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CASE REPORT

Trichotillomania with Trichophagia and Trichobezoars: A Case Report

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ABSTRACT

Trichotillomania is a condition when an individual is unable to resist the urge to pull out the hair on their head or in other places. Trichophagia is the repetitive swallowing of hair most usually connected with compulsive hair pulling. Trichotillomania with trichophagia is a rare psychiatric disease. Trichobezoar, on the other hand, has been documented in several surgical studies. It is unusual to see these symptoms appear at the same time. It can affect anyone at any age, but it is more common in adolescents, with a strong female predominance. This is a case report of an unusual case in an 11-year-old girl with all three disorders occurring at the same time and presenting with intestinal obstruction. This case report aims to identify signs and symptoms of trichobezoar, mainly in adolescents, to discover similar incidents that may link to psychiatric disorders like trichotillomania and trichophagia in medical practices.

INTRODUCTION

When compared to the previous DSM – IV, where it was classified under Impulse – Control Disorders Not Classified Elsewhere, trichotillomania is a condition in which an individual is unable to resist the urge to pull out the hair on their head or other places (DSM-4 Diagnostic and Statistical Manual, 2000). It is covered under the section on Obsessive-Compulsive and Related Disorders.

The criteria of an increased sense of stress shortly before ripping out the hair or when attempting to resist the activity were deleted in DSM-5. It has been superseded by repeated attempts to reduce or eliminate hair pulling (Christenson et al., 1991). The disease primarily affects females and adolescents, and they are subject to some but not all of the DSM-V criteria (DSM-5 Diagnostic and Statistical Manual, 2013). It is worth noting that hair-pulling can be automatic, occurring when the patient is unaware or attentive or completed when the patient is engaged in the pulling action (Grzesiak et al., 2017). Our goal is for early detection and precise diagnosis so that prompt action can be taken.

CASE PRESENTATION

An 11-year-old indigenous girl presented at the primary care clinic with acute epigastric discomfort and abdominal distension for four days. She had a history of haematemesis once but no alterations in bowel habits. She did not have a history of fever, a urinary tract infection, or an upper respiratory illness. She did not have melaena or loose stool. On examination, the abdomen was distended, with an epigastric mass measuring 4 × 5 cm and a spleen measuring 2 cm, but no hepatomegaly. Iron deficiency anaemia was discovered in initial blood testing, with haemoglobin of 8 g/dL and iron levels of 2.0 mmol/L. The blood result showed no evidence of infection with a white blood count of 10×10^3 per microlitre, neutrophils count of 50% and lymphocyte count of 40%. She was referred to the hospital for further management and plain abdominal imaging revealed the stomach and hepatic flexure appeared prominent, but there was no evident bowel dilation. Pneumoperitoneum was absent. As a result, an ultrasound abdomen was performed, which revealed an abundance of gas in the epigastric region, possibly due to gas in the pylorus or transverse colon. The CT abdomen and pelvis with oral contrast were then used to confirm the diagnosis. The CT abdomen and pelvis revealed a stomach distended with the presence of a mottled gas-patterned intragastric mass suspicious of a bezoar and a linear calcification was noted within. There was circumferential intramural wall thickening of the wall seen at the junction between the pylorus and duodenal cap suspicious of stenosis (Figure 1). The duodenum was also distended with a similar lesion which appeared to extend into the proximal jejunum (Figure 2). The CT scan concluded that the likely presence of gastric and proximal small bowel bezoar with features of gastric outlet obstruction is associated with long-standing chronic inflammation.

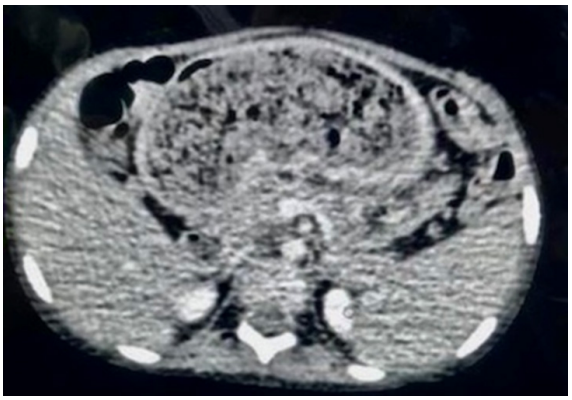

Figure 1 Axial view of abdominal computed tomography showed a hairball in the stomach

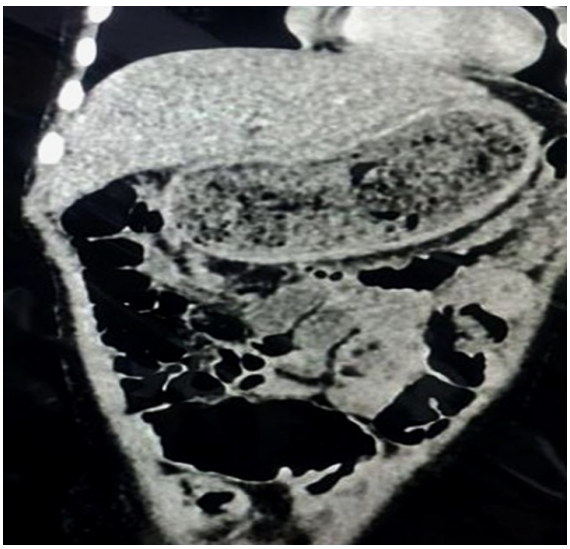

Figure 2 Coronal view of abdominal computed tomography showed the extension of a hairball from the stomach to the pylorus and duodenum

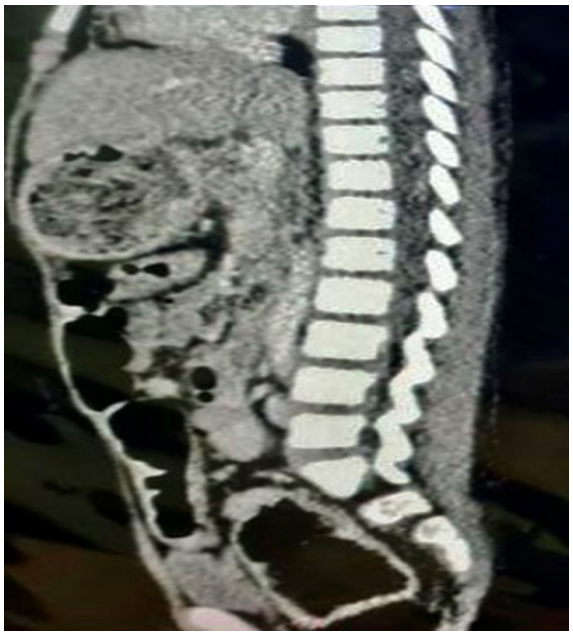

Figure 3 Sagittal view of abdominal computed topography

Pre-operatively, the patient's behaviours that may cause bezoars were thoroughly investigated. The mother noticed hair-pulling and hair-eating tendencies three years ago and tried to shave her hair to stop the behaviour but failed. The patient did not consume non-food items except for the hair. Then, an exploratory laparotomy was performed on the patient. A midline incision was made at the supraumbilical area, a gastrostomy and an enterostomy were performed, and a trichobezoar was delivered gently from the gastrotomy incision. A ball of hair from the stomach to the pylorus and duodenum (Rapunzel Syndrome) measuring 8 cm × 5 cm with an approximately 5 cm tail. Eight worms measuring 10 – 18 cm were evacuated through an enterostomy that was performed 30 cm from the duodenojejunal junction. The duodenum appeared normal, and stomach dissection revealed a 0.5 cm × 1.0 cm benign-looking ulcer on the body of the stomach with no biopsy performed. The incisions were repaired, and the rectus muscle and skin were closed with sutures. The parasitology findings of the worms resulted as *Necator americanus* is a species of hookworm infection.

During her psychiatric evaluation, she maintained a distance from the healthcare professionals while remaining close to her parents. Her assessment involved full background history, home environment, and mental state examination. On the social skills assessment, she did not interact with the doctor and appeared anxious when separated from her parents. She had poor eye contact and mostly talked to her mother. On further history, she has refused to attend school since she was eight years old for an unknown cause, and her mother has noted that she has acquired hair-pulling and chewing behaviour since then. Apart from the hair tugging and eating behaviours, the mother observed no other abnormality. A Goodenough Draw-a-Man test was performed on her with a score of 5.25. It indicated her mental age despite her actual age of 11. Her scalp was shaved with many coin-sized bald spots visible upon examination of her scalp. After the evaluation, she was diagnosed with trichotillomania, trichophagia, and borderline intellectual disability. She was started on a psychotherapy regime involving a psychiatrist, clinical psychologist, and occupational therapist, which included cognitive and behavioural therapy, reversal habit, and skill stimulation therapy. Her parents showed full cooperative support and affection throughout the process.

She and her parents attended all scheduled follow-ups throughout her follow-up and monitoring. She gradually reduced hair pulling, with no medicine prescribed for her. She was also able to restrain herself from eating the hair after pulling them with the help of close supervision from her parents. Her scalp examination showed old bald spots covered with minimal fine hair; no new bald spots were seen.

DISCUSSION

Trichotillomania commonly develops between 12 and 13 years old and is difficult to manage (Klobucar et al., 2018). The patient's atypical

symptoms delayed recognising the correct diagnosis and addressing the fundamental cause. Due to intellectual disability, acquiring an accurate history of the patient's disease is challenging. Her intellectual impairment, which hinders her from verbalising her feelings, may contribute to her.

Trichophagy has been found in more than half of trichotillomania cases (Malhotra et., 2008). In extreme trichotillomania cases, trichophagy and trichobezoar should be ruled out. In this case, a thorough history and examination were required to rule out trichophagy, and a USG abdomen revealed no trichobezoar. However, a CT scan of the abdomen was performed due to a mass over the stomach. It was revealing trichobezoar, allowing for the appropriate therapy for this patient. Rapunzel Syndrome, a rare form of trichobezoar extending from the stomach to the pylorus and duodenum, manifested late with gastrointestinal obstruction due to a low index of suspicion by healthcare personnel.

This girl's laparotomy revealed trichobezoar and eight worms ranging from 10 to 18 centimetres. Blood loss at the worm's intestinal attachment site caused this patient's iron deficiency anaemia. Iron and protein deficiency impairs growth and inhibits cognitive function which caused learning difficulties in this patient (Bagaskoro et al., 2017).

Worms and trichobezoar may cause intestinal obstruction. Pica is one cause of worm infestation. Pica causes people to consume non-food items like wormy dirt, causing worm infestations and iron deficiency anaemia (Al-Sharbati et al.,2003). However, this patient exhibited no signs of pica. Thus, her worm infection was likely attributable to low socioeconomic levels and water supplies. Worm infestation is still prevalent among indigenous Malaysians (Lim et al., 2019).

Trichotillomania is associated with intellectual impairment, depression, anxiety disorders, learning difficulties, enuresis, trichophagia, trichobezoar, and scalp abscess (Rehm et al., 2015). She also had comorbidities like learning disabilities and intellectual disabilities; however, she was not under any follow-up (Rothbart et al., 2013).

Habit reversal treatment (HRT) may comprise acceptance and commitment therapy and dialectical behaviour therapy for trichotillomania and trichophagia (Stanley et al., 1997). There are no first-line pharmacotherapies for trichotillomania. A recent Cochrane analysis found no evidence of a therapeutic effect for selective serotonin reuptake inhibitors (SSRIs) (Van et al., 2010). For this patient, her condition improved after behavioural treatment without pharmacological treatment.

In the treatment of trichotillomania, parents and primary care providers are critical. Early detection of intellectual disability should be improved at the primary care level to prevent the disorder from developing. As a result, the treating physician should be actively involved from the start (Woods et al., 2010).

This case demonstrates an unusual presentation of abdominal discomfort induced by trichotillomania and trichophagia complicated with trichobezoars, which should be considered while treating an adolescent with abdominal pain. Early diagnosis, intervention, and management, which include collaboration between a family medicine specialist, a paediatrician, a surgeon, and a psychiatrist, not to forget the patient's carers, as well as ongoing follow-up of the underlying ailment, are critical for averting disorder complications.

CONCLUSION

Trichotillomania should be evaluated as a differential diagnosis in a patient with sudden onset abdominal pain and a history of learning deficit. Focus history and examination are essential to aid in patient diagnosis and avoid delays in correct diagnosis and appropriate therapy, particularly with indigenous patients with a learning disability.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest to publish this case report.

CONSENT

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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FIGURE LEGENDS: