Casi clinici

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

## *Delirio condiviso: approfondimento storico e un case report sulla folie à deux*

PIERFRANCESCO MARIA BALDUCCI[1*], CHIARA GOBBICCHI[1], PATRIZIA MORETTI[1], ALFONSO TORTORELLA[1]

*E-mail: balducci.pierfrancesco@gmail.com

[1]Department of Medicine, Section of Psychiatry, University of Perugia, Italy

**SUMMARY.** Folie à deux (FAD) is a clinical condition that was first described by Lasègue and Falret in 19th century. They reported a rare condition where two or more people shared delusional ideas from a person to another. Nowadays a trace of this historical diagnosis and its theoretical framework, could be found on ICD-10 where FAD is translated in "Shared Psychotic Disorder". Given the lack of literature and a well-defined set of symptoms it is hard to detect the clinical limits of FAD. Furthermore, the complex of comorbidities could lead to a misdiagnosis. In this paper we report a peculiar case of FAD with an historical focus trying to give a wider point of view and tools to recognize this unconventional psychiatric diagnosis.

**KEY WORDS:** folie à deux, history, induced psychotic disorder, shared psychotic disorder.

**RIASSUNTO.** La folie à deux (FAD) è una condizione clinica osservata per la prima volta nella storia da Lasègue e Falret nel XIX secolo. Descrissero una rara circostanza nella quale due o più persone si ritrovarono a condividere idee deliranti gli uni dagli altri. Oggigiorno tracce di questa diagnosi del passato e del suo substrato psicopatologico si ritrovano nell'ICD-10, in cui la FAD viene codificata come "Disturbo Psicotico Condiviso". Considerando la scarsa letteratura in merito e la mancanza di una ben precisa definizione sintomatologica, risulta difficile circoscrivere i confini clinici della FAD. Inoltre, la presenza di comorbilità può portare a un'errata diagnosi. In questo articolo riportiamo un caso peculiare di FAD e un approfondimento storico, nel tentativo di fornire un punto di vista più ampio e degli strumenti atti a riconoscere questa diagnosi psichiatrica "non convenzionale".

**PAROLE CHIAVE:** folie à deux, storia, disturbo psicotico indotto, disturbo psicotico condiviso.

### 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[1]. Conditions like social isolation, intense emotional links and cognitive impairment or passive personality of the secondary patient seems to be risk factors for FAD[1-4].

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 recipients[1,7].

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 available[8]. Moreover, etiology, natural history and prognosis of the syndrome are still unclear[5,7].

According to Silveira & Seeman[9], women are more affected than males in the inducer group but, in the recipient group, males and females are equally affected[9,10]. 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 significant[10]. These outcomes are in contrast with earlier theses that females had a higher risk of being affected[11,12]. Recent studies also report that the age difference between primaries and secondaries might be not determinant[9]. Most cases of FAD involve people of the same family[5-7]. Married and common-law couples represent the largest proportion of cases, followed by sisters (50% twins) and by the parent-child dyad[1]. The most frequent psychiatric diagnosis in primary patients are delusional disorder, schizophrenia and affective disorder[13,14]. 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 disorder[9,15].

Patients usually show delusional symptoms and the most

frequent are persecutory delusions[2,10], followed by grandiose delusions[13]. Hallucinations are most common on inducers, whereas they seem to be less frequent and less intense in recipients[1,9]. For Lasègue and Falret[12] 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[10,11].

## 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[12]. In the paper called "La folie à deux ou folie communiqué", the authors provided descriptions of clinical cases and detailed factors that make possible any contagion[10,12] 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[7,12].

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[11], after examining 103 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[10,11].

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)[16,17]. This description seems to include only folie imposée-subtype of FAD without considering different types of induction[10]. Nowadays the latest edition of DSM, reaching its fifth edition, still has a specific diagnostic category for "shared psychotic disorder"[18]. 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 statistic as well as on observables and detectable phenomena some subtypes and criteria were necessarily excluded to avoid the occurrence of false positive risk[19]. 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[20]: even these diagnostic criteria emphasize the traditional theory of delusion induction from a more active inducer patient to a passive recipient individual[10].

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[21].

**Table 1.** Lasègue and Falret syndrome.

| |
|---|
| A syndrome prevalent among women living more or less confined, marked by: |
| • Coincidental appearance of psychotic symptoms in members of a family while living together<br>• Appearance of psychotic symptoms in two closely associated persons and retention of symptoms once initiated, in spite of separation.<br>• Transmission of psychotic symptoms from a sick person to one person or several healthy individuals who elaborate on the induced delusions. |

**Table 2.** Subtypes of FAD described by Gralnick[11].

| | |
|---|---|
| Subtype A | |
| *Folie imposée* (by Lasegue and Falret) | The delusional symptoms are transferred from a psychotic individual to a psychiatrically normal one. |
| Subtype B | |
| *Folie simultanée* (by Règis) | Identical psychoses (characterized by depression and persecutory ideas) appear simultaneously in two individuals, with no evidence of mental contagion. |
| Subtype C | |
| *Folie communiquée* (by Marandon and Montyel) | The secondary develops delusional symptoms after a long period of resistence. Symptoms remain even after separation. |
| Subtype D | |
| *Folie induite* (by Lehman) | New delusions are added to a psychotic individual's preexisting delusions under the influence of another patient. |

## 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.

Then the social and emotional withdrawal corresponded to a progressive psychological impairment resulting in a shared belief that all the people, except for AP and BR, were practicing evil spells toward them and all this was the real reason why such bad life events occurred in the family. Moreover, this belief became a structured delusion where all the negative events were correlated to actuality. So the husband's death was, for example, correlated to John Kennedy's murder or, on the other hand, some lyrics included the real truth about their bad luck.

From this moment on, they lived in seclusion, spending most of the time at home without any contact with the outside. In detail, AP was the only one of the couple to exit sporadically only to provide supplies and other first needs meanwhile BR waited at home without moving. That is, for 13 years BR hadn't been out of her house.

Mother and daughter both started to involve the authorities by writing claiming letters about the big plot they were in. Letters were sent to the local police department, to the mayor and even to the President of the Italian Republic interpreting the missing feedback as a part of the plot with a mutual enhancement of the delusional beliefs.

Their new general practitioner (GP), who they had never met and still refused to meet, collected some complaints from the whole neighborhoods because of strange construction work-like noises especially during the night hours so he decided to activate the psychiatric territorial service facility (CSM) of competence in order to plan an intervention. The director in charge, who is essentially appointed for outpatient management, went with GP thereby to set an evaluation although this attempt failed and they were rejected.

So thanks to the activation of compulsory treatment protocol, the psychiatrist and the GP, together with the police intervention, were able to get into the house and find a bad quality of life with poor hygiene conditions and just canned food and snacks as for diet.

Following the protocol ambulance and police carried both patients to our ward where they were admitted in involuntary status for 7 days as stated by the law.

During the hospitalizations psychometric tests were performed and they both resulted with a cluster A of personality, suggesting a possible evolution of the same premorbid patterns.

At their mental state examination, both patients showed an apparent age different from their real one especially for what concerns BR. She was in bad physical condition, with obesity and untreated personal care. She also showed an immature behavior for her age. They were not orientated to person and place with bad concentration due to their delusional thoughts with impairment on the reality exam showing no insight about their morbid condition.

Laboratory testing revealed uncontrolled diabetes mellitus with high percentage of glycated hemoglobin for AP who was introduced to an adequate insulin injection regimen and meal plan. Moreover, her daughter showed a glucose intolerance controlled with diet, suggesting a family trait. Both cases were referred to an endocrinologist for an accurate management over the time.

Patients were hosted in two different rooms trying to loosen their delusional link. They also received a psychopharmacological treatment to calm down anxiety subsequent to hospitalization and to overtake their delusional belief in order to obtain a better communication path providing social interventions.

That said, at the discharge AP was treated with haloperidol long acting formulation due to her metabolic status and her poor response to first line medications reporting also bad compliance.

On the other hand, BR had a better course showed important improvements with oral therapy with second generation antipsychotic and benzodiazepines.

A social network was created by the psychiatric territorial facilities which activated social workers for their social needs and planned special psychological sessions both single and coupled focused on their specific role on the folie à deux disease led by two different senior psychiatrists working in the same facilities where they were referred to. They were discharged on two different dates to provide a reinforcement of the improvements obtained.

## 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 an FAD arisen between consanguineous where the partners where 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 inducted 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[22,23].

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 model[25], BR had a better outcome, because as an "integrator" shown out to be more prone to communication and

to relate with care-givers than "sealer" patients like AP confirming the study from Poloni et al.[26].

As for the discharge program, the ward medical staff in collaboration with the colleagues of the territorial outpatient service discharged the two patients on different dates in order to reinforce the targeted interventions and according to the different course they had followed different pharmacological strategies. AP had Haloperidol LAI and BR received oral aripiprazole with good clinical response with no revolving door.

## 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


1. Arnone D, Patel A, Tan GM. The nosological significance of Folie à Deux: a review of the literature. Ann Gen Psychiatry 2006; 5: 11.
2. Talamo A, Vento A, Savoja V, et al. Folie à deux: double case-report of shared delusions with a fatal outcome. Clin Ter 2011; 162: 45-9.
3. Mazzoli M. Folie à deux and mental retardation. Can J Psychiatry 1992; 37: 278-9.
4. Ghaziuddin M. Folie à deux and mental retardation: review and case report. Can J Psychiatry 1991; 36: 48-9.
5. Yazar MS, Erbek E, Eradamlar N, Alpkan L. The Seven Sleepers: a folie à deux case originating from a religious-cultural belief. Transcult Psychiatry 2011; 48: 684-92.
6. Kashiwase H, Kato M. Folie à deux in Japan. Analysis of 97 cases in the Japanese literature. Acta Psychiatr Scand 1997; 96: 231-4.
7. Lew-Starowicz M. Shared psychotic disorder with sexual delusions Arch Sex Behav 2012; 41: 1515-20.
8. Ranjbar-Kouchaksaraei F, Norazar G-R, Mohaghghegi A. Shared psychotic disorder between a girl with her mother and younger sister (folie à trois). Arch Iran Med 2006; 9: 417-8.
9. Silveira JM, Seeman MV. Shared psychotic disorder: a critical review of the literature. Can J Psychiatry 1995; 40: 389-95.
10. Shimizu M, Kubota Y, Taichi M, Baba H. Folie à deux and shared psychotic disorder. Current Psychiatry Reports 2007; 9: 200-5.
11. Gralnick A. Folie à deux: the psychosis of association. Psychiatr Q 1942; 16: 230-63.
12. Lasègue C, Falret J. La folie à deux. Annals of Medical Psychology 1877; 18: 321-55.
13. Ferro FM, Pacilli AM. La folie à deux: il caso di una simbiosi madre-figlio. Imago 2003; 10: 57-68.
14. Wehmeier P, Barth N, Remschmidt H. Induced delusional disorder: a review of the concept and an unusual case of folie à famille. Psychopathology 2003; 36: 37-45.
15. Dodig-Curković K, Curković M, Degmečić D, Filaković P. Shared psychotic disorder ("folie à deux") between mother and 15 years old son. Coll Antropol 2008; 32: 1255-8.
16. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 3rd ed. Washington DC: American Psychiatric Association, 1980.
17. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 4th ed. Washington DC: American Psychiatric Association, 1994.
18. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Washington DC: American Psychiatric Association, 2013.
19. Biondi M, Bersani FS, Valentini M. The italian edition of DSM-5. Riv Psichiatr 2014; 49: 57-60.
20. World Health Organization. The ICD-10 classification of mental and behavioural disorders: clinical descriptions and diagnostic guidelines. Geneva: World Health Organization, 1992.
21. Guidi J, Fava GA. Emerging trends in clinical psychology. Riv Psichiatr 2014; 49: 227.
22. Mehta UM, Thirtalli J, Aneelraj D, Jadhav P, Gangadhar BN, Keshavan MS. Mirror neuron dysfunction in schizophrenia and its functional implications: a systematic review. Schizophr Res 2014; 160: 9-19.
23. Zaytseva Y, Bendova M, Garakh Z, et al. In search of neural mechanisms of mirror neuron dysfunction in schizophrenia: resting state functional connectivity approach. Psychiatr Danub 2015; 27 Suppl 1: S269-72.
24. Cuoco V, Colletti C, Anastasia A, Weisz F, Bersani G. A case of shared psychotic disorder (folie à deux) with original aspects associated with cross-cultural elements. Riv Psichiatr 2015; 50: 43-6.
25. McGlashan TH, Levy ST, Carpenter WT. Integration and sealing over: clinically distinct styles from schizophrenia. Arch Gen Psychiatry 1975; 32: 1269-72.
26. Poloni N, Diurni M, Buzzi A, et al. Recovery style, symptoms and psychosocial functioning in psychotic patients: a preliminary study. Riv Psichiatr 2013; 48: 386-92.