CLINICAL PERSPECTIVES

CLINICAL CASE ROUNDS IN CHILD AND ADOLESCENT PSYCHIATRY

Trichotillomania, Bipolar Disorder and White Matter Hyperintensities in a Six-Year Old Girl

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A six-year-old girl was referred to our child psychiatry outpatient clinic by the Pediatric Neurology Unit with a diagnosis of Attention-Deficit/Hyperactivity Disorder (ADHD) and trichotillomania. She had neither eyebrows nor eyelashes. The clinical picture was of irritability, frequent tantrums, and aggressive behaviour. During the following year she presented several brief episodes of intense mood changes, which typically started with night-time onset trichotillomania and sleep disturbance. The episodes lasted no longer than five days and recurred within one or two months. A diagnosis of pediatric bipolar disorder (BD) was made after the first months of clinical follow-up. An MRI showed focal white matter hyperintensities (WMH) in T2.

Key Words: Trichotillomania, pediatric bipolar disorder, white matter hyperintensities

six-year-old girl was referred to our child psychia-Atry outpatient clinic by the Pediatric Neurology Unit with a diagnosis of ADHD and trichotillomania. She had no family history of mental illness other than a maternal uncle who had suffered from alcoholism. The older of two siblings, she had a normal developmental history and lived with her parents and a four-year-old brother. Both parents were working, the family was not socially disadvantaged and there was no marital conflict. Trichotillomania started suddenly when she went on a school trip at the age of five and in one day pulled out all her eyebrows and eyelashes. The problem ceased after a few days of mood changes and tantrums. One year later, the clinical symptoms reappeared and she was then referred to our clinic by the Neuropediatric Unit, with a diagnosis of ADHD due to conduct disorder and trichotillomania. When we first attended her as a patient, she had neither eyebrows nor eyelashes. The clinical picture was of irritability, frequent tantrums, and aggressive behaviour. She was also sad and had depressive thoughts such as "I am ugly and silly", "my mother is mean" or "nobody loves me". During the following year she presented several brief episodes of intense mood changes, which typically started with night-time onset trichotillomania and sleep disturbance (she woke during the night). The episodes lasted no longer than five days and recurred within

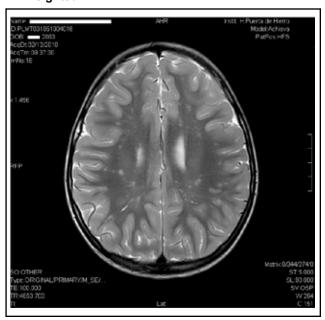
one or two months. Between episodes she would not pull out her hair and her eyebrows and eyelashes would grow normally. While attending our clinic, in a particular episode she showed manic symptoms such as grandiosity ("I am the owner of my house" "We have 8 bathrooms"), elated mood (she would try to make everybody laugh in the classroom and be very talkative), dysphoria, bizarre behaviour (trying to wear summer clothes in the middle of the winter, painting her entire body with markers), restlessness, diminished concentration, a decreased need for sleep and food refusal. She verbalized some thoughts repetitively ("I behave well and my mother doesn't love me"). However, no motor or vocal tics were observed. This episode had started the day before with night onset trichotillomania. Two days later, we saw her again and all the symptoms (including the trichotillomania) had ceased. After the episodes, symptoms completely disappeared and she would return to normal behaviour as a quiet, kind, attentive, reasonable and cooperative little girl. She had no ADHD symptoms (inattention, hyperactivity, impulsivity) and had stopped pulling out her eyebrows and eyelashes, letting them grow normally. In other episodes she had mixed manic and depressive symptoms. (The parents described how one day she would be dancing flamenco non stop and singing all day, being very talkative and saying happily how she had failed a school test, which

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Image 1. White matter hyperintensities in T2-weighted MRI

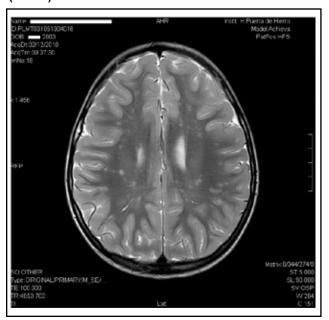


in normal circumstances would really worry her). In the periods when she seemed more depressed, she would accuse her teacher of spanking her (which proved to be false) and she would have several different complaints about her relatives not loving her and that she was ugly. She would say things like "I don't love my brother and I want him to die" or "I want to have my house full of money". During different episodes she was repeatedly obsessed about being rich and about her brother being obese. She would also wrap toys and give them away as gifts for other people. All complaints would disappear once the episodes were over. In one of her visits to our clinic she was able to depict her inner turmoil in a drawing (Image 3). The school reported frequent changes in her classroom behaviour. Sometimes she would behave normally and other days she would seem restless, distracted and wanting to be the center of attention. Her teacher was of the opinion that her behaviour was not a result of a psychiatric disorder but was only to seek attention.

The neurological examination, blood cell count, biochemistry, thyroid and prolactin hormone determination were normal, including ASO. Brain magnetic resonance imaging (MRI) was performed by sequences T1 Mprage sagital, T2 axial, flair axial and coronal and diffusion study. A faint spot pattern increase of the periatrial white matter signal intensity in strengthened sequences in T2 was observed, suggesting terminal mielination areas. Focal WMH were observed in T2, located in the superatentorial white matter. Perivascular spaces were prominent (Images 1 and 2).

A diagnosis of pediatric BD was made after the first months of clinical follow-up and this was explained to her parents. Several therapeutic options were initially offered, including

Image 2. WMH in fluid-attenuated inversion recovery (FLAIR) MRI



medication, but the parents refused pharmacological treatment. The patient was then followed up every fifteen days or on a monthly basis according to her symptomatology. Psychoeducation and symptom observation by the parents have been carried out over the last fourteen months and she has continued to have similar episodes which impair her academically, apart from their impact on family relationships and personal suffering. The parents are now considering pharmacological treatment with antipsychotics or mood stabilizers (Danielyan & Kowatch, 2005; Masi et al., 2010).

Trichotillomania is an impulse disorder that affects 1-3.5% of adolescents and young adults. However, the rates in younger children are unknown. Trichotillomania often occurs together with psychiatric comorbidity (Lejoyeux, Arbaretaz, McLoughlin, & Ades, 2002). Sah and colleagues described a subtype in preschoolers who often pull out their hair before going to sleep or while asleep (Sah, Koo, & Price, 2008). The diagnosis of BD in preschool children continues to create controversy despite the progress in validation and characterization made over the last ten years (Luby, Tandon, & Nicol, 2007). Pornnoppadol and Todd reported a case of trichotillomania and BD in a 13-yearold girl who also showed irritability and aggressive behaviour, which improved after treatment with lithium carbonate (Pornnoppadol & Todd, 1999). There have been reports of episodic trichotillomania responding to lithium treatment in young adults with type II BD (Berk, McKenzie, & Dodd, 2003; Sharma & Corpse, 2008). It has been suggested that valproate therapy may attenuate the formation of WMH in elderly patients (Yuan et al., 2009).

In the case of our patient, night-time onset trichotillomania was the first and salient symptom of manic or mixed

Image 3. Drawing done by the patient at the age of 6. She drew two houses in one: One was black, the other was pink with the rainbow, cloud and storm inside.



episodes of pediatric BD. This ceased whenever the mood returned to normality. Differential diagnosis with ADHD may be difficult and comorbidity is common, even up to 31%, according to some authors (Danielyan, Pathak, Kowatch, Arszman, & Johns, 2007). Elation, a decreased need for sleep and grandiosity are important for differential diagnosis. The same author describes a case of manic symptoms in a five-year-old boy. Racing thoughts and hypersexuality may be less frequent mania symptoms among bipolar preschoolers (Kowatch, Youngstrom, Danielyan, & Findling, 2005). Treatment with methylphenidate can worsen affective symptoms (Ross, 2006; Soutullo et al., 2002).

In BD an increased number of WMH, is one of the most consistently reported abnormalities (Mahon, Burdick, & Szeszko, 2010). In adults with BD, there is evidence which associates more severe WMH with a greater number of hospitalizations and poorer response to treatment. In pediatric BD an increased prevalence and severity of WMH has been reported (Lyoo, Lee, Jung, Noam, & Renshaw, 2002). In one study WMH were present in 67% of bipolar adolescent patients (Pillai et al., 2002).

In our case study, continuous psychoeducational work with the parents has been crucial for the acceptance of diagnosis and treatment. The findings in the MRI have helped the parents to accept the need for pharmacological treatment. However, the diagnosis of BD in young children is still a difficult task.

We believe that when dealing with trichotillomania, special attention should be paid to affective symptoms so as to rule out BD. This is particularly relevant in the treatment of very young children. We have also observed that although lithium treatment has been used in episodic trichotillomania and BD, in our experience valproic acid has also been equally effective, as in the case reported of elderly patients with WMH.

(Update: One year after commencing treatment with Sodium Valproate in monotherapy, the patient has been free of all symptoms for ten months and both trichotillomania and major affective episodes have completely disappeared.)

Acknowledgements / Conflicts of Interest

The authors have no financial relationships to disclose.

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