Folie à Deux in Monozygotic Twins with Cerebral Palsy

Dimitry Francois [1], Evan Bander [1], Mark D'Agostino [1], Alec Swinburne [1], Lauren Broderick [1], Michael B. Grody [1], Annaheta Salajegheh [1]

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Introduction

Folie à deux or shared psychotic disorder was first described by Lasègue and Falret in 1877 (1) as a "mental contagion" such that delusional ideas from one person were transferred to at least one other person closely associated with the primary affected person. Folie à deux is an uncommon condition which has long attracted clinical attention. Here we describe the first report of folie à deux in monozygotic twins with cerebral palsy who presented with a shared somatic delusion that they strongly smelled of feces.

Case Summary

Sister 1 and Sister 2 were born in 1980 as monozygotic twins. Per maternal history and medical record review the patients were identified as monoamnionic in their mother's womb, born 10 weeks premature, required NICU hospitalization, and were subsequently diagnosed with mild cerebral palsy. Their mother, father, and two brothers had no history of psychotic illness; there is a paternal aunt with bipolar disorder. Sister 2 underwent multiple orthopedic surgeries for complications of cerebral palsy (CP), leaving her with a profound limp. However, both twins have persistent, subtle right lower extremity weakness as a result of CP, but no apparent cognitive deficits. Per mother, both sisters were "bullied" through school but Sister 1 always tried to "protect" Sister 2. Both twins finished high school and college without special education, and worked in administrative positions. Neither had any prior evidence of psychiatric illness, neither married nor had children, and after moving from their parent's home were domiciled together in an apartment. There is no evidence or history of substance abuse in either twin.

In 2011, without a clear precipitating event, Sister 1 began to believe that she was unable to control the passing of flatus. She also experienced auditory hallucinations of a voice telling her "leave, leave." Her sister and co-workers reportedly did not notice any changes in her odor, but due to the insistent nature of her complaints, asked her to seek medical attention. She did seek medical attention, and over the course of the following two years she changed her diet in various ways, tried pelvic floor physical therapy, and eventually sought surgical treatment. She had two surgeries, a posterior colporrhaphy in October 2012 and a colon resection in February 2013. Her delusions persisted and intensified, and at the end of May 2013 Sister 1 sought evaluation for depression on her own. She was seen in a two-part evaluation because of her bizarre symptom cluster and concern for psychosis. However, further research into her medical complaints revealed that they were at least in part based in reality; she had gastrointestinal motility studies which demonstrated *paradoxical puborectalis contraction*, a problem with defecation which could be due, in part, to the CP. She was accepted to the clinic for treatment of anxiety, was not found to be depressed and was found to have moderate anxiety. We mainly accepted her for further evaluation and to ensure that she minimized medical harm to herself with further surgeries as we were concerned that her delusions might trigger unnecessary surgical procedures. She was pre-

[1]*Weill Cornell Medical College-Psychiatry, White Plains, NY*

Address for correspondence: Dimitry Francois, MD, Weill Cornell Medical College-Psychiatry, 21 Bloomingdale Road, White Plains, NY 10605
Phone: 914-997-5770; Fax: 914-997-5770;
E-mail: dif9013@med.cornell.edu

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scribed lorazepam initially for the anxiety and mirtazapine later for insomnia, as well as for self-reported depression in light of gastrointestinal problems. She never took either medication, claiming that her psychiatrist was lying to her because she told her that she didn't smell anything when she was in her office on several occasions. Sister 2, who lived with her sister in 2011 when the symptoms began, initially did not adopt her complaints or note any odor. During this time they worked on opposite schedules (day and night shifts respectively), and had minimal daily contact.

However, in December 2012 after Sister 1 stopped working in order to "protect" people at work from her odor, Sister 2 began to have similar delusions. Sister 2, the more "submissive" twin per the patient's mother, quickly reported auditory hallucinations, hearing "leave, leave, leave" periodically emanating from heating vents in the ceiling. Sister 2 stopped working within a few months. They reported frequenting emergency departments to seek "cures" for their odors, and spent days isolated in a park in order to spare others from their fecal odor. Without income, they were eventually evicted from their apartment and domiciled in a shelter. For both Sister 1 and Sister 2 there were no reports of any hostile or aggressive behavior, neurovegetative depressive symptoms, or evidence of mania preceding hospitalization. They were brought into the psychiatric emergency department by their father for further evaluation.

Both twins were admitted to a psychiatric hospital, diagnosed with schizophrenia and placed on different units so as not to potentiate symbiotic pathological behavior (see discussion for more details). During the initial two weeks of their hospitalization both twins were started on risperidone. Despite separation, both continued to manifest similar somatic and persecutory delusions, as well as ideas of reference. Of note, Sister 1 was noted to become irritated and agitated following telephonic conversations with her sister. As a result, her treating psychiatrists recommended a complete cessation of contact. After two weeks on risperidone—titrated up to 6 mg orally daily—Sister 2 noted a decreased frequency of auditory hallucinations. However her somatic delusions remained firmly fixed and she continued to isolate: sitting by the windows, avoiding eating meals in common areas and not attending groups without active coaxing due to a stated intent of minimizing others' exposure to her "odor." MRI obtained showed white matter changes consistent with CP that were unchanged from prior imaging, and no evidence of temporal lobe seizures or pathology. She was cross-tapered to olanzapine, and the dose was titrated up to 15 mg orally daily over the next two weeks. Concurrent with the titration she had no contact with her sister. Within these two weeks her auditory hallucinations ceased, her affect brightened, and her delusions, while still present, were far less fixed. She was discharged to a partial hospital program and endorsed a willingness to return to work.

Sister 1, who as above was more dominant in the relationship, had less difficulty socializing, and was able to attend most groups despite initially insisting that she was malodorous and that other patients were talking badly about her. Despite disagreeing with the treatment team, she remained pleasant and in good behavioral control. Risperdal was titrated up to 6 mg orally daily with good effect, with no cross-taper required. Weeks ahead of her sister, Sister 1 had a cessation of auditory hallucinations, demonstrated insight into her delusional thoughts, and endorsed improved mood. She was discharged to the shelter and her outpatient therapists, also with an intent to return to work.

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Discussion

Dewhurst and Todd (2) proposed the following criteria when diagnosing folie à deux:

1. intimate association between partners in the delusion;
2. identical delusional content between partners; and,
3. partners share, support, and accept both their own delusion and that of their counterpart.

In this dynamic of transference, the dominant/principle initiates the delusion and the submissive/associate acquires the delusion secondarily. According to Soni and Rockley (3), most cases of folie à deux occur in pairs with social isolation. Additionally, the principle often has clinical features consistent with paranoid schizophrenia, while the associate presents with socio-clinical personality vulnerabilities including "seclusiveness, suggestibility, or dependence on the principle." Such personality vulnerabilities can be of a "prepsychotic" pathology, including "suspicious, histrionic, dependent, or antisocial traits" (4). Specifically, Soni and Rockley (3) postulate that the associate tends to rationalize and identify with the principle to reduce their own anxiety. As such, both ego defense mechanisms and inherent personality traits appear to underlie an associate's vulnerability to

folie à deux. Additionally, there appears to be evidence for a biological correlate for folie à deux. Folie à deux appears to be more common in genetically related individuals (5). In the case of identical twins, there appears to be a powerful link between genetics, complex psychodynamic relationships, and shared environmental forces that could promote the acquisition of folie à deux (6). Specifically, there appears to be a "genotype-environment correlation" such that "any delusional thinking in one twin would tend to be reinforced by the other" (7). Given this dynamic positive feedback loop, the psychological similarity, parallel life experiences and unique interpersonal experiences combined with the shared genetic loading, folie à deux in monozygotic twins creates a unique interplay of both nature and nurture in the presentation of shared psychosis (6). The primary and recipient/induced individuals in folie à deux often demonstrate close emotional associations. In this case, the two patients were identical twins, whose close relationship was further entrenched due to their shared medical diagnosis of cerebral palsy, their social isolation, cohabitation in a shelter, and the emotional stress of being bullied from a young age due to their physical disabilities. Perhaps unsurprisingly, the primary sister was considered the "stronger," more dominant sister. From a young age, the primary sister required fewer surgeries after their premature birth and was characterized by their mother as the protector of the induced sister. This relationship dynamic likely established a psychological framework ripe for the development of the shared delusion in the induced sister (4).

Gralnick (5), in reviewing a number of cases of folie à deux, was able to separate folie à deux into four subtypes: folie imposée (the dominant person with delusions imposes his or her delusions on a younger, more submissive person), folie simultanée (the simultaneous appearance of an identical psychosis occurs in two intimately associated and morbidly predisposed individuals), folie communiquée (the recipient develops psychosis after a long period of resistance and maintains the symptoms even after separation), and folie induite (new delusions are adopted by an individual with psychosis who is under the influence of another individual with psychosis). The folie communiquée subtype best characterizes these two sisters, since the induced patient was initially skeptical of the primary's delusion, but once adopted, the delusion proved difficult to extinguish. Folie à deux in identical twins has been identified as particularly treatment resistant (8). In this case we believe the treatment was successful (at least until before the patients were discharged) due to the proper recognition of the condition and situation, the fact the sisters were treated by physical separation into different units of the hospital and by the use of an atypical antipsychotic for both patients. Risperidone was chosen because the option for intra-muscular administration was ideal for the medication-naive and potentially noncompliant sisters. The primary sister's delusions resolved more rapidly than the induced patient, who continued to perseverate on the delusion despite frequent reality testing.

This example of shared psychotic disorder in monozygotic twins with cerebral palsy is a good illustration of the interactions between common genetic make-up, common and individual environmental agents and the nature of relationship between the inducer and the induced.

The problem of chronic organic brain dysfunction and its etiological role in various psychiatric diagnoses, whether presenting classically or in an atypical manner, has been a point of much discussion and controversy. Rutter (9) in his report to the Third W.H.O. Seminar on Psychiatric Diagnosis pointed out that the diagnosis of cerebral palsy does not leave room for differentiation "between a child with cerebral palsy, but no psychiatric problems, and a child with cerebral palsy who also showed a psychosis or neurosis." There are few descriptions of presentation of acute psychosis in this population, and this is the first report that we know of regarding a shared psychotic disorder involving two individuals with CP. Some descriptions (10, 11) describe acute onset, florid and fluctuating symptoms successfully treated with antipsychotics. If the nonprogressive brain lesions that characterize cerebral palsy play an etiological role in various complex psychiatric presentations, such as the folie à deux in this case, the question beckons—by what underlying, and perhaps characteristic, mechanisms?

Considerations could include the lesions themselves, the associated intellectual impairment and effect on sensorimotor and speech/language functioning, environmental protective and risk factors, and psychosocial defensive and adaptive styles which may be more prone to psychotic-level manifestations. The successful management with neuroleptics in this and other cases seems to point to a standard that would not deviate from treatment of such symptoms in populations without evidence of early developmental nonprogressing brain dysfunction. Reviewing the literature, Shiwach and Sobin found that delusional disorder is the most common primary diagnosis in folie à deux of twin pairs, as compared to schizophrenia in non-twin pairs. Psychosis

secondary to the general medical condition of cerebral palsy must also be considered in the sisters. However, based on EEG and MRI, the contribution of cerebral palsy was less likely one of biology as opposed to psychosocial priming of parallel life experiences and emotional identification.

Conclusion

Folie à deux is a rare phenomenon. This example of shared psychotic disorder in monozygotic twins with cerebral palsy is a good illustration of the interactions between common genetic make-up, common and individual environmental agents and the nature of relationship between the inducer and the induced. Our case suggests that treatment with risperidone can lead to a good clinical outcome with a remission of the psychotic symptoms.

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