age 17, she started to fear that she had asked different men to have sex with her and had been contaminated with HIV and/or hepatitis. When leaving a room, she would pay careful attention to other people's emotional expressions and statements to ensure that no sexual contact had taken place.

Two years after the birth of her only son, Mrs. A started to obsess about his safety. Specifically, she feared having made a "deal" that involved donating him for sexual practices or having him kidnapped after minor arguments with friends, coworkers, or doorkeepers, among others. She started to suspect that someone had substituted her son with an identical clone to bamboozle her. Although she was not 100% sure most of time, occasionally she felt like "it was real". This would lead her to spend hours daily checking his body for minor scars, so that she could confirm that her baby was actually her son. Her score on the Yale-Brown Obsessive-Compulsive Scale was 38 (minimum-0; maximum-40). Major depressive disorder was present. MRI scan and organic work-up were normal. Although showing an initial response to high dose serotonin reuptake inhibitors (SRI) and cognitive-behavioral therapy, and remaining under treatment on a long-term basis, Mrs. A OCD eventually relapsed. This time, however, her OCD and Capgras syndrome proved to be treatment resistant to different high-dose SRI potentiated with several strategies (including different atypical antipsychotics).

Mr. B, a 30 year-old single men with high school education, OCD and a pervasive developmental disorder (PDD) not otherwise specified, sought treatment for fearing being raped by a gang of sexual abusers. As a kid, he used to have no friends, to walk in circles, to stutter, and to display frequent outbursts of rage

towards family members. His mother reported that Mr. B started to show symptoms consistent with OCD, i.e. turning light switches off using his elbow, at age 9. While current OCD diagnosis was confirmed using the SCID, PDD was diagnosed on clinical grounds, based on qualitative impairments in social interaction and odd prosody.

Since late adolescence, Mr. B repeatedly asked family members, including mother, father, and sister, whether they had witnessed him having any type of sexual intercourse with strangers. Concurrently, he started to fear that one abuser was disguised to appear as different family members to reassure him that he was not at any risk of sexual abuse (while, in fact, he thought he was at risk). Violence against his mother and sister was common, particularly when they refused to provide reassurance or, more recently, when Mr. B's concerns regarding the abuser's disguise became more intense, reaching a delusional level. However, after assaulting his mother and sister-in-law, he would generally show regret and full insight regarding the absurdity of his symptoms, arguing that they were illogical but uncontrollable. His Y-BOCS score was 39. Mr. B displayed mild depressive symptoms but did not fulfill criteria for a current major depressive disorder and did not have a history of mania. Despite a history of positive and short-lived response to lithium carbonate (prescribed by another clinician for aggressiveness), Mr. B's more recent clinical picture (dominated by OCD and Fregoli syndrome) proved to be treatment resistant to several high dose SRIs augmented by atypical antipsychotics.

DISCUSSION

Our findings highlight an interesting phenomenon, i.e. the occurrence of DMS among patients with OCD. Although it is presently unclear whether these conditions are rare or actually underreported in OCD, our report adds to the current discussion regarding the existence of a continuum between obsessions and delusions or, alternatively, between OCD and psychotic disorders. In fact, although the rates of DMS in OCD are unknown, the distinctiveness of our series raises a range of diagnostic, etiological, and management issues, which we discuss below.

From a diagnostic point of view, we believe that our patients exhibited obsessional variants of DMS. Both of our patients displayed fluctuating levels of insight towards their OCD symptoms, a phenomenon that has been systematically described in OCD [12]. Further, while it could be argued that the co-occurrence of DMS and OCD represents a chance association, we feel there are reasons to believe in a continuum between these conditions. For instance, DMS could be conceptualized as an understandable progression of contamination (Mrs. A) and sexual/aggressive (Mr. B) obsessions facilitated by premorbid conditions, such as PPD and PDD, respectively [13]. Also, both OCD and Capgras syndrome have been characterized by failures to generate or experience “feelings of knowing” [14] or “rightness” [15], thus suggesting some sort of phenomenological overlap between these conditions. Interestingly, there are previous reports of OCD patients displaying a fear of turning into someone or

something else or taking on unwanted characteristics [16], a phenomenon that could be considered an obsessional variant of “reverse intermetamorphosis”, a rare type of DMS [17].

From an etiological perspective, it is difficult to conjecture on the mechanisms through which some patients with OCD might develop misidentification syndromes. However, in our particular cases, DMS might result from a combination of obsessive fears and preexisting cognitive bias/deficits, such as mistrustfulness (in PPD) or poor theory of mind (in PDD). It is interesting that autism (a PDD) has been associated with increased rates of PPD in OCD patients [18]. Accordingly, while paranoid tendencies might have shaped OCD symptoms in one case [19, 20], damaged egocentric representations of familiar persons (and its replacement by OCD-related representations) might have played a role in the other [21].

While face recognition deficits are intrinsic to the concept of DMS [3], it is also remarkable that emotional facial recognition deficits have been noted in DMS (i.e. Capgras syndrome [22]), PDD [23], and OCD [24]. For instance, in one study, patients with high-functioning autism were found to be less accurate at processing a range of basic emotional expressions, particularly disgust, anger and surprise [25] whilst in another investigation, 33% of OCD patients were impaired in their ability to recognize disgust expressions [24]. Therefore, one could speculate that these conditions may exist on continuum of severity or that there is an overlapping facial recognition deficit linking them.

Our patients raise important questions in terms of therapeutic management. Although poor insight OCD has been reported to improve after conventional treatment with serotonin reuptake inhibitors (SRIs) [20, 26], our cases have shown resistance to several trials of SRIs augmented or not with antipsychotics. Accordingly, it is unclear whether DMS lead to increased treatment resistance or, in contrast, results from a chronic and refractory OCD condition. In fact, while cluster A personality disorders (including PPD) have been described as a predictor of treatment resistance in OCD [27], DMS, poor theory of mind and belligerence have led to increased “family accommodation”, a phenomenon that has been associated with poorer response to pharmacotherapy and cognitive-behavioral treatment in OCD [28].

In summary, we have described what we believe is a rare complication of patients with severe and treatment resistant OCD. Future studies should be performed to identify (i) prevalence rates of Capgras syndrome and other DMS in OCD; (ii) the OCD phenotype generally associated with DMS, and (iii) what are the neurobiological mechanisms associated with DMS along different non-psychotic psychiatric conditions.

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