Cotard’s Syndrome after breast surgery successfully treated with aripiprazole augmentation of escitalopram: a case report

DOMENICO DE BERARDIS1,2, MAURIZIO BRUCCHI2, NICOLA SERRONI1, GABRIELLA RAPINI1, DANIELA CAMPANELLA1, FEDERICA VELLANTE1,2, ALESSANDRO VALCHERA3, MICHELE FORNARO3, FELICE IASEVOLI6, MONICA MAZZA7, GIULIANA LUCIDI8, GIOVANNI MARTINOTTI2, MASSIMO DI GIANNANTONIO2

E-mail address: dodebera@aliceposta.it

1Department of Mental Health, Psychiatric Service of Diagnosis and Treatment, Hospital “G. Mazzini”, ASL 4, Teramo, Italy
2Department of Neuroscience and Imaging, Chair of Psychiatry, University “G. D’Annunzio”, Chieti, Italy
3UOSD Senology, Hospital “G. Mazzini”, ASL 4, Teramo, Italy
4Hermanas Hospitalarias, FoRpsi, Villa S. Giuseppe Hospital, Ascoli Piceno, Italy
5Department of “Scienze della Formazione”, University of Catania, Italy
6Laboratory of Molecular Psychiatry and Psychopharmacotherapeutics, Section of Psychiatry, Department of Neuroscience, University School of Medicine “Federico II”, Naples, Italy
7Department of Health Science, University of L’Aquila, Italy
8Director, School of Nursing, MeSvA Department, University of L’Aquila, Italy

SUMMARY. In 1880 the French neurologist Jules Cotard described a condition characterized by delusion of negation (nihilistic delusion) in a melancholia context. Recently, there has been a resurgence of interest in Cotard’s syndrome (CS), but the nosographical figure of CS remains unclear. It isn’t determined if it pertains to the delusional themes area or if it is related to the sense of imminent ruin in some depressive episodes. For these reasons CS has recently been supposed to be an intermediate form. Furthermore, since even less is known about secondary CS in subjects who had never suffered of psychiatric disorders, in the present case we report the development of a secondary CS in a female patient who underwent a lumpectomy for the removal of a benign fibroadenoma. The patient responded well to aripiprazole augmentation of escitalopram and totally remitted.

KEY WORDS: Cotard’ Syndrome, nihilistic delusion, melancholia, negation, depression, aripiprazole, augmentation, escitalopram.

INTRODUCTION

In 1880 the French neurologist Jules Cotard described a condition characterized by delusion of negation with corporeal themes in a melancholia context. At first he formulated it as a new type of depression characterized by: anxious melancholia, idea of damnation or rejection, insensitivity to pain, delusions of nonexistence concerning one’s own body, and delusions of immortality. Cotard categorized it as Lypemanie, a kind of psychotic depression described by Esquirol. In 1882, he introduced the term délire de négations.

After some acknowledgments by Séglas, Regis and Toulouse, several findings, although dissonant, have succeeded especially by French clinicians who, even with critical
acumen, preferred to keep the traditional image. Most recent studies about this “uncommon syndrome”, have instead considerably drifted away from them, giving new psychopathological interpretations.

However the nosographical figure of Cotard’s Syndrome (CS) remains unclear. It isn’t determined if it pertains to the delusional themes area or it is related to the sense of imminent ruin in some depressive episodes: patients that belong to both these psychotic areas may express experiences of somatic loss associated with psychomotor arrest, a kind of deep melancholic state. For these reasons CS has recently been supposed to be an intermediate form. Furthermore, as even less is known on secondary CS in subjects who had never suffered of psychiatric disorders, in the present case we report a development of a secondary CS in a female patient who underwent a lumpectomy for the removal of a benign fibroadenoma, successfully treated with aripiprazole augmentation of escitalopram.

CASE REPORT

A 38-year-old female white-collar married with one daughter came to our observation at the outpatient facility of Psychiatric Service of Diagnosis and Treatment of Teramo (Italy) in January 2013, referred by her primary care physician.

About seven months before our visit, the patient underwent a lumpectomy for the removal of a painful breast mass diagnosed as a benign fibroadenoma after breast biopsy. The surgical intervention was executed without pre- and post-operative problems and without leaving relics. The histological examination revealed no signs of a cancer. However, her husband noted that the patient immediately before the surgical intervention became more ruminative and less active, but she told him she was worried about the possibility to have a malignant tumor.

After intervention, depressive symptoms gradually manifested and worsened within one month. She refused to go to work complaining of feeling generally unwell, “stressed”, anxious, less concentrated and expressed the belief to have a malignant cancer (decreased appetite and impaired functioning).

The first evidence-based classification of CS was made by Berrios and Luque in 1995. After a retrospective factorial analysis of 100 cases, they described three types: 1) Psychotic depression: included patients where overhang the picture of melancholia in comparison of nihilistic delusions; 2) Cotard type I: included patients where overhang the picture of melancholia in comparison of nihilistic delusions; 2) Cotard type II (mixed group): anxiety, depression, auditory hallucinations, delusions of immortality, nihilistic delusion, and suicidal behavior are the prominent features. Our patient showed characteristics compatible with a Type I CS, explaining why the clinical picture radically improved with aripiprazole augmentation.

Moreover, CS can appear after a prodromic period (germination stage) characterized by a vague feeling of anxiety, feeling of derealization and depersonalization, hypochondria and delusion of guilt. After this stage, the syndrome develops around three classic themes: denial of body part, delusions of immortality, délire d’énormité together with melancholia and ideas of damnation and possession that may increase self-agressive behaviors.
However, the problem of the present case was the development after a breast surgery in a subject without previous psychiatric problems. Several cases of secondary CS have been published and almost all evidences suggest the possibility of perceptual alterations due to central nervous system (CNS) lesions in such cases. In their comprehensive article, Debruyne et al. reviewed the co-occurrence of CS with other rare psychiatric syndromes and with several organic conditions, but, in our case, all possible causes were ruled out and a diagnosis of a pure single episode Type I CS secondary to stressful life event (breast surgery) was made.

To our knowledge, there is only one published report of CS that developed after abdominal surgery. Therefore it is possible to hypothesize that surgical interventions may be a possible independent risk factor for development of CS even in healthy individuals. It should be noted, in the present case, that a breast surgery, even if not too destructive, may be particularly distressing for a young woman more than the abdominal surgery, as involves body image and self-esteem. In fact, it has been demonstrated that younger women, particularly those with poor body image, are at an increased risk for pre- and post-surgical emotional distress. Therefore, these women may benefit from pre-surgical assessment and interventions designed to improve body image or to address emotional distress and negative attributional styles that may both contribute to the development of severe depressive symptoms. In fact, the patient with CS, whose attributional style may be introjective, might interpret emotional distress and strange sensations of depersonalization or derealization in terms of a change in herself but not in the external world.

There are several reports of successful pharmacologic treatment of CS and combination strategies (antidepressants plus antipsychotics) are often used. The aripiprazole monotherapy has been used with good results in a case of CS and was effective as augmentor in the present case, as it has been demonstrated a hyperactivity of dopamine systems in CS. The effect of aripiprazole on the dopamine systems may be attributed to its targeting of presynaptic autoreceptors and post-synaptic D2 receptors explaining why aripiprazole was effective in this case. On the other hand, as also depressive symptoms improved, it is possible that an indirect facilitation of dopamine transmission through 5-HT receptor-mediated pathways may be involved in the therapeutic response, potentiating the effect of escitalopram.

In conclusion, CS may develop after breast surgery even in women who never suffered of psychiatric disorder. Therefore a pre- and post-surgical assessment of psychiatric status may be useful especially in young women who undergo breast surgery, even if not destructive. The aripiprazole addition to antidepressant treatment may be a therapeutic option in SSRI-refractory CS, but this was only a case-report and further studies are necessary.

REFERENCES

De Berardis D et al.


