Diagnostic conundrum: A rare case of psychosis in leptospirosis among siblings with folie à trois

Tengku Mohd Saifuddin Tengku Kamarulbahri[1], Rosliza Yahaya[1]
[1]M. D., Deparment of Psychiatry & Mental Health, Faculty of Medicine, Universiti Sultan Zainal Abidin, Malaysia
https://orcid.org/0000-0002-9770-5685-https://orcid.org/0000-0002-5729-2193

SUMMARY

Folie à Trois is a rare presentation of psychosis. It has been described in several case reports, but there is a lack of studies to understand the mechanism and management of this disease. We presented a case of three siblings who share the same delusional idea initially induced by one of the sisters who had neuropsychiatric manifestations of leptospirosis. The siblings exhibited a shared pattern of aggressive behaviors, auditory hallucinations, and persecutory delusions toward each other. Intriguingly, two of the sisters displayed improvement even without pharmacological intervention. The case highlights the complexity of differentiating shared psychotic disorder from organic etiologies, emphasizing the importance of multidisciplinary assessments in unraveling intricate clinical presentations. The distinctive temporal resolution of psychiatric symptoms among the siblings underscores the need for nuanced diagnostic considerations in shared psychotic disorders. This case report contributes to the understanding of the interplay between infectious diseases and psychiatric manifestations, urging clinicians to exercise meticulous scrutiny in cases of shared psychotic disorders masquerading as organic illnesses.

Keywords: Folie à trois, Leptospirosis, Shared delusion

INTRODUCTION

Folie à deux, or shared psychotic disorder, demonstrates inherent distinctions when juxtaposed with other psychiatric conditions (1). This uncommon entity is characterized by the transmission of psychotic symptoms from one individual (the inducer) to another (the induced) (2). This condition may arise in individuals living in close emotional proximity to those with mental illness, particularly psychotic disorders (3). Building upon this framework, folie à trois, an extension of the concept, describes a scenario where the shared delusion involves three individuals (4). The diagnosis known as "Shared Psychotic Disorder" was included in the DSM-III and DSM-IV. However, in the DSM-5, it no longer exists as an independent diagnosis but is instead classified under other specified schizophrenia spectrum disorders (5). The concept has since evolved, and according to the ICD-10, the diagnosis is now based solely on phenomenology. However, there is still a lack of information on the prevalence, natural history, and optimal treatment of folie à deux, and its etiology remains unknown (6).

We presented a case report of three siblings who presented with psychosis. One of them was positive for leptospirosis. Two of them had no evidence of leptospirosis or brain infection; however, they still manifested psychotic symptoms that resolved later, even without antipsychotics.

Leptospirosis, a globally distributed zoonosis, is prevalent in tropical and temperate regions, particularly in emerging nations in Oceania, the Caribbean, Latin America, sub-Saharan Africa, and South and Southeast Asia. These geographical areas experience elevated mortality and morbidity rates associated with the disease (7). However, the occurrence of neuropsychiatric manifestations in leptospirosis is infrequent, as documented in the literature through case reports and case series.

Despite the rarity of neuropsychiatric manifestations in leptospirosis, this case report aims to explore the potential link between leptospirosis and shared psychotic disorder. Our hypothesis is that leptospirosis might contribute to or exacerbate the presentation of psychotic symptoms in individuals with preexisting close emotional ties, thereby

DOI: 10.5505/kpd.2024.87528

Cite this article as: Tengku Kamarulbahri TMS, Yahaya R. Diagnostic conundrum: A rare case of psychosis in leptospirosis among siblings with folie à tois. Turkish J Clin Psych 2024; 27:331-334

The arrival date of article: 04.05.2024, **Acceptance date publication:** 31.07.2024

Turkish J Clinical Psychiatry 2024;27:331-334

influencing the development of folie à trois. By examining this unique case of three siblings, including one diagnosed with leptospirosis, we aim to shed light on the possible neuropsychiatric effects of leptospirosis and its impact on the dynamics of shared psychotic disorder. This study intends to contribute to the existing literature by providing insights into the relationship between infectious diseases and psychotic manifestations, potentially guiding future clinical assessments and management of similar cases.

CASE REPORT

Mrs. A, a 26-year-old woman residing with her sisters, Mrs. B (38-year-old) and Mrs. C (35-year-old), presented to the emergency department with aggressive behavior for three days before admission to the medical ward. Initially observed in Mrs. A and subsequently in Mrs. B and Mrs. C, all three siblings exhibited similar symptoms, including self-directed conversations, wandering, disrupted sleep, poor appetite, auditory hallucinations (commanding in nature, instructing them to eat and inflict harm), and aggressive behaviours such as biting. The siblings harbored persecutory delusion towards each other, accusing one another of malicious intentions, leading to verbal altercations. They also firmly believe that the villagers are plotting bad things toward them. They had no symptoms of manic or depressive symptoms. Mrs. A has no known history of pre-existing medical conditions. The family history indicated no known occurrences of mental illness.

Upon admission, the mental state examination revealed that the patients admitted to experiencing second-person auditory hallucinations and visual hallucinations of seeing ghosts. Additionally, the patients exhibited paranoid delusions.

They showed no evidence of delirium based on the Confusion Assessment Method (CAM) and lacked signs of infection except for Mrs. A, who had a three-day fever prior to admission. Mrs. A was tachypneic with low-grade fever, while other siblings' vital signs and temperature were normal. Their neurological examinations were normal, and there were no signs of neck stiffness.

Baseline investigations, including total white cell counts, were unremarkable, except for elevated liver function tests in Mrs. A. Mrs. A also had increased creatinine kinase, erythrocyte sedimentation rate, and C-reactive protein. Screening for HIV, Hepatitis B, Hepatitis C, and blood malarial parasites yielded negative results. MRI brain scans and lumbar punctures were both unremarkable.

A notable finding was the positive serum IgM Leptospirosis test in Mrs. A. It was done because all of them come from remote areas known for several cases of leptospirosis. Hence, Mrs. A was admitted to the medical ward. Her siblings were also admitted to the medical ward for observation. Intravenous Acyclovir and Ceftriaxone were administered to all three sisters to address potential encephalitis and leptospirosis. Subsequently, the intravenous Acyclovir and Ceftriaxone were off for Mrs. B and Mrs. C on day three of admission because of no sign of infection, and the psychotic symptoms also improved. The psychiatric team diagnosed Mrs. A with a psychotic disorder due to leptospirosis. Concurrently, the psychiatric team also identified shared delusional disorder (folie à trois) for Mrs. B and C. The psychotic symptoms were managed with Haloperidol (up to 5 mg/day) for Mrs. A. However, antipsychotics were not given to Mrs. B and C. Mrs. B and C, separated from Mrs. A in the ward, demonstrated improvement in psychotic symptoms even without antipsychotics and were discharged earlier. Mrs. A's psychosis resolved on day 20, leading to her discharge. Follow-up assessments revealed no psychotic symptoms in all of them, who functioned normally without antipsychotics.

DISCUSSION

Leptospirosis, a biphasic febrile illness prevalent in tropical and subtropical regions, poses a substantial global health concern as a neglected tropical zoonotic disease (8, 9). The clinical spectrum of leptospirosis manifests across a range of severity, from mild influenza-like symptoms to severe syndromes indicative of multiorgan failure (9). The disease's potential for misdiagnosis, exacerbated by nonspecific symptoms, underscores the gravity of its impact, particularly in the severe form known as Weil's disease, which often leads to fatal outcomes, primarily due to complications such as renal failure

(8). While primary neuroleptospirosis is a rare occurrence, it has been documented in the literature through case reports and case series (10).

The pathophysiological mechanisms underpinning leptospirosis remain incompletely understood, with a dearth of knowledge in the literature regarding the intricate cellular interactions facilitated by Leptospira (7, 8). Leptospira species exhibit a notable capacity to adhere, invade, and replicate within host cells, necessitating an urgent refinement of research efforts to elucidate their precise pathophysiological framework (8). The leptospiral endotoxin, glycolipoprotein (GLP), targets Na/K-ATPase at the molecular level, inducing lipotoxicity that inhibits Na/K-ATPase activity. This mechanism contributes to diverse clinical manifestations in various organs and tissues, compromising nerve impulse generation and conduction in neurons and excitable cells (7).

Diagnosing leptospirosis relies on the recovery of the organism through culture, macro agglutination tests, and dark field microscopy. Recognition of manic and psychotic symptoms, along with fever and elevated transaminase as well as CK levels, particularly in high-risk occupational groups during rainy periods, should alert physicians to the potential presence of leptospirosis (11). Notably, healthcare professionals working in regions with high leptospirosis incidence must be attuned to cases where the primary presentation is neurological, facilitating prompt diagnosis and implementation of appropriate treatment strategies (9).

Mrs. A was diagnosed by the psychiatric team with a psychotic disorder due to leptospirosis. Simultaneously, the team identified folie à trois in the case of Mrs. B and C. Folie à trois, also referred to as shared psychotic disorder, presents an atypical psychiatric condition wherein delusional convictions are transmitted from one individual to one or more susceptible individuals closely connected. As of now, this remains an infrequent yet intricate psychiatric diagnosis (12). This rare psychotic syndrome, alternatively termed Folie à trois, Shared Psychosis, or Induced Delusional Disorder, involves the transfer of delusional beliefs or aberrant behavior from one individual to another or to others closely associated with the primary affected person (3). Notably, only the inducer experiences an authentic psychotic disorder, while the induced individuals typically recover following separation from the inducer (2).

Folie à trois, an infrequent mental disorder, manifests in situations where close emotional bonds exist among two or more individuals, with only one of them experiencing a genuine psychotic disorder. The delusions originating from the inducer are transmitted to individuals who come into contact, ceasing upon separation from the inducer (3). Recognized by a collective adherence to delusional beliefs, shared psychotic disorders typically arise within pairs or groups characterized by close relationships and social isolation. Significantly, the cognitive and emotional functions of those affected by shared psychotic disorders generally remain unaffected, contributing to the scarcity of identification, diagnosis, and treatment. The clinical manifestation of this disorder encompasses a multifaceted aspect, extending beyond the traditional focus on delusions, potentially elucidating the conflicting outcomes observed with various treatments (1). Mental illness in the dominant individual often assumes a schizophrenic nature, with initial fixation and induced delusions exhibiting a chronic nature and often involving ideas of persecution, influence, poisoning, or grandeur. Deluded thoughts are transmitted under specific circumstances, characterized by close group contacts and isolation from alternative languages, cultures, or geographies (3).

Folie à trois has emerged as a condition marked by heterogeneity, characterized by a complex etiopathogenesis (1). Notably, there exists a lack of consensus among researchers regarding the uniformity of opinions concerning the incidence of this disorder in relation to gender, age groups, and various interpersonal relationships, such as those between partners, siblings, parents, and children. However, a prevailing consensus acknowledges long-term social isolation as a common risk factor, alongside dominance and potent power of suggestion in one partner, complemented by passivity and susceptibility to suggestion in the other (2). The phenomenon of induced delusions within this context can be perceived as a manifestation of a "learning error," wherein the recipient becomes persuaded by delusional interpretations of events due to distorted perceptions. Functional imaging analysis reveals disparate activities in the same areas of the

cerebral cortex between the inducer and the recipient. Specifically, there is an augmented volume and localization of activation in the cerebral cortex of the inducer compared to the recipient (13). This intricacy underscores the multifaceted nature of Folie à deux and invites further exploration into its underlying mechanisms.

Induced delusions, autonomously and without therapeutic intervention, may dissipate upon the separation or isolation of the affected individuals from the mentally ill person who triggered the onset of psychotic symptoms (3). The therapeutic approach for shared psychotic disorders encompasses the separation of individuals involved, coupled with pharmacotherapy utilizing antipsychotics. This case introduces a distinctive ethical dilemma wherein the psychiatric team was summoned to assess a patient and discovered both the patient and another individual exhibiting symptoms. In conclusion, the consideration of a shared psychotic disorder becomes pivotal in the differential diagnosis when encountering cases of psychosis characterized by delusional systems on medical floors (14). The multifaceted nature of this disorder necessitates a comprehensive approach to understanding and addressing its intricacies.

In conclusion, this comprehensive case study highlights a unique association between leptospirosis and shared psychotic disorder (folie à trois) in three siblings, a connection not previously documented. Unlike other studies, which typically do not link infectious diseases with shared psychosis, our findings suggest leptospirosis can trigger psychotic symptoms. The successful resolution of psychosis in two siblings without antipsychotics, solely through separation and supportive care, further underscores a novel, non-pharmacological approach to managing such conditions. This study contributes valuable insights into the differential diagnosis and treatment of psychotic disorders in endemic regions.

Correspondence address: M.D., Tengku Mohd Saifuddin Tengku Kamarulbahri, Deparment of Psychiatry & Mental Health, Faculty of Medicine, Universiti Sultan Zainal Abidin, Malaysia drtgsaifuddin@gmail.com

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