Diagnostic challenges in shared delusional disorder (folie à deux). Case presentation

**Ligia Vereșanu, Vasile Chiriță, Irina Dobrin,
Cristinel Ștefănescu, Roxana Chiriță**

Ligia Vereșanu - M.D., residency training in psychiatry, Socola Institute of Psychiatry, Iași, Romania

Vasile Chiriță - M.D., PhD, Professor, Honorary Member of the Academy of Medical Sciences Iași, Romania

Irina Dobrin - M.D., PhD, lecturer, University of Medicine and Pharmacy "Grigore T. Popa" Iași, Senior psychiatrist Socola Institute of Psychiatry, Iași, Romania

Cristinel Ștefănescu - M.D., PhD, Professor of Psychiatry University of Medicine and Pharmacy "Grigore T. Popa" Iași, Senior psychiatrist Socola Institute of Psychiatry, Iași, Romania

Roxana Chiriță - M.D., PhD, Professor of Psychiatry University of Medicine and Pharmacy "Grigore T. Popa" Iași, Senior psychiatrist Socola Institute of Psychiatry, Iași, Romania

ABSTRACT

Psycho-social factors profoundly influence the meaning and nature of symptoms in mental disorders, therefore the cultural and family framework can correctly interpret behaviors and reduce diagnostic errors and diagnostic ambiguity in clinical situations. Abnormal beliefs are the main focus of delusional disorders, often finding paranoid symptoms in dysfunctional families. This paper presents the case of two brothers with a dysfunctional family, in which psychological stress and social isolation are the main important factors in the aetiology of shared delusional disorder, a rare psychiatric condition, characterized by uncritically accepting delusional beliefs of another person. This pathology has a chronic evolution because the people involved have an unusually close relationship and are isolated from the surrounding world from a cultural point of view.

KEY WORDS:

Shared delusional disorder, diagnostic, psychosocial factors.

INTRODUCTION

Regarding the social environment, recent studies show that psychiatric disorders occur

more frequently among disadvantaged people. Evolutionary theory highlights the role of social hierarchies that influence the social

choices and expectations of others regarding a person's behavior. The type of psychosocial information has a direct impact on physiological and psychological states: repeated negative information and behaviors can lead to deviant behaviors and vice versa (1, 4).

According to C.C. Hughes, family environment becomes a configuration of images and other symbolic elements (such as language) widely shared by its members, in which it serves as a matrix of communication and tends to coordinate and sanction behaviors. Thus, the family becomes a normative framework that defines normality and promotes preferred values and behaviors, while outlawing others, leading to healthy adaptation or preventing development by causing maladaptation (2, 4).

Induced (shared) delusional disorder or folie à deux was first described in 1877 by Lasegue and Falret as a rare clinical condition in which there are usually two protagonists. There are cases, in which delusion was shared by three or even more persons, sometimes the whole family, these persons being relatives or people who have a long-term intimate contact (two brothers, mother-child, father-child, husband-wife) (8).

A specific characteristic of the shared delusional disorder is the relationship of dominance and obedience, so the dominant partner is more affected, in terms of the severity of the disease and usually suffers from schizophrenia or another psychotic disorder, thus representing the origin of the delusion that passes to the other partner (inductive delusion). The recipient of delusions is not known as a psychotic patient in the records of the psychiatric network, the manifestations appearing after the onset of delusion in the dominant partner (induced

delusion). In the most common form, the individual who has the first delusion (the primary case) is often chronically ill and is typically the influential member of the intimate relationship with a more suggestible person (the secondary case) who also develops delusions. The secondary case is often less intelligent, more gullible, more passive, with low self-esteem. The most common delusion themes are persecutory, followed by hypochondria, grandeur and sexuality, all contributing to the hostile perception of the outside world which closes a vicious circle, the two partners isolating themselves, becoming even more interdependent and more motivated in developing delusions (3). The condition is usually chronic and both the dominant and submissive individual share the original delusions, which could be of any type (5).

CASE REPORT

We present the case of two brothers that meet the criteria for diagnosing Shared Delusional Disorder (secondary case), who underwent medical treatment and had an unfavourable evolution due to psychological stress inside their family.

• *Family health history*

The social family history of the two brothers revealed that their neuropsychic and psychiatric development in early childhood was normal. They have a dysfunctional family (their father has chronic alcoholism and their mother developed schizoid personality traits). They live in four bedroom house, located in the rural area, unmarried and childless. The older brother (the dominant case) graduated from the Faculty of Agronomy Iași in 2015, but never had a job because he retired on medical grounds, just like his brother did (the secondary case), who never went to high-school. The two brothers are occasional drinkers, non-smokers.

It is worth noting that their mother manifests an overprotective behaviour, the interdictions and associated fears incapacitating them to perform age-appropriate activities and isolating them from the outside world (she goes with them at the park or shopping). Their alcoholic father did not get involved in their education and therefore created an additional negative impact on the family.

- *Medical history – case 1 (the dominant case)*

It is a male patient, aged 33, with no pre-existing conditions, was diagnosed in 2014 with Acute Psychotic Disorder (treated with antipsychotic medication Risperidone 6mg/day oral suspension and Valproic acid 1000mg/day). He was admitted to the „Socola” Institute of Psychiatry in 2015, accompanied by his mother, for psychoproduktive symptoms. In 2016, he returned to the hospital, accompanied by his mother and brother for worsening symptoms and he was diagnosed with Paranoid Schizophrenia. He continued his initial treatment with Risperidone and Valproic acid and was also prescribed Clonazepam 2mg/day. Two years later, he was re-admitted to the hospital, along with his brother, due to non-adherence to treatment and his medication was switched to Olanzapine 20mg/day, Bromazepam 6mg/day and Valproic acid 1000mg/day. Once again, in 2020, his symptoms aggravated due to alcohol consumption and along his other medication, he is also prescribed Zopiclone 7,5mg/day. His latest admission to the hospital was earlier this year, accompanied by his mother and brother, for aggravated symptoms in a conflictual context and he presented psychomotor agitation, irritability, auditory hallucinations, suspiciousness, bizarre behavior, delusion of harm and persecution and interrupted sleep.

The psychiatric evaluation revealed a partially cooperating patient, dressed appropriately, with a suspicious attitude, good hygiene, low-pitched voice, detached look, less body language and facial expressions and unmotivated laughter. He had a tendency to depression, irritability, anhedonia and spontaneous and voluntary hyperprosexia. Regarding perception, auditory hallucinations were observed („I hear two people that judge me and criticize me because I am teaching my brother how to be strong”). His thinking is relatively consistent with the flow of ideas at an accelerated pace, with disturbances of content in the form of delusions of prejudice and persecution („Everyone wants to hurt me because I have the courage to educate my brother alone”). The consciousness of the disorder is absent, he is temporal and spatial oriented.

- *Medical history – case 2 (the obedient case)*

The other male patient, aged 30, with no pre-existing conditions, was diagnosed in 2015 with Acute Psychotic Disorder, after multiple admissions to the „Socola” Institute of Psychiatry (just two months after his brother's onset of symptoms), for psychomotor agitation, irritability, auditory hallucinations („I hear two voices that criticize my behaviour and clothing when I go to church with my brother” , „Last night, there were patrol officers with dogs outside my window, ready to take me to the police station because I don't know how to behave in society and because I protect my brother”), delusion of harm and persecution and interrupted sleep. He was prescribed Risperidone 6mg/day oral suspension and Valproic acid 1000mg/day. One year later, after many re-admissions to the hospital, he was brought in by his mother and brother (who was also admitted that day) for worsening psychotic symptoms due to lack of adherence to treatment. His

medication was switched to Amisulpride 600mg/day and Valproic acid 1000mg/day and Aripiprazole 30mg/day, but he stopped taking his medication on many occasions, therefore in 2020 his medication was switched again to Risperidone 6mg/day oral suspension, Valproic acid 1000mg/day, Trihexyphenidyl 4mg/day and Zopiclone 7,5mg/day. His latest admission to the hospital was earlier this year, accompanied by his mother and brother, for aggravated symptoms in a conflictual context and he presented irritability, irascibility, clastic behaviour, mixed insomnia and bizarre behaviour.

The psychological examination revealed a disharmonious configuration of paranoid personality in a delusional and hallucinating context and an integrative-relational deficit in the social and family environment.

The psychiatric evaluation revealed a cooperating patient, dressed appropriately, with a suspicious attitude, his body hygiene was in accordance with the hospital environment, low-pitched voice, detached look, less body language and facial expressions. Regarding perception, auditory hallucinations were observed („I hear a voice that threatens me and judges me for my actions”). His thinking is incoherent and at an accelerated pace, with disturbances of content in the form of delusions of prejudice and persecution („Some bad people want to steal my house and assets”). He had a tendency to depression and interrupted sleep. Clinical examinations, neurological evaluations and laboratory testing (blood tests, EEG, ECG, brain scan, toxicology screen, endocrine panel) did not reveal any pathological modifications for the two brothers.

The positive diagnostic criteria for SPD according to DSM-IV and induced delusional

disorder (IDD) according to ICD-10 are shown below. Guidelines for diagnosing SPD and IDD do not differ significantly.

- *ICD-10 Induced delusional disorder(6)*
 - A. A subject must develop a delusion or delusional system originally held by someone else with a disorder classified in F20–F23
 - B. The two people must have an unusually close relationship with one another, and be relatively isolated from other people;
 - C. The subject must not have held the belief in question prior to contact with the other person, and must not have suffered from any other disorder classified in F20–F23 in the past.
- *DSM-IV-TR Shared psychotic disorder(7)*
 - A. Delusion develops in an individual in the context of a close relationship with another person(s), who has an already-established delusion;
 - B. The delusion is similar in content to that of the person who already has the established delusion;
 - C. The disturbance is not better accounted for by another psychotic disorder (e.g., schizophrenia) or a mood disorder with psychotic features and is not due to the direct physiological effects of a substance (e.g., a drug of abuse, a medication) or a general medical condition.

DIFFERENTIAL DIAGNOSIS (THE SECONDARY CASE)

The differential diagnosis could be ruled out based on the history of the association between both partners. The onset of the condition usually precedes the onset of the shared delusions. The diagnosis of shared psychotic disorder should always only be made after ruling out any organic causes or substance induced.

• **Schizophrenia/Schizoaffective:** This could be differentiated if the case reported other findings that are not being influenced by the primary, hallucinations, disorganized speech, grossly disorganized or negative symptoms. In the case of schizoaffective, an affective component should be present.

• **Mood Disorder with Psychotic features:** This condition has a specific delusion which is mood congruent and not shared but expressed independently.

The prognosis of shared psychotic disorder is challenging to estimate, as it depends on multiple risk factors including the primary mental disorder and the secondary biopsychosocial predisposing factors. Taking into account the family environment and lack

of adherence to treatment, with multiple relapses, the long-term prognosis is reserved.

• Factors associated with a poor prognosis are:

- Dysfunctional family;
- Alcohol abuse;
- Brother interdependency;
- Presence of psychotic behaviours (hallucinations, delusions);
- Lack of adherence to treatment.

• Factors associated with a good prognosis is:

- Family with financial resources.

TREATMENT

• *Pharmacological treatment* – needed because of the severity of symptoms and it is individualised (tab 1).

Table 1. Pharmacological treatment for the two brothers

The primary case	The secondary case
Paliperidone 6mg/day Olanzapine 20mg/day Sodium Valproate 1000mg/day Bromazepam 3mg/day Zopiclone 7,5mg/day	Risperidone oral suspension 8ml/day Valproic acid 600mg/day Trihexyphenidylum 2mg/day Zopiclone 7,5mg/day

• Psychotherapy

The separation of the induced person from the dominant partner is essential, so the induced patient becomes accessible and rational, abandoning the delusional system.

Given that patients return to the same environment, other therapeutic measures are needed, including family therapy, based on the theory that this is a system that tries to maintain homeostasis. Therapy techniques include focusing on the family, not the patient who was identified as „ill”. These techniques include: re-framing and reformulating the problem in another context, trying to change maladaptive behaviors, understanding the

dynamics between generations, achieving a balance between individualization and cohesion, increasing direct communication. It also aims to develop activities and interests outside the relationship with the dominant partner.

The exact cause of shared delusional disorder is still unknown. However, certain risk factors associated with its development, include (5):

- *Length of a relationship:* Numerous studies highlight the role of the long relationship duration as an essential factor for developing this condition. It is crucial to understand that the attachment with the primary case

plays a key role in adopting the delusion.

- *Nature of the relationship:* The majority of cases reported were among family members. The commonest relationship was between married or common-law couples and the second most common group was between sisters.
- *Social isolation:* Most cases reported poor interaction with society. An individual who is confused and perplexed can undergo influence under frightening conditions in the absence of social comparison. This information received by the secondary individual is in harmony with what the primary individual felt. The conviction to certain ideas will eventually prevail as the only solution to maintain a mutual relationship.

- *Personality disorder:* Individuals usually show features of a personality disorder. The usual description for them is as neurotic, introvert and emotionally immature. Some case reports noticed features of premorbid personality disorders especially dependent (passive), schizoid and schizotypal type.
- *Life events:* Stressful life events that affect the relationship could influence the behavior in the individual to accept certain delusions or lessening the ability to resisting the feelings/emotions.
- *Communication difficulties:* Having difficulties in sharing ideas could be a reason for preferring isolation.

CONCLUSIONS

Psycho-social factors influence the evolution of psychotic symptoms, so exposure to harmful behaviours (alcohol consumption, hyperprotective mother) tends to increase the risk of long-term relapses. In this case, a psychological cause is possible, because it includes a socially isolated relationship, so delusion becomes a way of communicating with the partner, which compensates for the rupture with the external reality. The passive partner desperately seeks the care and acceptance of the dominating person, so for the moment, the delusion seems a compromise that allows him to continue a dependent relationship. The content of aberrant thinking, delusional ideas and themes seem to depend on the environment in which patients have developed and live and on their individual and social concerns. The delusional system entails poor social functionality and often the behavior occurs as a reaction to the beliefs of the parents, in the case presented, of the mother.

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REFERENCES

1. Arnone D, Patel A, Tan GM. The nosological significance of Folie à Deux: a review of the literature. *Ann Gen Psychiatry*. 2006;(8):5:11.
2. Fennig S, Fochtmann LJ, Bromet EJ. Delusional disorder and shared psychotic disorder. *Kaplan and Sadock's comprehensive textbook of psychiatry*. Lippincott Williams and Wilkins. 2005:1525–1533.
3. Aniculăseși C.D., Radu D.A., Boloș A., Untu I., Chiriță R. Catatonic syndrome - positive and differential diagnostic controversies. Case study. *Buletin de Psihiatrie Integrativă*. 2019;3(82):105-110.
4. Trujillo M. Cultural Psychiatry. *Comprehensive Textbook of Psychiatry*. Lippincott Williams and Wilkins. 2000.