CASE REPORT

Trichotillomania with Trichophagia Complicated with Intestinal Obstruction Secondary to Trichobezoars Treated with Antidepressant: A Case Report of Rapunzel Syndrome

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Abstract

Trichotillomania with trichophagia is a bizarre psychiatric condition which is rarely reported. Trichobezoar on the other hand has been seen in various surgical reports. It is uncommon to encounter these conditions presenting concurrently. Trichotillomania which is a focus of this case report is a chronic, mental disease of impulse control, characterized by repetitive, compulsive, and self-induced hair pulling. It can occur at any age but is observed more often in adolescents, with a strong predominance in females. Diagnosis of trichotillomania may be difficult, and its effective treatment challenging. This is a case report regarding a rare case of all three conditions occurring simultaneously; of a young girl of native origin who had the recurrent urge to pull her hair and consume it. She developed two episodes of intestinal obstructions consequent to the trichobezoars which was secondary to the uncontrolled hair consumption. She had to undergo two separate laparotomies to relieve the obstruction.

Keywords: Trichotillonania, Trichophagia, Trichobezoar

Introduction

Trichophagia is classified under the Obsessive Compulsive Disorder spectrum of disorder as defined by DSM-V [1]. Trichotillomania or TTM is almost always accompanied with trichophagia and is and accounts for majority of cases [2]. This condition is classified under NORD (National Organisation of Rare Diseases) [3]. Most of these patients report the obsession which appears as a thought or urge which is difficult to resist or suppress. As a result, they act out on the compulsion by removing the hair by various means. It can be seen in depression, obsessive compulsive disorder, body dysmorphic disorder, eating disorder, anxiety disorder, alcohol and substance abuse and particularly trichotillomania. Trichophagia can lead to trichobezoar (hair ball) in worst case scenario and cause intestinal obstruction which requires surgical removal as hair is indigestible in human gastrointestinal tract. Trichobezoar can extend into small or large intestine and this condition is known as Rapunzel syndrome. It is named after a girl Rapunzel in a fairy tale [4]. This occurs

predominantly in young women with psychiatric disorders as compared to the regular obsession which affects both sexes equally.

Case Report

Miss R is a 16 years old Orang Asli girl From Gerik. She is single and dropped off from school since 15 years old and has been living with her family. She was presented to psychiatry during January 2020 on the fourth day after she underwent exploratory laparotomy for removal of bezoar from small bowel in Hospital Taiping. This was her second laparotomy during her encounter with us. Mental state revealed a young moderately built Orang Asli girl, who spoke mainly in Tagalog; their native language and minimal Bahasa Melayu. There was no tics or abnormal movements noted. Fortunately, her father was present for translation purpose. Both father and child were cooperative during the examination. She was calm and was not in any form of distress. Physical inspection revealed multiple patches of hair missing over her head, especially the parietal, temporal and occipital regions with certain regions showing signs of hair growth. She did not pull her hair from other body region eg groin or axilla. Physical examination was grossly normal. There were no abnormal facial or other forms of physical deformities. She did not show any signs of malnourishment or abuse.

According to her, this habit started when she was 12 years old. She had the habit of pulling out her hair and collected them into hair ball before swallowing them. Hair was removed by fingers and was non-instrumental in nature. She did it three to four times per day on a daily basis when no one was around. This incident went undetected at home for years as she did it secretively in the toilet. It worsened after bath or when she was combing her hair. She also admitted that the behaviour worsened at times of stress. She described the unmistakable sense of urge and apprehension to pull the hair and admitted that it was an unhelpful behaviour. It appears as a thought and a persistent urge to carry out the act. The obsession and the discomfort which came with it subsided after the act of pulling and swallowing the hair. However, it would quickly return a few hours later and the cycle of pulling and swallowing would repeat.

There appears to be no form of psychosis or culture bound practice which could influence her behaviour. Besides eating her hair, she did not take other non-nutritive objects. She did not appear to be malnourished in any way and her BMI was within normal range. Apart from her peculiar behaviour, she consumed regular diet comprising meat, rice, dairy products and vegetables. According to her father, patient was also a slow learner and could not study beyond form three in her secondary school. However, her family was unable to furnish us with detailed developmental history. She was diagnosed with Trichotilomania with trichophagia and underlying mild intellectual disability. Miss R was treated with T.Fluoxetine 20mg OD and her condition improved. Her father reported that her condition improved during subsequent visits and we noted signs of recovery and regeneration at previous hair patches. She admitted that she could not reduce the obsession and compulsion entirely, but the frequency had reduced to once a week which was a significant improvement. Now, she would extract her hair and play with it between her fingers before discarding it.

Discussion

The psychology underlying trichotillomania remains intriguing and bizarre till this day. Two TTM subtypes have been identified and is characterised based on pulling style: automatic and focused hair pulling [5]. Automatic hair pulling is characterized by the act of pulling without full behavioural awareness and is generally performed during sedentary or leisurely situations (e.g., reading or watching television). Focused hair pulling is thought to be a driven effort with full behavioural awareness and triggered by intense emotions or unpleasant internal experiences. Most patients report a mixed pulling style both automatic and focused [5, 6]. After pulling, individuals often engage in rituals, such as examining and toying with the hair/hair bulb, , biting off the root, mincing or eating the hair or hair bulb [6]. The act may worsen during times of stress, one pattern which is also consistent with the case of Miss R [2].

Biological studies have suggested that structural or functional abnormalities of the brain include subtle changes (compared to 'control' groups) in the putamen, cerebellum and cortical regions such as the anterior cingulate and right inferior frontal gyri [7, 8]. These brain regions are important in how prone we are to develop habitual behaviours, and how able we are to suppress inappropriate or unwanted habits once they occur. The specific structural or functional brain abnormalities associated with trichotillomania and the role that they play in the development of trichotillomania require more research to understand, because findings differ between studies [7]. Physical consequences from hair pulling include hair loss, scalp irritation, carpal tunnel syndrome as well as dental and gastrointestinal problems

Miss R has been treated with T.Escitalopram which is an antidepressant titrated to 20mg per day. Her response appears to be favourable. Our next approach is to ensure adherence to therapy with the aid of her father [9]. Whilst many literature encourage intensive psychological intervention for example dialectic behavioural therapy, such intervention was challenging in view of the language barrier encountered[9]. The fact that her village was deep in the jungle also proved as a therapeutic challenge as she had tendency to default follow-ups on certain occasions. Most of our psychological intervention were supportive in nature and were directed at improving patient's insight and understanding of the illness.

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