Delusional sharing: a history focus-on and case report of folie à deux

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INTRODUCTION

Folie à deux (FAD) – or Shared Psychotic Disorder (SPD) or Induced Psychotic Disorder (IPD) – is a relatively rare clinical condition marked by the transference of delusional ideas from a “primary” affected individual to one or more “secondaries” in close association. Conditions like social isolation, intense emotional links and cognitive impairment or passive personality of the secondary patient seem to be risk factors for FAD1-4.

In FAD we are used to refer to the primarily affected patient as “primary”, “inducer” or “dominant partner”; the patient “influenced” by the primary is usually called “secondary”, “recipient”, “induced”5,6. It usually involves only two people - rarely three or more - an inducer and more recipients5,7.

Shared psychotic disorder is a rare syndrome and most of the studies on the topic consist in case reports. A limited amount of information, therefore, about its prevalence and incidence is available6. Moreover, etiology, natural history and prognosis of the syndrome are still unclear5,7.

According to Silveira & Seeman9, women are more affected than males in the inducer group but, in the recipient group, males and females are equally affected9,10. These results are not confirmed by Arnone et al.1; for the authors the difference between men and women in the inducer and recipient group is not statistically significant10. These outcomes are in contrast with earlier theses that females had a higher risk of being affected11,12. Recent studies also report that the age difference between primaries and secondaries might be not determinant1. Most cases of FAD involve people of the same family5-7. Married and common-law couples represent the largest proportion of cases, followed by sisters (50% twins) and by the parent-child dyad1. The most frequent psychiatric diagnosis in primary patients are delusional disorder, schizophrenia and affective disorder13,14. In the secondary, FAD is usually the primary diagnosis, but comorbidity is highly described: the most common co-morbid diagnosis seems to be schizophrenia, followed by depression, cognitive impairment (dementia, mental retardation) and bipolar disorder9,15.

Patients usually show delusional symptoms and the most
Frequent are persecutory delusions, followed by grandiose delusions. Hallucinations are most common on inducers, whereas they seem to be less frequent and less intense in recipients. For Lasègue and Falret separations from the primary can cause disappearance of the delusional symptoms in the secondary. The question seems to be more complex and most recent studies show that separation by itself is insufficient: treatment of inducer and recipient needs also a psychopharmacological approach with antipsychotic drugs.

**HISTORICAL PERSPECTIVE OF FOLIE À DEUX**

Even though Harvey, in 1651, had described a case of pseudocyesis associated with an induced psychosis in two sisters, the term *folie à deux* was first coined by Lasègue and Falret in 1877. In the paper called “La folie à deux ou folie communicante,” the authors provided descriptions of clinical cases and detailed factors that make possible any contagion as reported in Table 1. According to the authors, only the inducer suffers from an established psychotic disorder and separation will cause the abandon of the delusions by the recipient.

From that moment on, the concept has been elaborated and a new definition and different subtypes of FAD have been introduced. On his review Gralnick, after examining 105 cases gave a definition of FAD as “a psychiatry entity characterized by the transference of delusional ideas and/or abnormal behavior from one person to one or more others who have been in close association with primarily affected patient” and described four subtypes of FAD (Table 2), using European historic concept.

More than a century after Lasègue and Falret study, standardized diagnostic criteria for FAD were proposed in DSM-III as “shared paranoid disorder” and then in DSM-IV, as “shared psychotic disorder”: FAD is defined as the development of delusion in a person (secondary) as a result of a close relationship with an individual with an already established delusion (primary). This description seems to include only folie imposée-subtype of FAD without considering different types of induction. Nowadays the latest edition of DSM, reaching its fifth edition, still has a specific diagnostic category for “shared psychotic disorder”. This diagnosis can be included in “Other Specified Schizophrenia Spectrum Disorder and Other Psychotic Disorder” as “Delusional symptoms in partner of individual with delusional disorder” stressing so just part of FAD nosography. Since the DSM from the third edition were based on statistical as well as on observables and detectable phenomena some subtypes and criteria were necessarily excluded to avoid the occurrence of false positive risk. In ICD-10 FAD is defined as “Shared Psychotic Disorder” which it is applicable to clinical conditions like folie à deux, induced paranoid disorder or induced psychotic disorder, even these diagnostic criteria emphasize the traditional theory of delusion induction from a more active inducer patient to a passive recipient individual.

This is an evidence of how the necessity of a global and reliable diagnostic system based on symptomatic assessment does not match the psychopathology of a mental disorder in its complexity that should promote decisions in psychiatry.

**CASE REPORT**

AP, female, 72 years-old, is the mother of BR, female 46 years-old, formerly single. They used to live together in the same apartment since BR’s childhood, with the whole familiar nucleus composed by mother, father and two kids, both females. AP and BR were known to have good social and work functioning in their neighborhood, a small peripheral town in the countryside in Umbria, Italy. As a matter of fact, BR reached graduation and was employed and AP reached retirement.

In the patient’s personal history, they had both normal delivery and good psychomotor development through childhood and adolescence and no psychiatric history is mentioned. Furthermore, alcohol and substance consumption is denied although they both meet DSM-5 criteria for tobacco use disorder.

AP was married to BR’s father, who died of an acute myocardial infarction, and a few years later another loss deranged the family: AP’s youngest daughter, BR’s sister, died at the age of 23 because of a hematologic cancer.

Those traumatic events shocked AP and BR and became the turning point of their lives. That is they started to get isolated from their neighbors and little by little they cut down all their family ties.
DISCUSSION

FAD represents an unconventional psychiatric diagnosis that has a long history but it is still unclear in terms of pathogenesis, assessment and treatment.

Our case reported a FAD arisen between consanguineous where the partners were mother and daughter, respectively inducer and recipient as for the criteria stated by.

Regarding this condition, the genetic heritage represents a specific diagnostic challenge for the psychiatrist who might manage a similar condition, it could be hard to detect whether the secondary patient really suffered from an induced delusional belief or might have independently developed a psychotic condition due to multifactorial vulnerability.

Recent evidences supported by some electrophysiological findings report that abnormalities in the mirror neuron activity (MNA), might underlie some symptoms involved in psychotic spectrum disorder like ego-boundary disturbances, social cognition impairments and negative symptoms.

That said, these pathways might be particularly involved in FAD where the boundaries between the persons involved are particularly loosen and the delusional beliefs are specularly internalized by the recipient subject. As reported by Cuoco et al.24, some cultural traits and religious beliefs could represent additional stressors enhancing the underlying vulnerability and represent a trigger or contributory cause for this rare event.

In our patients there was a peculiar timing course where AP was first affected by persecutory delusions, grandiose ideas, fear of being cursed or spelled by other family subjects, and only on a second time the recipient started to passively share these beliefs and slowly become fully inducted and showed the same psychotic features.

Compulsory treatment was carried out because of their bad social functioning and scarce health condition. On this perspective we were able to detect important medical comorbidities such diabetes and activate proper interventions under specialist consultation.

We reported a better improvement in BR than AP in the moment they were separated in the ward, and even though they were able to interact during the day, we stated a benefit by approaching them singularly with a day-by-day meeting session. The inducer was indeed hard to be treated showing more resistance during the meetings as well as to the treatment, on the other hand the inducted was more help-seeking and easy to manage.

According to the McGlashan et al. integration/sealing over model25, BR had a better outcome, because as an “integrator” shown out to be more prone to communication and...
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Conclusions

In conclusion, even though this report could be heuristic, we suggest that an accurate background mapping could help to set a proper treatment strategy and overtake most of the problems that a rare psychiatric condition like FAD can bring.

Conflict of interest: the authors declare no conflict of interest.

References