

# From trichotillomania to Rapunzel syndrome – a case report

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## Introduction

Trichotillomania is a disorder of impulse control, resulting in an urge and intense desire to pull out one's hair. Rarely some with this condition chew or swallow their hair leading to a sense of relief or pleasure. This condition is then known as trichophagia (1).

In trichophagia, rarely a mass of hair may build up in the gastrointestinal tract. This mass is known as a trichobezoar. It may cause bowel obstruction requiring urgent intervention. Rapunzel syndrome is a rare presentation of trichobezoar, in which the trichobezoar extends from the stomach to the small intestine and beyond. In these patients a proper psychiatric evaluation, treatment and follow up aids better post-operative outcomes.

In this case, we describe a 10 year old girl with Rapunzel syndrome. This case report highlights the course and progression of untreated trichotillomania to Rapunzel syndrome and possible contributory factors.

## Case Report

This 10-year old girl had been investigated for on and off abdominal pain for around one year, and on the most recent presentation had features of intestinal obstruction. She underwent emergency laparotomy, during which a trichobezoar was removed. The trichobezoar was seen to be extending from the stomach to the small intestine.

History revealed that her preschool teacher had first noticed her ingesting hair around the age of 4 years. Although this was noticed again at 7 years, she herself only recalled pulling out and ingesting hair at the age of 8 years, but not previously or after that age. Others too had not noticed such behavior after 8 years of age.

She reported that, in the past, the urge to pull her hair usually happened when she was on her own. She described pulling her hair out with the intention of ingesting it, and thereafter swallowing it deliberately. She reported obtaining pleasure from doing so. There was no history of pulling hair from other people, pets or dolls. However, she reported a desire to ingest hair fallen on the floor. There was no history of avoidance of social situations to engage in this behavior.

She also reported that she had engaged in this behavior when she was relaxed and not when she was distressed. There were no features of any other obsessive compulsive disorder, tic disorder, depression or anxiety. These behaviours appear to have stopped when the child was about 8 years of age. As such, the child had presented with intestinal obstruction due to a trichobezoar 2 years after the cessation of trichophagia.

She had an average school performance, and she and her family did not describe any major relational issues or difficulties. However, her mother lived abroad and visited them only once every 3 months. She appeared closer to her father and lived with him, her grandmother and younger sister. No abnormality in sleep or appetite was reported. There was no significant history of psychiatric disorders in the family.

Her mental state was stable at the time of examination. On physical examination there was no alopecia, friar tuck sign or hairs of varying length, suggestive of recent hair pulling. No other skin abnormalities or inflammation was noted.

As there was no reported trichotillomania or trichophagia at the time we saw her, the management was mainly focused on support and prevention. Psycho education, maintaining a thought diary, and discussing strategies (including habit reversal) if the urges were to reoccur were discussed.

## Discussion

The literature suggests that the prevalence rate of trichotillomania in children is around 0.05% (2). Trichophagia is even rarer, and trichobezoar does not occur in all children with trichophagia (2). Hence, the occurrence of Rapunzel syndrome is quite rare.

Early onset of hair pulling in those less than 6 years of age is usually benign and self-limiting (3). The natural history of trichotillomania is that in most children with onset before the age of 8, they outgrow the disorder by adolescence (3). In this girl, there was some evidence of trichotillomania and trichophagia since the age of 4 years onwards, which had progressed till 8 years and resulted in a trichobezoar at the age of 10 years. In clinical practice trichotillomania is commonly seen around 13 years of age, and is more commonly seen in females (4,5). As



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such the case described was suggestive of an early onset of trichotillomania.

Only a few studies have attempted to assess the relationship between trichotillomania and trichophagia. In one such study, out of 68 trichotillomania patients, 14 (20.6%) reported current trichophagia, and an additional 9 (13.2%) had ingested their hair episodically in the past (6). But the differences between individuals with trichotillomania who ingest hair, and those who don't, are not well described. The literature is not clear on the age of onset of trichophagia, although case reports contain cases from 5 years up to around 30 years of age. Among the case reports of children with early onset trichobezoar or Rapunzel syndrome, a considerable number is linked with mental retardation, although it was not the case here (7, 8).

Even though the literature does not clearly mention whether Rapunzel syndrome occurs while trichophagia is ongoing or not, in this case it appeared to have occurred two years after trichophagia had ceased.

Trichotillomania is a clinical entity with a high rate of co-morbidity with mood or anxiety disorders, most commonly depression (28.6%) and obsessive compulsive disorder (10.7%) (9). In our case, no clear contributory factors could be identified, except that the child's mother had lived abroad since the child was very young. Relationship issues are known to be associated with trichotillomania.

There are also no major studies to suggest the occurrence of significant psychological disturbances in the immediate post-operative period or intermediate period after the surgery for trichobezoar. In our patient as well, no major psychological or psychiatric issues were encountered in this period.

The literature on recurrent trichobezoar is very scarce although there are a few cases reported (10). Thus, the child described in this case report was offered reviews at least every three months.

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## Conflicts of interest

None reported.

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