

Pseudocyesis Leading to Folie-à-Deux

Amit B. Nagdive¹ Ravi Singh Bhainsora¹ Rouchelle Fernandes¹ Prakash B. Behere¹ Siddharth Sethi¹

¹Department of Psychiatry, Jawaharlal Nehru Medical College, Sawangi, Wardha, Maharashtra, India

Address for correspondence Rouchelle Fernandes, MBBS, Department of Psychiatry, Jawaharlal Nehru Medical College, Wardha, Maharashtra, India (e-mail: rouchelle795@gmail.com).

J Neurosci Rural Pract 2021;12:419–423.

Abstract

Pseudocyesis or false belief of pregnancy is the emergence of classical manifestations of pregnancy—nausea, breast enlargement and pigmentation, abdominal distention, amenorrhea, and labor pains—in a nonpregnant woman. It is a multifactorial disease and its development is influenced by many different elements such as neuroendocrine, social, psychodynamic, and cultural issues. “Folie-à-deux,” is shared psychotic disorder, describes a syndrome in which delusion is transferred to another person who is more susceptible. Both individuals are closely related or know each other for a long time and typically live together in relative social isolation. In its commonest form, the individual who first develops the delusion (the primary case) is often chronically ill and typically is the dominant member in a close relationship with a more suggestible person (the secondary case) who also develops the delusion. Treatment options should also be kept in mind as antipsychotics themselves can increase prolactin levels and can lead to amenorrhoea and galactorrhea and can further strengthen patient’s belief about her pregnancy. This case highlights that the most important therapeutic step in the treatment of folie-à-deux is separation of the inducer and the induced. Here we describe a case of folie-à-deux of a married couple in which the female had delusional pregnancy while the husband shared and supported her delusion against substantial medical evidence.

Keywords

- ▶ delusion of pregnancy
- ▶ delusional disorder
- ▶ shared psychotic disorder

Introduction

Pseudocyesis or false belief of pregnancy is the emergence of classical manifestations of pregnancy—nausea, breast enlargement and pigmentation, abdominal distention, amenorrhea, and labor pains—in a nonpregnant woman. Pseudocyesis illustrates the ability of the psyche to influence the soma, most likely through central input at the level of the hypothalamus.¹

Extensive analysis on the subject revealed that most of the cases originated in developing countries. Eighty case histories were found.²

Pseudocyesis is a multifactorial disorder and its evolution is influenced by many different elements such as neuroendocrine, social, psychodynamic, and cultural issues.³ Few case reports have also mentioned pseudocyesis in relation to anti-psychotic-associated hyperprolactinemia⁴⁻⁷ and metabolic syndrome.⁸

Although in cases of pseudocyesis, single fetus is most commonly reported but often patients may report more than one.⁹

“Folie-à-deux,” was devised by Lasegue and Falret in 1877, also known as shared psychotic disorder, is a syndrome in which delusions are transferred from one individual to another.¹⁰

published online
March 24, 2021

DOI <https://doi.org/10.1055/s-0041-1726615>
ISSN 0976-3147

©2021. Association for Helping Neurosurgical Sick People. This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>). Thieme Medical and Scientific Publishers Pvt. Ltd. A-12, 2nd Floor, Sector 2, Noida-201301 UP, India

Both pseudocycosis and folie-à-deux are considered to be rare mental illnesses but are often reported in developing countries. It frequently occurs when a married couple was infertile and living in a society that gave much more emphasis on having children. The infertility is often blamed on the woman, due to which she is exposed to social discrimination and substantial distress.

We describe a case of folie-à-deux of a married couple in which the female had delusional pregnancy, while the husband shared and supported her delusion against substantial medical evidence.

Case Report

The case presented to a tertiary care center in Wardha district of Maharashtra state, India, on February 12, 2019. Mrs. A, 37-year-old married woman, housewife, was referred from gynecology department with her husband and sister-in-law. She had visited the hospital five to six times in the past 2 months claiming that she was 6 months pregnant with a single child. Her urine pregnancy test (UPT) and beta-human chorionic gonadotropin were negative and after multiple ultrasonography (USG) abdomen done both in our hospital as well as multiple other centers were not indicative of pregnancy, she was still not willing to believe that she was not pregnant. She believed she could feel the baby kicking in her womb; her belief was further strengthened by her husband who said he could feel the baby kicking whenever he touched her abdomen. Husband also complained that USG was not done properly as radiologist had not scanned the whole abdomen. Sister-in-law after seeing the reports had questioned the couple regarding pregnancy but kept quiet when the couple became adamant and aggressive. They had consulted four times in the gynecology department. After initial consultation and detailed workup when no evidence of pregnancy was found, the patient was subsequently referred to psychiatry department that she refused as she believed she was not mentally ill. But after persistent counselling and on the advice of her sister-in-law she reluctantly agreed.

Mrs. A was the youngest of her three siblings, only female child out of the three and described herself as a shy kid who preferred to stay at home. Her father passed away when she was 12 but refused that it impacted her as she always felt that her father was more close to her brothers. She claims that she was close to her mother and was average in studies.

Mrs. A was married 10 years ago after she ran away from home with her husband. As they belonged to different caste, their parents were not willing to get them married. Mrs. A had known her husband and had an affair for 12 months while they were studying in same BSc. course prior to eloping. Eventually her-in-laws accepted her but her parents never spoke to her again. She got pregnant a year later but had spontaneous abortion in 3 months. In the past 9 years, the couple had tried five times but

her pregnancy would not last for more than 3 months. Her-in-laws were supportive of her but gradually they started blaming her as she could not have a child. Her husband always supported her, and when she could not tolerate further verbal abuses from in-laws, much to the protest of her husband, they moved away and started living separately and it had been 2 years since they have been living separately. Her in-laws complained in the past 2 years, the couple had become increasingly isolated and would seldom meet other family members. They also alleged that the husband had always been inclined toward the patient from the beginning and would not listen to anyone else on any other matters. He even left home at her advice and started living separately.

She described her marital relationship as adequate but had noticed that in the past 1 year their interaction had declined. There was no history of any substance use by the husband. Over the past 6 months, the patient had developed increase in appetite and consequently weight gain of ~8 kg in the initial 3 months. She often felt nauseated and developed bloating sensation. She could also feel the baby kicking and moving in her womb. There was also an enlargement in breast and visible pigmentation. The husband also felt the same way about the patient and took her to various gynecologists. Whenever a doctor would disprove pregnancy, they would just change the doctor, and eventually, they reported to our hospital.

On further interview the patient claimed that she could intermittently hear voice of her baby who would call her maa. Husband also claimed that whenever he would rest his head against her abdomen, he could hear faint sounds/noises that he believed were coming from the baby and it further strengthened his belief about the pregnancy.

The patient also had a sad mood, mainly due to the constant disapproval of her pregnancy. However, the patient denied any other symptoms of a major depressive episode including loss of interest in previously pleasurable activities, fatigability, ideas of hopelessness and worthlessness, frequent crying spells, death wishes, and active suicidal ideation.

During interview, the patient did admit that she had irregular menstrual cycles with scanty bleeding that she owed to her doing regular household work during her pregnant state. She blamed her UPTs were faulty and her husband supported this by showing different blogs and reports that he had found on internet claiming that UPT and other routine blood tests did not always detect pregnancy. He also blamed radiologist for not conducting USG properly. The couple alleged that the world had been against them and their relationship from the start and this was a conspiracy and hospital was a part of it. They refused to cooperate and left hospital, but sister-in-law assured us that she would convince them and bring them back for consultation.

They presented a week later (19-02-2019) with in-laws (brother-in-law, sister-in-law, mother-in-law, nephew) and were admitted into psychiatric ward.

Medical Examination and Laboratory Findings

The laboratory findings of Mrs. A is given in ► **Table 1**.

Table 1 Mrs. A—Ultrasonography abdomen revealed uterus of 60 x 35 mm in size, empty cavum, clear endometrium 8.4 mm was seen. The adnexa was echo free.

Test	Specimen	Value
Acid phosphatase	Serum	0–0.07 μ kat/L
Activated partial thromboplastin time	Plasma	31 s
Adrenocorticotrophic hormone	Serum	8 pmol/L
Albumin	Serum	39 g/L
Alkaline phosphatase	Serum	0.9 μ kat/L
Alpha fetoprotein	Serum	13 pg/L
δ -Aminolevulinic acid	Serum	1.25 μ mol/L
Aminotransferase, alanine	Serum	24 U/L
Aminotransferase, aspartate	Serum	28 U/L
Ammonia	Plasma	32 μ mol/L
Amylase	Serum	86 U/L
Antidiuretic hormone	Plasma	0.07 pmol/L
Bilirubin		
Direct	Serum	0.2 mg/dL
Total	Serum	0.8 mg/dL
Cholesterol		
High-density lipoprotein (HDL-C)	Plasma	43 mg/dL
Low-density lipoprotein (LDL-C)	Plasma	95 mg/dL
Total (TC)	Plasma	172 mg/dL
Cortisol		
At 5 PM	Serum	7 μ g/dL
At 8 AM	Serum	11 μ g/dL
After overnight suppression test	Serum	3 μ g/dL
C-peptide	Serum	2.1 ng/mL
C-reactive protein	Serum	0.1 mg/dL
C-reactive protein, highly sensitive	Serum	0.07 mg/L
Creatine kinase	Serum	58 U/L
Creatinine	Serum	0.9 mg/dL
Erythrocyte sedimentation rate	Blood	11 mm/h
Erythropoietin	Serum	10.5 IU/L
Estradiol	Serum	102 pg/mL
Ferritin	Serum	98 μ g/L
α -Fetoprotein	Serum	12 pg/L
Folate (folic acid)		
RBC	Blood	330 ng/mL
Serum	Serum	9.2 ng/mL
Follicle-stimulating hormone	Serum	11.4 mIU/mL

(Continued)

Table 1 (Continued)

Test	Specimen	Value
Glucose		
2-h postprandial	Plasma	120 mg/dL
Fasting	Plasma	98 mg/dL
Hemoglobin	Blood	13.5 g/dL
Hemoglobin A _{1c}	Blood	5.2%
Human chorionic gonadotropin, quantitative	Serum	3 mIU/mL
Luteinizing hormone	Serum	14 IU/L
Parathyroid hormone	Serum	38 ng/L
Parathyroid hormone-related peptide	Plasma	1.2 pmol/L
pH	Blood	7.40
Platelet count	Blood	180 \times 10 ³ / μ L
Potassium	Serum	4.1 mEq/L
Progesterone	Serum	8 ng/mL
Prolactin	Serum	11 μ g/L
Testosterone	Serum	55 ng/dL
Thyroid-stimulating hormone	Serum	2.4 mIU/L
Thyroxine (T ₄)		
Free	Serum	1.2 ng/dL
Total	Serum	8 μ g/dL

The findings laboratory tests of Mr. B is given in ► **Table 2**.

Table 2 Mr. B—Routine blood test apart from slightly raised liver enzymes (OT: PT 1:3) was within normal limits.

Test	Specimen	Value
Alkaline phosphatase	Serum	0.7 μ kat/L
δ -Aminolevulinic acid	Serum	1.32 μ mol/L
Aminotransferase, alanine	Serum	28 U/L
Aminotransferase, aspartate	Serum	91 U/L
Ammonia	Plasma	38 μ mol/L
Amylase	Serum	94 U/L
Antidiuretic hormone	Plasma	0.09 pmol/L
Bilirubin		
Direct	Serum	0.3 mg/dL
Total	Serum	1 mg/dL
Cholesterol		
High-density lipoprotein	Plasma	36 mg/dL
Low-density lipoprotein	Plasma	112 mg/dL
Total (TC)	Plasma	194 mg/dL
C-reactive protein	Serum	0.3 mg/dL
C-reactive protein, highly sensitive	Serum	0.09 mg/L
Creatine kinase	Serum	64 U/L
Creatinine	Serum	0.7 mg/dL
Erythrocyte sedimentation rate	Blood	9 mm/h
RBC	Blood	330 ng/mL
Serum	Serum	9.2 ng/mL

(Continued)

Table 2 (Continued)

Test	Specimen	Value
Glucose		
2-h postprandial	Plasma	112 mg/dL
Fasting	Plasma	85 mg/dL
Hemoglobin:	Blood	14 g/dL
Hemoglobin A _{1c}	Blood	4.2%
Platelet count	Blood	192 × 10 ³ /μL
Potassium	Serum	4.5 mEq/L
Testosterone	Serum	755 ng/dL
Thyroid-stimulating hormone (TSH)	Serum	2.8 mIU/L
Thyroxine (T ₄)		
Free	Serum	1.6 ng/dL
Total	Serum	10 μg/dL

Physical Findings

Height 1.61 m, weight 64 kg, cardiorespiratory compensation. The skin and visible mucosa were well perfused, abdomen was slightly above the chest level, slightly distended, soft and insensitive, increased pigmentation along the medial line (linea nigra) was seen. Neurological status was normal, no neurologic events.

No significant physical finding noticed in husband Mr. B

Mental Status Examination

Mrs. A—conscious, oriented toward herself, other persons, to place, and oriented in time. Verbal communication established, with some delay in response, giving short answers. Thought revealed delusion of pregnancy and delusion of persecution. Mood was sad and affect was depressed. Perceptual abnormality in the form of auditory hallucination 2nd person of her baby calling to her (“maa”) from womb heard in both the ears, clearly and equally. Insight was absent.

Mr. B—conscious oriented to time, place, and person. Verbal communication established. Thought revealed delusional belief that his wife was pregnant. Mood and affect were anxious. Insight was absent.

Course during Hospital Stay

The couple was admitted in psychiatric ward and was separated. Mrs. A was started on Tab. Quetiapine and dose was gradually titrated to 600mg during her 3 weeks stay. In the initial 7 days, no improvement was seen and patient refused to see her relatives and would become irritable on seeing them. She often refused food. During second week of admission, patient became much calmer and agreed to see her relatives. Gradually toward the third week in planned psychotherapy sessions conducted with Mrs. A alone, sessions along with Mr. B, patient's delusion seemed to falter along when diagnostic blood reports and USG reports that she had not believed earlier were explained to her. Mr. B played a

significant role during sessions (who had improved) in convincing the patient against her false belief. Mrs. A admitted that she felt alone when her husband left for work and often blamed herself for her inability to sustain pregnancy. She often felt inadequate during family functions and therefore had stopped attending them. Significant improvement was noted in patients toward the end of 3 weeks, her auditory hallucinations had stopped, mood had improved and she no longer believed that she was pregnant.

She was discharged on March 13, 2019 when the patient and relatives requested that they wanted to continue treatment on outpatient basis.

Mr. B was started on Tab Olanzapine and dose was gradually titrated to 15 mg. Mr. B initially was angry on separation from his wife but was much more co-operative. Improvement was seen in the initial 10 days when in psychotherapy session Mr. B admitted that it was his deep-seated desire to be a father, and he often felt frustrated and depressed when he saw his coworkers talk about their kids or when he attended their birthday parties. When confronted with test reports indicative that his wife was not pregnant, Mr. B initially was confrontational but eventually came to terms with reality. He played a much bigger role during planned psychotherapy sessions with the wife and toward the end of 3 weeks stay Mr. B was relatively asymptomatic and was discharged on request with his wife.

After 2 months, Mrs. A with her husband moved back to her in-laws and have been regularly compliant. On subsequent follow-ups first fortnightly and then monthly both have been relatively asymptomatic.

Discussion

Folie-à-deux is characterized by the transfer of delusions from one person to another. Both persons are closely associated for a long time and typically live together in relative social isolation. In its most common form, the individual who first has the delusion (the primary case) is often chronically ill and typically is the influential member in a close relationship with a more suggestible person (the secondary case) who also develops the delusion. The person in the secondary case is frequently less intelligent, more gullible, more passive, or more lacking in self-esteem than the person in the primary case. If the pair separates, the secondary person may abandon the delusion, but this outcome is not seen uniformly.¹

The duo who presented clearly fit the diagnosis of folie-à-deux with Mrs. A, the wife, who is clearly the dominant partner in the relationship. In this case, it is clear that Mrs. A was the inducer who first developed delusional pregnancy, while Mr. B was induced one who was over dependent on the wife. Gradually as they begin to live in social isolation, away from family members and friends may have influenced the development of shared psychosis.

Treatment options should also be kept in mind as antipsychotics themselves can increase prolactin levels and can lead to amenorrhoea and galactorrhea and can further strengthen patient's belief about her pregnancy.

Of note was the significant and early improvement in Mr B when the duo was separated.

Conclusion

Perhaps the most important therapeutic step in the treatment of folie-à-deux is separation of the inducer and the induced. This case clearly demonstrates that treatment options and separating the duo lead to significant improvement in the induced and the induced can have a significant role to play in breaking the delusional belief system of the inducer.

Conflict of Interest

None declared.

References

- 1 Sadock BJ, Kaplan HI, Sadock VA, Kaplan & Sadock's Synopsis of Psychiatry. Philadelphia: Lippincott Williams & Wilkins; 2007
- 2 Seeman MV. Pseudocyesis, delusional pregnancy, and psychosis: the birth of a delusion. *World J Clin Cases* 2014;2(8): 338–344

- 3 Azizi M, Elyasi F. Biopsychosocial view to pseudocyesis: a narrative review. *Int J Reprod Biomed (Yazd)* 2017;15(9):535–542
- 4 Grover S, Sharma A, Ghormode D, Rajpal N. Pseudocyesis: a complication of antipsychotic-induced increased prolactin levels and weight gain. *J Pharmacol Pharmacother* 2013; 4(3):214–216
- 5 Ahuja N, Moorhead S, Lloyd AJ, Cole AJ. Antipsychotic-induced hyperprolactinemia and delusion of pregnancy. *Psychosomatics* 2008;49(2):163–167
- 6 Cramer B. Delusion of pregnancy in a girl with drug-induced lactation. *Am J Psychiatry* 1971;127(7):960–963
- 7 Shankar R. Delusion of pregnancy in schizophrenia. *Br J Psychiatry* 1991;159:285–286
- 8 Manjunatha N, Saddichha S. Delusion of pregnancy associated with antipsychotic induced metabolic syndrome. *World J Biol Psychiatry* 2009;10(4 Pt 2):669–670
- 9 Bera SC, Sarkar S. Delusion of pregnancy: a systematic review of 84 cases in the literature. *Indian J Psychol Med* 2015;37(2): 131–137
- 10 Lasègue C, Falret J. La folie à deux. *Ann Med Psychol (Paris)* 1877;18:321–355