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Case report
Prikaz slučaja
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FOLIE À DEUX – A CASE REPORT

FOLIE À DEUX – PRIKAZ SLUČAJA

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Summary

Introduction. Shared psychotic disorder, or folie à deux, is a rare entity characterized by the transmission of psychotic symptoms from one patient (the inducer) to another (the induced). Delusional disorder is a type of mental illness (International Classification of Diseases, Tenth Revision, Clinical Modification, diagnosis code F24; it was moved from the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, to Other Specified Schizophrenia Spectrum and Other Psychotic Disorders). Only one person (the inducer) suffers from a real psychotic disorder, and the other/s is/are induced, and most often recover/s after separation from the inducer. **Case Report.** Based on the medical records and available literature on this condition, we present a case of a mother and son with this disorder, where the son with a paranoid psychosis transfers it to his mother. They lived alone in the same household for years, socially isolated from the others. **Conclusion.** In general, the research has shown that there are no uniform opinions among authors regarding the incidence of the disorder in males and females, at younger and older age, as well as in relationships between partners, siblings, and between parents and children. Most agree that long-term social isolation is among the most common risk factors, as well as dominance and strong power of suggestion in one partner, and passivity and tendency to suggestion in the other.

Key words: Shared Paranoid Disorder; Risk Factors; Mental Disorders; Social Isolation; Delusions; Interpersonal Relations; Suggestion; Mother-Child Relations; Treatment Outcome

Sažetak

Uvod. Udvojeni psihotični poremećaj je retka pojava koju karakteriše transmisija psihotičnih simptoma sa jednog pacijenta (*induktor*) na drugog (*indukovani* partner). Ovaj poremćaj je tip mentalne bolesti (šifra u ICD-10 sistemu klasifikacije je F24, a u DSM-V je premešten pod: „drugi specifični shizofreni spektar i drugi psihotični poremećaji"). Jedna osoba (induktor) boluje od pravog psihotičnog poremećaja, a drug(e)a je(su) indukovan(e)a, i najčešće ozdrav(e) i kada dođe do separacije od induktora. **Prikaz slučaja.** Koristeći medicinsku dokumentaciju, kao i literaturu dostupnu za temu, prikaz slučaja opisuje sina i majku sa ovim poremećajem, pri čemu sin boluje od paranoidne psihoze, koju „prenosi" na majku. Oni dugi niz godina žive sami u domaćinstvu, socijalno izolovani od okoline. **Zaključak.** U osnovi, istraživanja su pokazala da ne postoje ujednačena mišljenja među autorima u pogledu broja poremećaja uočenih kod muškaraca i žena, u mlađem i starijem uzrastu, kao i u odnosu partnera, braće i sestara, odnosu roditelj−dete. Većina se slaže da je najznačajniji faktor dugotrajna socijalna izolacija, kao i dominacija i jaka moć sugestije kod jednog, a pasivnost i sklonost sugestiji kod drugog partnera.

Ključne reči: udvojeni psihotički poremećaj; faktori rizika; mentalni poremećaji; socijalna izolacija; zablude; međuljudski odnosi; sugestija; odnosi majke i deteta; ishod lečenja

Introduction

”Folie à Deux is a rare syndrome that has attracted much clinical attention. There is increasing doubt over the essence of the condition and the validity of the original description, such that it remains an elusive entity difficult to define” [1]. According to Shimizu M. et al., shared psychotic disorder (SPD) was first discovered and described by Jules Baillarger as “folie a communique” in 1860 [2]. In 1877, Lasegue and Falret coined the name “folie à deux” or “insanity or psychosis of two” in their paper “La folie a deux” (also called “Lasègue-Falret syndrome”) [3] **(Table 1).** In his paper from 1942, Gralnick defined SPD as the transfer of delusional ideas, and/or abnormal behavior from one person to another, or one person to several others, related or unrelated, who have been in close association with the primary affected person [4]. If the disorder is shared with more than two people, it may be called folie à trois, folie à quatre, folie à famille… [5]. The number of reported and recognized cases of SPD is relatively small [6]. According to Schoenhals and his 14-year-experience, only four cases on SPD (around 0.028% of the total number of his patients) were admitted to the hospital [7]. In addition, an extensive review of the literature showed that many cases of folie à deux were not reported and remained unrecognized, so our impression of a low incidence of the illness is false. Specifically, Dušan Kovačević’s movie “Balkan Spy” is a popular dramatization of this subject matter. The main character transmits his

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Abbreviations
SPD – shared psychotic disorder

paranoid psychosis on his inmates. It is common knowledge that under normal circumstances, psychotic symptoms are not transmissible, and do not affect neither healthy nor mentally disturbed individuals. Accordingly, transmission may occur in specific cases: if people have lived together in isolated environments for a long time, spending most of their lives dependent on each other, if the "primary" patient or an inducer is a dominant partner, and if the "secondary" partner is a passive and a susceptible person, or possibly if delusion-like ideas are closely related to a potential turn of events in their environment [8].

The most common risk factors for SPS are 1) length of a relationship plays a significant role in the development of this disorder, 2) intimacy of the relationship (usually in family members or married couples), 3) social isolation, an individual in conditions of lack of social interaction may be confused and under the strong influence of another person from the environment, 4) comorbid personality disorder (such as neurotic or introvert persons with emotional immaturity), 5) mental disorder (usually untreated) in the nuclear family (untreated person with a mental disorder may have an influence on the other members), 6) cognitive disability and impairment, that disrupts rational reasoning, 7) stressful life events can influence the judgment of the individual, 8) age (usually elderly being a dominant while the young being submissive), 9) gender (more common in females) [9–13]. A paper found that 98% of cases reported between 1993 and 2005, occurred within the nuclear family, usually between married or common-law couples (52%) or sisters (24%), 50% of them in sister-sister and mother-daughter relationships and 8% in non-consanguineous patients or friends [14]. Some clinical characteristics arise, such as frequent mother-daughter associations and diagnosis of schizophrenia in inducing subject [15]. Scharfetter reported that the most common age range of the dominant partner is 16 to 82 years (average 38.5 years) and that of the submissive partner is 4 to 79 years (average, 36.2 years) [16]. Some cases of Ekbom syndrome, also known as delusional parasitosis (in which an individual harboring the delusion of being infested with insects or parasites), are described with the combination of folie à deux [17]. The first case was described by Skott in 1978. Patients may present with skin excoriations and crusting in an attempt to eliminate the underlying pathogens [18]. Also, there are a few reports on Incubus syndrome known as demonic lover, which is a dissociative disorder like dissociative hyperventilation, trance and demonic possession disorder, and dissociative amnesia [19] and Fregoli syndrome (a rare delusional belief that one or more familiar persons, usually persecutors following the patient, repeatedly change their appearance) can be found within folie à deux psychosis [20].

According to Guirvach et al. homicides may arise from all forms of folie à deux and they are most often committed with great violence. Two types of homicide are mainly described: manslaughter (no intent to kill, but death happens due to delusions and hallucinations) and intentional homicide. Research shows that a combination of mystical and persecutory delusions in SPD is associated with a high risk of homicide [21]. Delusional disorders can be induced by dopaminergic agents like psychoactive substances (PAS). Studies show that the most common PAS that cause delusional disorders are cannabis, cocaine, and amphetamines [22]. There are a few reports on the use of cannabis and amphetamines in folie à deux, which can also be combined with delusional infestations in some cases [23]. In his paper: "Folie à deux - the psychosis of association" Gralnick suggested a classification of four types of folie à deux: 1) Folie impose, 2) Folie simultanee, 3) Folie communique – a communicated insanity, 4) Folie induite [4].

Case Report

A single 39-year-old male, currently unemployed, tradesman by profession, without children, has been

Table 1. Terminology and definition of folie à deux
Tabela 1. *Terminologija i definicija folie à deux*

Serbian	Indukovano sumanuto duševno oboljenje
French	Folie á deux
English (DSM-V)	Induced delusional disorder DSM-5 does not consider Shared Psychotic Disorder as a separate entity, but rather, the physicians should classify it as a "Delusional Disorder" or "Other Specified Schizophrenia Spectrum and Other Psychotic Disorders". *Indukovani psihotični poremećaj* *DSM-5 ne smatra udvojeni psihotični poremećaj zasebnim entitetom, već bi lekari trebalo da ga klasifikuju kao Psihotični poremećaj ili kao Specifični poremećaj iz spektra shizofrenije ili drugi psihotični poremećaj.*
Latin	Psychosis paranoides inducta

Legend/*Legenda:* DSM-5 - Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition/*Dijagnostički i statistički priručnik za mentalne poremećaje, peto izdanje*

living with his 66-year-old mother in the same household for many years. He had type 1 diabetes and hypothyroidism since his childhood and there was no evidence of genetic mental illness in the family. The mother is divorced, has no more children, has a primary education and works at the market. There is no information if she was treated by a psychiatrist. The young man was treated at the Psychiatry Clinic on multiple occasions. The first hospitalization was in 1999, when he was diagnosed with opioid addiction. He underwent abstinence treatment more than once and enrolled in short-term methadone treatment. Before the end of 2017, he visited the Outpatient Clinic of the Clinical Center, accompanied by his mother, and upon his arrival complained of several somatic symptoms which were the main reason for his visit. They both looked decent and were well dressed. During the conversation, he revealed a dissociative disorder characterized by a wide range of alterations in the flow of thinking and presented with a series of non-systematized delusions. Accordingly, besides exhibiting megalomaniacal ideas and opinions (e.g. he said that he was engaged in the world's music star production, that he was the founder of a children's foundation which has raised millions of dollars to support children, and among other things he was going to marry the princess of England and he was in the Federal Bureau of Investigation witness protection program...), he demonstrated paranoid delusional ideation (he reported that some people from the casino want to hurt him because he has a children's foundation and that his mobile phone was tapped and cameras were set up to monitor him in his house...), as well as delusional ideas of reference (when passing by someone he always heard the sound of coins and messages communicated to him through the TV and radio station...). Among sensory delusions, he experienced cenesthopathic hallucinations (such as a built-in chip in his head that controls him, mobile applications which regulate the work of his heart...). At the same time, he did not affectively communicate his ideas. Instead, he was accompanied by his mother who fully confirmed the delusional ideas of her son (she said that she was recorded and followed whenever she left the house and that her son was controlled by someone who entered his head through the computer... that the phone rings strangely at night as someone was recording them...). She had a defensive attitude, and showed mood-congruent delusions (she was irreplaceable). Initially, the disorder was recognized as an induced delusional mental illness, where the young man agrees to be admitted to hospital, but his mother refuses it, as well as being prescribed any drugs, claiming that she is not crazy and that the only reason she agreed to let her son be treated is to regulate his blood sugar levels.

An antipsychotic therapy (fluphenazine) was administered to the young man. Satisfactory response to the therapy was reflected in gradual disappearance of psychotic symptoms, and ultimately by complete dissociation from the delusions present on admission to hospital. Conversations were held with his mother almost on daily basis and she regularly visited her son. At first she was still suspicious and did not trust the doctors (she said she thought doctors were involved), but there was a gradual reduction in delusions after separation from her son. After some time, when the son's discharge was planned, his mother said that she may have overstated and exaggerated things a bit and that she supported the fantasies of her son.

In our case, a young man and his mother have lived together for many years, and most of the time they depended on each other, living relatively socially isolated and without employment. During the treatment, on the grounds of paranoid psychosis, the son underwent treatment with antipsychotic medication. After the disappearance of symptoms, psychotherapeutic treatment was recommended to both of them, which they both accepted. The young man was recommended to take prescribed antipsychotic medications regularly, checking the status of mental health regularly, as well as finding a job, if possible a part-time job, to prevent further social isolation. So far, no recurrence of the disease has been recorded. We may conclude that this is the case of a folie impose - imposed psychosis, which is also supported by the fact that after separation from her son, the mother's symptoms disappeared.

Discussion

There is limited research examining SPD. One of the reasons is that SPD may be a rare condition. Another possibility is that patients with shared psychotic disorder rarely seek psychiatric treatment on their own, because of the lack of insight. They become visible if they commit a crime or if their strange behavior is noticed by others [24]. Etiologically, folie à deux has been proposed to underlie either psychogenic, sociogenic or biogenic genesis [25]. Most studies, explaining the psychopathology of this disorder, have mentioned the role of genetic susceptibility, but the occurrence of the disorder in friends and spouses also suggests an additional environmental factor as an exogenous factor [26]. Gender differences in the prevalence of folie à deux differ in the observations of several authors. Few authors believe that this disorder is more prevalent among females due to a subordinate social status, while the others think that there is equal prevalence in both genders, as well as at a younger and older age. It is considered to be evenly distributed among married couples, siblings, as well as in parent-child relationships (long-term relationships). Also, it seems that the most important factor is social isolation of the couple; it involves the transfer of delusions from the dominant (primary) partner to the second, because they usually have identical content of the delusional system and evidence that they share [27–29]. If the second person is exposed to the primary, it may be a trigger for a transient psychotic phenomenon, and the secondary subject will develop a psychotic episode (the young man's mother confirmed that she supported the fan-

tasies of her son) [30]. Usually, the secondary patient sees the primary patient as credible and trustworthy [31]. Association is found between psychosis and adverse childhood experiences such as neglect by parents or other caregivers or traumatic personal experiences (physical abuse, suicidal attempts, alcohol abuse in the family or close environment) [32]. In our case, we do not have substantial data, with the exception of an absent father.

Folie à deux seems to include a syndrome including such disorders as schizophrenia, paranoid/delusional disorder, and reactive psychosis [2]. Folie à deux, in most cases involves delusional beliefs, there is no evidence of psychomotor disturbances, flattened affect, or formal thought disorder, and it is transmitted to the secondary patient. Similary, except for the belief in her son's delusions (she spontaneously produced only the paranoid and interpretative ones) [33]. The basic therapeutic principle in psychiatry, as well as in other areas of physical medicine, claims that it is necessary to diagnose the disorder as early as possible and start treatment, use a proper dosage of the medication, and apply the medical treatment for the required amount of time [34]. Fortunately, the contact with a psychiatrist, although accidental in our case, was established early and the outcome was fairly positive. Treatment depends on the severity of the case. Old data suggest that separation is the primary method, but new data show that it may be insufficient or may aggravate the psychosis. In our case, however, separation proved to be a very helpful intervention. Drug treatment alone or combined with other types of treatments may improve the condition. If the disorder is treatable with medications only, it indicates the severity of the clinical case. Psychotherapy, as a way of treatment, can be done as mono-therapy, or both can be used. It is very difficult to predict the prognosis of this disorder. The adherence to the management plan in both partners may provide a better outcome than being untreated [9]. In our case, the psychotic symptoms in the induced penitent, inducers mother, were significantly reduced by separation from the primary patient, whereas the primary patient was treated with antipsychotics in our clinic, which led to the remission of the disorder. The young man was recommended to take prescribed antipsychotic medication regularly, checking the status of mental health regularly, as well as finding a job, if possible a part-time job to prevent social isolation.

Conclusion

In general, the research has shown that there are no uniform opinions among authors regarding the incidence of the disorder in males and females, at younger and older age, as well as in relationships between partners, siblings, and between parents and children. Most authors agree that long-term social isolation is among the most common risk factors, as well as dominance and strong power of suggestion in one partner, and passivity and tendency to suggestion in the other. We can conclude that this case was "folie impose" psychosis, supported by the fact that mother's symptoms disappeared after separation from her son.

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