Case Study

“Ansa” is an eleven year-old girl who lives with Mrs. J, her mother; her parents have been divorced for years, and she has little contact with her father. Ansa’s early childhood history was marked by her mother’s numerous unstable relationships which led to the mother and daughter moving many times before Ansa was seven. Ansa was unknown to any child psychiatry service. From age six to eight, Ansa was on methylphenidate for suspected attention-deficit/hyperactivity disorder (ADHD); there were no beneficial effects, and the medication was discontinued. Otherwise, Ansa’s past psychiatric history is negative. She has never had physical illnesses, has never taken drugs, alcohol, or medication. Family history is positive for Mrs. J’s borderline personality disorder and her uncle’s possible but unconfirmed diagnosis of bipolar disorder.

Ansa was admitted to our service for grandiosity, agitation, and distractibility that had appeared in the last two days. She had no sleep, spending the nights cleaning her room. She had excessive projects, including becoming a rock star. These symptoms rendered her totally unable to attend school and even take care of her basic needs, such as hygiene. There were no recent stressors. The mental status examination showed a child unable to remain seated (spending the whole interview moving from object to object), an
irritable and labile affect, logorrhea, accelerated speech, and flight of thoughts. She expressed countless projects although there was no delusional grandiosity. She denied hallucinations and suicidal ideations. Baseline metabolic tests were negative, including complete blood count, electrolytes, thyroid levels, and urinalysis. A physical evaluation by the pediatrician yielded no significant findings. An electroencephalogram and brain magnetic resonance imaging (MRI) were negative. Ansa was diagnosed with acute manic episode with no identifiable organic etiology but with a possible biological vulnerability for emotional dysregulation, given the mother’s borderline personality disorder.

After no improvement with quetiapine 12.5mg and olanzapine 5mg trials, Ansa was given aripiprazole 5mg. With the medication and strong presence of nurses and psychoeducators, her condition was stabilized in less than five days. Due to sedation, aripiprazole was lowered to 2mg, resulting in a manic relapse. Aripiprazole was increased to 3mg, and the manic symptoms quickly resolved, again in less than five days. The hospitalisation lasted one month in order to monitor Ansa’s symptoms and level of functioning during progressive returns to school. They were ultimately successful, and Ansa was discharged.

The following week, despite compliance to medication, Ansa suffered a relapse of the identical symptoms and was readmitted. This coincided perfectly with turbulent changes in her environment. Indeed, Ansa and her mother had just moved in with family friends: there were frequent unexpected visits from various acquaintances, accompanied by daily verbal violence. Mrs. J admitted to her fragile emotional state and that Ansa would usually internalize her mother’s feelings. Since no other cause was identified, the manic relapse was attributed mainly to the unstable environment with a child in a symbiotic relationship with her mother, who suffers from borderline personality disorder but whose parental capacities were considered preserved. Given the apparently clear temporal relationship of the symptomatic relapse with the stressors, an adjustment disorder was diagnosed as part of the differential diagnosis. However, the clinical presentation – especially the mental status examination as previously described – clearly corresponded to the severity of a mood disorder. In addition, the patient’s personal and longitudinal history revealed no signs of conduct disorder or oppositional defiant disorder. The relapse also heightened the prior hypothesis of Ansa’s biological vulnerability to emotional dysregulation.

No pharmacological change was made and with the presence of the multidisciplinary team, Ansa’s condition remitted in four days. The total hospital stay lasted three weeks in order to once again observe Ansa’s evolution during progressive returns to school, which were successful. Ansa was then discharged with a follow-up by a social worker and psychiatrist. After a year of follow-up, she is doing well, and the family situation remains stable.

**Early-onset Bipolar Disorder**

Studies have shown that bipolar disorder usually begins with an index episode of depression: positive family history (Pavuluri, Birmaher, & Naylor, 2005), clinical severity, psychotic symptoms, and psychomotor retardation are well documented predictors of bipolarity. Approximately 20% of youths with a first major depressive episode will develop a manic episode. Prodromal symptoms of bipolar disorder – hyperactivity, anxiety, dysphoria – have been identified but remain nonspecific (American Academy of Child and Adolescent Psychiatry – AACAP, 2007). Ansa’s case illustrates this diagnostic complexity since prodromal symptoms must be taken in the context of environmental instability.

Early-onset bipolar disorder is often considered atypical because of the fluctuating course of symptoms and lack of clear episodes, which defines the classic phenotype (AACAP, 2007). Some authors have introduced the concept of “broad phenotype” of bipolar disorder for youths with extreme irritability, explosiveness, mood variability, and functional impairment. In bipolar disorder literature, one particularity for the pediatric population is the introduction of the concepts of “ultrarapid cycling” (hours to days) and “ultradian cycling” (minutes to hours). Geller et al. (2000) described a prepubertal and early adolescent bipolar disorder (PEA-BP) phenotype which includes both types of cycles.

Although continuity between this “juvenile mania” and adult bipolar disorder has not been established (Duffy, 2007), the prevalence of bipolar disorder diagnoses in children and adolescents is increasing dramatically, with a fortyfold increase in 1993-2004 in the USA (AACAP, 2007). The prevalence of the classic phenotype in prepubertal children is unknown but is considered rare. Ansa represents our service’s earliest manic episode and one of the few prepubertal cases.

The rate of comorbidity between juvenile bipolar disorder and ADHD has been previously described as high in numerous studies. However, a recent review by Anne Duffy (2012) stated that ADHD is not a reliable predictor for the development of juvenile bipolar disorder. The strong overlap of symptoms between the two conditions (distractibility, hyperactivity, talkativeness) raises questions about diagnostic specificity. Thus, DSM-V work groups for bipolar disorder have proposed specifying in the “B” criteria of (hypo)mania that symptoms must absolutely represent a change from baseline (American Psychiatric Association, 2012b). This proposal could help reduce the double-counting of symptoms towards ADHD and bipolarity. “Disruptive Mood Dysregulation Disorder” is a proposed additional diagnosis to target youths suffering from sustained irritability (American Psychiatric Association, 2012a; Margulies, Weintraub, Basile, Grover, & Carlson 2012).
In our opinion, the debate on early-onset bipolar disorder remains important. The AACAP states in its most recent practice parameters that “caution must be taken before applying this diagnosis in preschool children” (AACAP, 2007). The stigma of psychiatric diagnoses and the consequences of mislabeling juveniles should not be minimized. Iatrogenic risks must be weighed against risks of inappropriately treating ill juveniles especially since mood stabilizers are associated with potential serious adverse effects (Parens & Johnston, 2010). Nonetheless, a recent meta-analysis concluded in the efficacy and safety of antipsychotics in early-onset bipolar disorder, leading to the US Food and Drug Administration’s approval of several antipsychotics in juveniles, including aripiprazole (Liu et al., 2011).

Ansa’s case is striking in her fragility to psychosocial stressors and quick improvement with environmental stability and medication. There is little conclusive information on the impact of the environment on the evolution of early-onset bipolar disorder. This is a crucial point since the therapeutic approach might have to target better psychosocial follow-ups with the entire family rather than focus on individual treatments. Ultimately, these issues illustrate the need for more scientific research to better identify and treat patients with bipolar disorder.

Consent

The patient and parent have both given written consent to the publication of this paper following an explanation of the procedure. Patient anonymity has been protected.

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The authors have no financial relationships to disclose.

References


