CASE REPORT

DOUBLE JEOPARDY - A CASE OF TRICHOTILLOMANIA AND INTELLECTUAL DISABILITY

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Abstract

Objective: The commonest age of onset for Trichotillomania is between 9 to 13 years of age and at times triggered by Depression and stress. According to literature, the best treatment is a combination of clomipramine and Cognitive Behaviour Therapy. The objective of this index case is to present a patient with an older age of onset, and with co-morbid intellectual disability and the challenges faced during treatment because of the comorbidity. Methods: A case of Intellectual disability who presented with Trichotillomania, and Trichophagia was chosen and followed up for a period of three years. *Results:* The hair-pulling behaviour in the index case was due to a strong urge, which was relieved by the behaviour and was not secondary to other symptoms. The course of trichotillomania was independent of the course of aggression as improvement in aggressive symptoms was not accompanied by improvement in hair-pulling behaviour, which responded to administration of Imipramine, though it did not improve with clomipramine and citalopram, which are the medications of choice. Conclusion: The index case suggests customizing treatment according to individual suitability, and choosing a medication that the patient is comfortable with, is important. It also suggests true comorbidity between Intellectual Disability and Trichotillomania, as the symptom of trichotillomania was not secondary to the aggressive behaviour exhibited by the patient. ASEAN Journal of Psychiatry, Vol. 16 (2): July – December 2015: XX XX

Keywords: Intellectual disability, Trichotillomania, Trichophagia

Introduction

Trichotillomania is the compulsive urge to pull out one's own hair leading to noticeable hair loss, distress, and social or functional impairment. In Diagnostic and Statistical Manual of Mental Disorders– 5^{th} Edition [1], it is classified under Obsessive-Compulsive and Related Disorders and is often chronic and difficult to treat. [2] The commonest age of onset is between 9 to 13 years of age and at times triggered by depression and stress [3, 4]. It is usually confined to one or two sites but can involve multiple sites on the body [5,6]. The scalp is the most common site, followed by the eyebrows, eyelashes, face, arms, and legs [7] Some less common areas include the pubic area, underarms, beard, and chest [8]. People who suffer from trichotillomania often pull only one hair at a time, and these hairpulling episodes can last for hours at a time. They can go into remission - like states where the individual may not experience the urge to "pull" for days, weeks, months, and even years. Some of these patients may eat the pulled hair (trichophagia) causing intestinal obstruction due to the hair ball (trichobezoar). In the following case report, we focus in the management of a patient with Moderate Mental Retardation with Trichotillomania with Trichophagia. Trichotillomania has been associated with Trichobezoar, [9] pica, obsessive-compulsive disorder, [10] Tourette syndrome [11].

Case Report

Ms. S is a 19-year-old Indian female with no family history of mental illness. As a child, she displayed delayed milestones and thus an IQ assessment was done (Full-Scale IQ Scale IO - 49, Verbal IO - 48, Performance IO - 53) prior to her school admissions at the age of five. She was found to have Intellectual Disability, Moderate [12] and thus was sent to a special school. An IQ test was repeated when she was 10 years old, and the results were similar to that mentioned above. Her present visit with the psychiatrist occurred when her family noticed that she was displaying aggressive behavior at home for the past four months. She was attending a vocational school for young people with special needs and was coping fairly well. There were no complaints of aggression from the school. She would hit her family members without any provocation. When asked why she did so, she would shout at them or leave the room and made no attempt to explain herself. Two months after the onset of abnormal behaviour, her mother noticed that she had developed a bald patch on the right occipito-temporal area. When queried, the patient denied any knowledge about it. She several centres specializing visited in treatment for baldness, but the patch continued to increase. When she was seen in the clinic, during the first assessment, the patch was round in shape, about 3 centimeters in diameter. There were no other bald patches, and her eyebrows and eyelashes appeared normal. There was no deterioration in her activities of daily living. After the mother noticed the bald patch and after visits to the hair treatment centres, she kept a closer look on her and did notice occasional hair pulling. Teachers and other family members did not notice any hair pulling, though the father and siblings were away from the house at work most of the time.

Physical examination was unremarkable apart from the bald patch and Full blood count and basic metabolic chemistry panels were normal, which were done to rule out anemia and thyroid dysfunctions, which may lead to hair loss. Chest x-ray was done to rule out any systemic involvement like the lungs and heart as the differential diagnosis of Sarcoidosis, though unlikely, was thought of. X-ray was unremarkable. Mental state examination revealed a young lady with a bald patch on the right occipito temporal area. She displayed a tendency to be over familiar and asked certain questions repeatedly despite the questions being answered. She could answer simple questions usually with short relevant answers. Her mood was euthymic, and she did not verbalize or display any self-harm or aggressive behavior at the time of assessment. There were no psychotic symptoms. An IQ test was repeated (Full-Scale IQ - 51, Verbal IQ - 49, Performance IQ - 53) which was similar to the previous one done when she was ten years old. As her mother was concerned about the patient's aggression, she was prescribed oral Haloperidol 0.5mg twice a day. Oral Lorazepam 0.5mg twice a day was also added, and mother was instructed to give it to her only when the patient was agitated. She came for follow up every three weeks, and her aggression showed a reduction over the next two months. However, the bald patch kept increasing in size and during the 3rd visit (six weeks after the haloperidol and Lorazepam was started) it was noticed that she had developed another bald patch 3cm x 2cm on the parieto-frontal area, and the original patch in the right occipito-temporal area had increased in size to 4.5cm x 5 cm. Other physical examinations were unremarkable. There was no gastric tenderness or lump. She was less agitated, and lorazepam was completely stopped and haloperidol was reduced to 0.5mg in the afternoon for three weeks and then stopped. An X-ray abdomen was done to look for hair balls (trichobezoar). None were detected. She was reassessed after 4 weeks. The aggression was absent; she was euthymic and continued to do her activities of daily living as usual. The size of the bald patches though, increased. The patient finally admitted to pulling out her hair and hiding most of it under the cushions at her residence. She would throw the collected hair once, in three to four days. She mentioned that occasionally she would also swallow the pulled out hair. Thus, she was diagnosed with Trichotillomania with mild trichophagia.

She was referred for Cognitive Behaviour Therapy and Clomipramine 25mg at night was started, which was increased to 50mg after 2 weeks. Her family members and school teachers were requested to monitor her hairpulling behavior but not interfere in it. When she was seen after 4 weeks, she appeared sedated and there was a very minimal decrease in the symptoms. It was thought CBT strategies like helping the patient develop an increased awareness of the times of day, emotional states, and other factors that promote hair pulling would help to control the behavior. However, she stopped CBT after one session and was unwilling to continue. She was convinced to see the therapist once more, but she walked out of the room after 5 minutes and refused to return. In view of the sedation with Clomipramine, the medication was changed to Citalopram and it was started at 20mg but the patient complained of headache and dryness of mouth. She was also noticed to be pacing around the house and having poor sleep at night. The possibility of these new symptoms occurring as a result of side effects of Citalopram was considered, and thus she was started on Imipramine 10 mg at night for two weeks, which was then increased to 25mg at night for another 4 weeks. When reviewed after 4 weeks, the patient was alert and mother reported a decrease in the observable pulling out of her hair. Imipramine was kept at the same dose, and she was seen again after a period of 4 weeks. There was new growth of hair over the bald patches, and medication was continued at the same dose. She was seen regularly for a period of three years, and imipramine dose was gradually reduced and stopped after 6 months. Thus, she received imipramine for 40 weeks. However, 2 months after completely stopping imipramine, there was relapse of her hair-pulling symptoms and thus imipramine was restarted at 25mg and continued for 12 months at the same dose. Thereafter, it was reduced gradually over a period of three months and stopped. She has been on follow up for past three years without medications and there was no relapse of her hair-pulling symptoms or aggression.

Discussion

According to literature, the peak age of onset of Trichotillomania is 12–13 years, and the disorder are often chronic and difficult to treat [13]. Although rising tension and subsequent pleasure, gratification, or relief is part of the current diagnostic criteria for trichotillomania, many people with hair pulling to do not experience these symptoms. [14,15]. The above-mentioned patient was not a typical presentation as she was older. She was almost 19 years when the symptoms started. There were no symptoms of hair pulling as a child. Furthermore, she did not endorse any of the above emotions but her increased aggression without any apparent precipitant could have been an expression of the rising tension culminating in hair pulling. Her inability to verbalize her feelings even when specifically queried could be a possible reason for the increased aggression. We also queried childhood trauma, obsessive-compulsive symptoms in the past as hair pulling is often associated with post-traumatic stress disorder [16] and obsessive-compulsive disorder [17]. After she was diagnosed with trichotillomania, the treatment of choice was clomipramine, as this compound had proven to have the highest efficacy on a number of trials [18, 19]. This was later substituted with an SSRI which was also efficacious according to literature [20, 21] in conjunction with CBT, which emphasized on habit reversal [22, 23]. Unfortunately, she rejected CBT and refused to see a therapist after the first session. It was a challenge to convince her to do so again, though she did see the therapist once more, but it was impossible to establish a therapeutic alliance with her, partly because of her intellectual disability and also because she walked out of the room after 5 minutes into the session and refused to return.

Clomipramine that was started and continued for 4 weeks caused minimal reduction of hair pulling but made her drowsy. She was found to be dozing during her vocational training class and thus medication needed to be changed. This was followed by a trial of citalopram at 20mg/day as it may be preferable given the superior safety and tolerability of this drug class for related conditions, such as OCD, and the positive results reported for an open-label study with trichotillomania patients [24]. Due to side effects, this had to be changed. Though clomipramine was found to be more efficacious in trichotillomania than desipramine (an imipramine metabolite) in a randomized clinical trial, [19] the patient was started on imipramine because the other two medications, clomipramine and citalopram that were tried were not efficacious and caused side effects. With 50mg of clomipramine, she had shown minimal improvement, and we thought that if there was a possibility of

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optimizing the dose. she may have amelioration of her symptoms. However, since this was impossible due to her and her family's insistence on discontinuing it due to the side effects (drowsiness) experienced, another medication with similar structure and efficacy was selected. Imipramine was started with a low dose of 10mg and titrated up to 25mg. She responded well to the medication without significant side effects. Some trials have suggested the efficacy of olanzapine [25] or venlafaxine [26] in conjunction with Behaviour therapy. In this patient, because of her intellectual disability, it was difficult to engage her in therapy. However, despite most trials supporting clomipramine as superior to imipramine and desipramine, it is important to customize treatment according to individual suitability, and choosing a medication that the patient is comfortable with, is important. In this patient, presenting her with reasonable arguments and showing her evidence of superior efficacy of clomipramine was not helpful as she was unable to process the information and was insistent that we change to another medication. She did respond to Imipramine and cooperated with the treatment with subsequent remission of symptoms.

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