

Case Report

Gastric trichobezoar and Rapunzel syndrome: a case report

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ABSTRACT

Giant gastric trichobezoars are unusual form of foreign body found in stomach of mostly young adolescent females which may lead to morbidity and high mortality 30%, if goes unnoticed. These females have history of trichophagia or trichotillomania. This report is of a 12- year old young adolescent female presented with epigastric pain and mass. An exploratory laparotomy with anterior gastrotomy was performed and a giant trichobezoar with a very large tail of 2.5 ft was removed, weighing 2.52 kg. She recovered well and was discharged on 7th post-operative day. Patient was advised for psychiatric follow up.

Keywords: Gastric trichobezoar, Rapunzel syndrome, Trichophagia, Trichotillomania, Anterior gastrotomy

INTRODUCTION

Gastric trichobezoar is a mass or collection of undigested and accumulated foreign material usually own hair into the stomach. The term "bezoar" is derived from Arabic badzehr from Persian panzehr, both meaning counter poison or antidote.^{1,2}

Trichobezoars are often associated with trichotillomania (hair pulling), and trichophagia (hair swallowing). Trichotillomania may be unconsciously or unintentionally done and is part of the DSM IV psychiatric classification of impulse control disorders.^{3,4} In up to 18% of patients with trichotillomania, trichophagia occurs; one third of patients with trichophagia develop trichobezoars.⁵ Trichobezoars most commonly occur in adolescent females.⁶ The site of hair pulling is most commonly from the scalp, but can occur from the eyelashes, eyebrows, and pubic area.⁷ The first description of a post mortem human bezoar was by Swain in 1854.⁸

The name "Rapunzel" syndrome comes from the Grimm Brothers' fairy tale of a 12-year-old princess who was

shut into a tower with neither stairs nor doors by an enchantress who climbed up the tower's walls with the help of Rapunzel's long tresses.⁹

The prevalence of bezoars in humans is low, if treatment is not administered, associated mortality rates may be as high as 30% primarily because of gastrointestinal bleeding, destruction, or perforation.¹

CASE REPORT

A 12 years old girl presented with mild epigastric pain in our OPD. On physical examination patient had a palpable, non-tender epigastric mass which was crescent shaped, size approximately 10×15 cm, hard in consistency and was not freely mobile.

Plain radiograph was not conclusive, ultra sonography of abdomen revealed an echogenic mass giving acoustic shadow in stomach and extending beyond pylorus. Patient later on gave history of hair eating (trichotillomania and trichophagia). A provisional diagnosis of gastric trichobezoar was made and

exploratory laparotomy with anterior gastrotomy was planned.



Figure 1: Gastric cast trichobezoar removed from stomach.



Figure 2: A gastric trichobezoar with very long tail.

Operating finding

Anterior gastrotomy revealed a giant gastric trichobezoar of size 12×17 cm and the tail was extending up to the

ileum approx. 2.5 ft from the pylorus, slow and gentle removal of tail was performed without breaking it. Weight was 2.52 kg.

DISCUSSION

Trichobezoar commonly occurs in adolescent females, often with underline psychiatric disorders. The presentation may vary and these patients may not give history due to social stigma. However, this is a rare condition and in numerous cases high mortality rate has been reported.

We assume that the frequent location of these casts in the stomach is due to the holdup by the pylorus and the churning action of the stomach which helps to entangle new hair into the already formed cast. The bezoar takes on a glistening shiny surface from the mucus that covers it. Decomposition and fermentation of fats give the bezoar, and the patient's breath, a putrid smell.¹⁰ The acidic contents of the stomach denature the hair protein and give the bezoar its black colour.^{11,12}

Rapunzel syndrome is a rare form of trichobezoar, and various criteria have been used in its description in the literature. Some define it as a gastric trichobezoar with a tail extending up to the ileocecal junction.^{13,14} Some describe it as a simple trichobezoar with a long tail, which may extend up to the jejunum or beyond; and some still define it as a bezoar of any size which can cause intestinal obstruction.¹³

The diagnosis of a gastric trichobezoar can be confirmed by radiography or endoscopy. Plain films of the abdomen may reveal amorphous, granular, calcified, or whirlpool-like configurations of solid and gaseous material within the stomach.¹⁵ In some instances, the bezoar is so compact that a layer of air envelopes it and, in light of the long term accumulation, calcification is often observed. Upper gastrointestinal studies with use of contrast medium confirm the presence of a bezoar and may outline a concomitant gastric ulcer. On ultra-sonography, the echogenic arc of air between the bezoar and the gastric wall is pathognomonic and may be enhanced if fluid is administered concomitantly during the examination. Computed tomography vividly demonstrates trichobezoars as free-floating filling defects within the stomach, especially in the presence of orally administered contrast medium.

The most common of these complications that have been reported over the years include gastric mucosal erosion, ulceration, and perforation of the stomach or the small intestine, gastric outlet obstruction, intussusception, obstructive jaundice, protein-losing enteropathy, pancreatitis, and death.¹⁶⁻²¹

Management options include endoscopic removal, laparoscopic removal, or via laparotomy. Gorter et al, in a retrospective review of 108 cases of trichobezoar,

evaluated the available management options; it was noted that whereas 5% of attempted endoscopic removals were successful, 75% of attempted laparoscopies were successful.²² However, laparotomy was 100% successful and thus favoured as their management of choice in our case.

After trichobezoar removal, prognosis is good if psychiatric therapy to control habitual trichophagia is successful. In patients who have undergone gastrectomy, however, the recurrence rate of phytobezoars is 13.5% despite preventive measures.²³ Pica is another condition that necessitates psychiatric evaluation to prevent recurrence. Although risk factors for pica (female gender, childhood, mental retardation, and African-American and Aboriginal race) are known, this behaviour is not usually identified until a bezoar has been diagnosed. Thus, a bezoar should be considered in the differential diagnosis in a child with symptoms of gastrointestinal obstruction and a painless upper abdominal mass.

CONCLUSION

Trichobezoar must be considered as a differential diagnosis in a young female with epigastric mass. A thorough history of pica, trichotillomania and trichophagia must be obtained. As in our case history confirmed the diagnosis and thus contrast computed topography and upper GI endoscopy was not performed. In giant trichobezoar, exploratory laparotomy with anterior gastrotomy gave better result as the tail may reach up to ileo-caecal junction.

These patients must undergo psychiatric evaluation and treatment for better outcome and prevention of recurrences after surgery.

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