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CASE REPORT

Delirious mania in an adolescent with Bardet-Biedl syndrome : A case report

Syeda Rawnak Jahan[1] | Tanbir Ahmed[1] | Afsana Benta Anowar[2] | Faisal Rahat[1] | Md. Munaim Reza[3]

[1]Department of Psychiatry, Bangabandhu Sheikh Mujib Medical University (currently, Bangladesh Medical University), Dhaka, Bangladesh
[2]Department of Psychiatry, National Institute of Mental Health and Hospital, Dhaka, Bangladesh
[3]Department of Psychiatry, Enam Medical College and Hospital, Savar, Bangladesh

Correspondence
Tanbir Ahmed
tanbir200@gmail.com

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Responsible editor
Md Nahiduzzamane Shazzad
0000-0002-8535-4259

Reviewers
MM Jalal Uddin
0000-0003-0402-7457
Hurjahan Banu
0000-0002-8115-1761
Abed khan
0000-0002-4151-2817

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Introduction

Bardet-Biedl syndrome is a multisystemic autosomal recessive disorder characterised by a range of phenotypic manifestations. Its frequency is 1 in 120,000 in North America, 1 in 160,000 in Europe, and 1 in 36, 000 in Kuwait [1]. Bardet-Biedl syndrome is marked by a set of core features as retinal dystrophy, obesity, polydactyly, hypogonadism and cognitive impairment historically referred to as the "Bardet-Biedl syndrome pentad." These manifestations are commonly attributed to underlying defects in primary cilia function [2, 3].

It is often associated with psychomotor slowness, infantile behaviour, and emotional instability in response to stimuli [3]. Delirious mania has been rarely documented [4, 5], although other mental symptoms of Bardet-Biedl syndrome have included hallucinations, delusions, depressed mood, manic symptoms, and catatonic features [4]. Delirious mania is regarded as a variant form of classical bipolar disorder [6]. This severe but frequently undiagnosed neuropsychiatric illness is characterised by the abrupt development of psychosis, manic symptoms, and delirium [5].

This example illustrates the challenge of recognising and treating a patient exhibiting perplexity and manic excitement, particularly in those with complex comorbidities.

Case description and management

A 17-year-old girl, the first child of consanguineous parents presented at Bangabandhu Sheikh Mujib Medical University (currently, Bangladesh Medical University), Dhaka, Bangladesh with characteristics that were typical of Bardet-Biedl syndrome. Phenotypically, the diagnosis was determined based on the presence of distinctive clinical characteristics such as developmental delay, post-axial polydactyly, obesity, and retinal degeneration (Figure 1). She had significant mental symptoms, hypertension, diabetic renal disease, hypothyroidism, and type-2 diabetes mellitus also, among other comorbidities for last 2-years.

The patient had auditory hallucinations, affective disturbances like mood swings, increased energy, crying spells, and emotional instability, cognitive impairment, and behavioural issues like irritability, self-harm, insomnia, treatment non-adherence, violent behaviour, and refusal of medical intervention, which worsened over the past week. Her psychiatric symptoms worsened, deteriorating her overall health and complicating her comorbidities. Her severe psychiatric symptoms and refusal of treatment necessitated an urgent referral to psychiatry. The patient was irritable, talkative, aggressive, had poor eye contact, and was agitated during the mental state examination. Speech was disorganised and incoherent. She was easily distracted and skipped topics, making sustained attention and sequencing tasks difficult. Although her extremities showed negativism, a motor examination did not find any catalepsy, echophenomena, grimacing, or stereotypies. Although denied, she

Key messages

Psychiatric issues in adolescents with conditions like Bardet-Biedl Syndrome and neurodevelopmental delay can be challenging to diagnose because symptoms like mood swings, hallucinations, and aggression can mix with their existing cognitive and behavioral problems. Delirious mania is often hidden by this overlap. Early detection and multidisciplinary care are crucial for accurate diagnosis, effective treatment, and improved health and compliance.

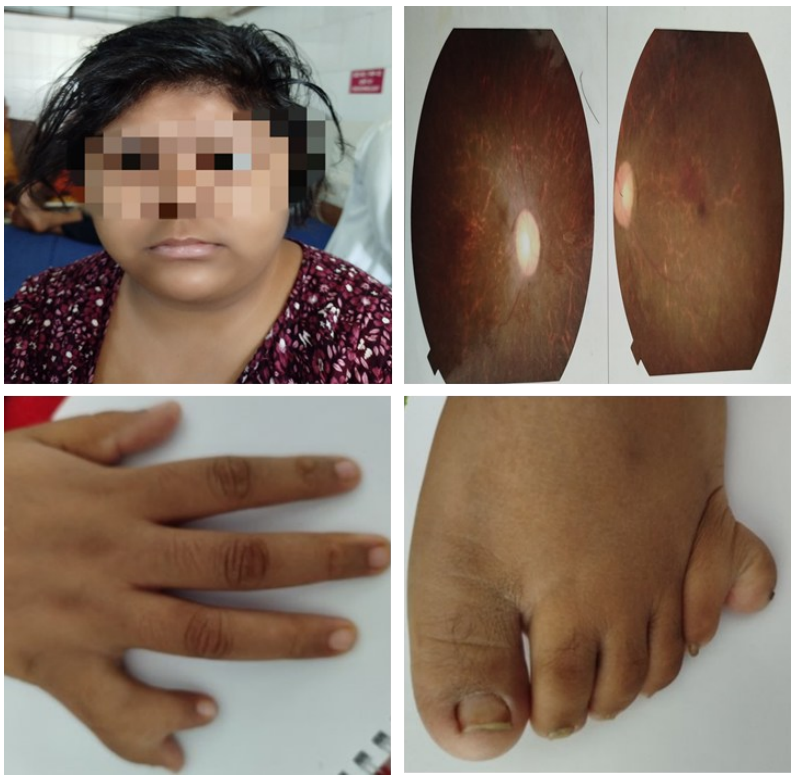

Figure 1 An adolescent girl with Bardet-Biedl syndrome

demonstrated active symptoms of hallucinations, such as self-laughing and self-muttering, indicating a lack of insight. Her attention was poor, and she was disoriented to time and place. Previous records showed that her IQ was 65. Mini–mental state examination score was 22. She had epilepsy managed with valproate for six years, remained seizure-free for five years, and had been off medication for the past year. There was no family history of Bardet-Biedl syndrome. Her medical conditions were treated according to standard protocols by the endocrinologist and nephrologist. Laboratory investigations and organic delirium or catatonia were ruled out on clinical grounds. The psychiatric diagnosis was clinical delirious mania, characterised by the simultaneous presence of both delirium and manic symptoms. Interventions, including rapid tranquilisation, de-escalation strategies, pharmacological management, and psychoeducation for the caregivers, were initiated.

She was prescribed lorazepam (2 mg), quetiapine (100 mg), aripiprazole (10 mg), topiramate (25 mg), and melatonin (3 mg), resulting in significant improvement in her psychiatric symptoms. Over the course of a three-month follow-up, including an initial two-week inpatient stay, her agitation, hallucinations, and disorientation resolved, as confirmed by mental state examination. Her mini–mental state examination score increased from 22 to 26, behavioural disturbances lessened, and treatment adherence improved. Despite experiencing mild mood symptoms, her attention, insight, and cooperation were enhanced, allowing her to safely transition to outpatient psychiatric and endocrinological care.

Discussion

She exhibited mixed delirious symptoms (both hyperactive and hypoactive) [6] along with mixed manic symptoms. When cases of delirious mania arise at the convergence of psychiatry and medicine, it can be difficult to balance the needs of behaviour, mental health, and medicine [6]. Delirium, which can have infectious or metabolic causes, was included in the differential diagnosis for this illness. However, relevant investigations and clinical examinations ruled out these. Confusion, increased hallucinations, and a noticeable escalation of manic symptoms are characteristics of the shift from mania to delirious mania [7]. Abruptly stopping medication raises the risk of mania in people with Bardet-Biedl syndrome and concomitant chronic diseases, particularly chronic kidney disease [6] as occurred here. The extremely quick onset of delirious mania is especially common among children and adolescents [7]. Literature showed the majority of patients with delirium and mania were female, younger, and had a history of bipolar disorder [8]. Of all individuals with acute mania, 5–20% exhibit delirium symptoms [7].

Bardet-Biedl syndrome genes may affect cortical development and cause major mental illness, with over 30% prevalence in psychiatric conditions [9]. Increased dopamine activity and decreased acetylcholine may affect melatonin release, circadian rhythms, and sleep patterns. These symptoms improved with the above-mentioned pharmacological treatment, which targets neurotransmitter imbalances [5, 8].

No diagnostic classifications or treatment guidelines exist for delirious mania, which makes diagnosis and treatment challenging. Given a history of Bardet-Biedl syndrome with psychosis, along with current symptoms of delirium and mania, the symptom profile and medical history can assist in therapeutic decision-making.

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Author contributions

Manuscript drafting and revising it critically: TA, SRJ, ABA. *Approval of the final version of the manuscript:* TA, MMR, SRJ, FR, ABA. *Guarantor accuracy and integrity of the work*: TA, MMR, SRJ, ABA, FR.

Conflict of interest

We do not have any conflict of interest.

Data availability statement

We confirm that the data supporting the findings of the study will be shared upon reasonable request.

Supplementary file

None

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