



A Rare Case of Rapunzel Syndrome Causing Small Bowel Obstruction.

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ABSTRACT:

Rapunzel syndrome is a rare form of bezoar in which trichobezoar is seen mostly in sufferers with records of psychiatric disorders, trichotillomania (dependency on hair pulling) and trichophagia (morbid addiction to chewing hair), consequently developing gastric bezoars. The fundamental signs are vomiting and epigastric ache. In this case report, we describe this syndrome in a younger female.

Introduction

A bezoar is a mass, ingested material that gets trapped in the gastrointestinal tract. Bezoars can be made up of vegetable or fruit fibres (phytobezoars), milk curds (lactobezoars) or any indigestible material that is ingested. Trichobezoar is the most common type of bezoar, composed of human hair, and is typically found in the stomach.[1]

This syndrome is named after the woman with the long tresses in the fairy tale. The Rapunzel syndrome was first described in the literature by Vaughan et al. in 1968 [1] and was named for the length of the hair and the uniqueness of the situation, both of which are characteristic of the fairy tale and of the clinical cases described in the report.

There are numerous different styles of presentation of this syndrome; but, in popular, it entails the presence of a gastric trichobezoar with a protracted tail extending beyond the duodenum, as found in the case pronounced right here. The component chargeable for this syndrome is the compulsion of patients to drag out their hair and swallow it, approaches referred to as trichotillomania and trichophagia, problems that affect young ladies with or without recognised psychiatric issues. [1]

Case report

A young female with psychiatric symptoms presented with acute abdominal pain and vomiting in the emergency department. There were similar episodes in the patient's past medical history.

The patient was slightly pale, the only abnormality noted during the physical examination. She had a history of slightly retarded neural development. The patient's mother admitted that she had a habit of pulling out her hair and secretly swallowing it.

Ultrasound of the patient was done and revealed dilated stomach and small bowel loops showing echogenic masses with intense internal shadowing. CT abdomen of the same patient revealed an intragastric well-circumscribed homogeneous mass consisting of a 'mottled gas pattern' due to the presence of entrapped air and food debris. Similar masses were seen in the terminal ileum, causing small bowel obstruction. The distal large gut is collapsed. No contrast enhancement was seen within the visualised masses. Findings were suggestive of trichobezoars causing small bowel obstruction. The patient was taken for surgery to relieve a small bowel obstruction. On surgery, trichobezoars were found lodged in the antrum region of the stomach and the distal ileum. A rare case of Rapunzel syndrome causing small bowel obstruction was reported.



Fig 1. Ultrasound showing characteristic echogenic content with dense internal shadowing.

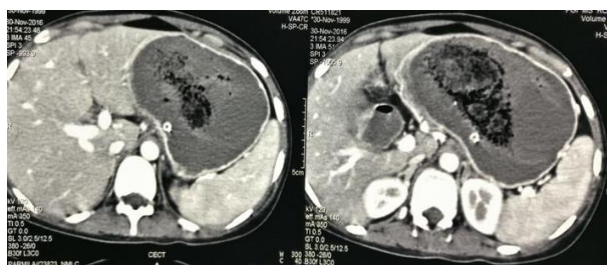


Fig 2. CT images showing a dilated stomach with trichobezoars within the lumen



Fig 3. CT scan of the same patient shows intragastric and ileal contents showing

Characteristic mottled appearance with small bowel obstruction.



Fig 4. Of the patient reveals dilated small bowel loops with internal hair filled lumen.

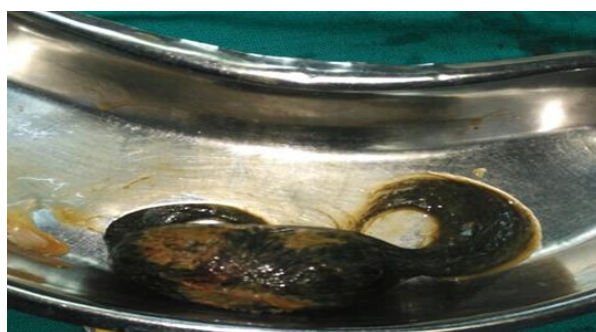


Fig 5. Reveals hair ball i.e. trichobezoar removed from bowel.

Discussion

Trichobezoar, a hairball within the proximal gastrointestinal tract, is an unusual condition primarily seen in younger girls. [1][2] Human hair is resistant to digestion and peristalsis. Therefore, it accumulates among the mucosal folds of the belly. Over time, continuous ingestion of hair leads to impaction by hair, along with mucus and food, resulting in the formation of a trichobezoar. In most instances, the trichobezoar is limited to the stomach. In a few cases, the trichobezoar extends via the pylorus into the jejunum, ileum or even colon. This condition is known as Rapunzel syndrome [3] [4]. Its part can break off and pass into the small intestine, causing intestinal obstruction [5] [6]. The frequent place of bezoars within the belly is due to the holdup by the stomach, which allows entangling new hair into the already formed solid. The bezoar takes on a glistening, vivid floor from the mucus that covers it. Decomposition and fermentation of fats deliver the bezoar and affect the person's breath. [6] The acidic contents of the belly denature the hair protein, supplying the bezoar with its characteristic black colour.[7]The causes of a bezoar can be due to the presence of indigestible fabric inside the lumen, gastric dysmotility (such as preceding surgery, which includes vagotomy and partial gastrectomy, and so on). Bezoars are found mainly in young ladies and are so named because of the substances which are swallowed and produce them, which include hair (trichobezoar), phytobezoar (vegetable fibres), diospyrobezoar (persimmon fibres), or pharmacobezoar (tablets/semi-liquid masses of drugs). [8]

Gastric bezoars must be differentiated from the pictures produced by retained food particles within the belly. The marked acoustic shadowing in the back of the echogenic band produced by a bezoar is different from "grimy" shadowing generated by ingested food and fuel in the stomach on ultrasonography. [9] On ultrasound, the featured appearance persists irrespective of the angulation of the transducer, the alteration of the patient's location, or the management of clean water. This excludes the pancreatic pseudocyst, a splenic or renal mass, a noncalcified gastric tumour, a gastric duplication cyst, or a gastric outlet obstruction. [10]On CT, a cellular intraluminal mass along with "compressed concentric jewellery" is seen. There is a combined density sample due to the presence of entrapped air and meal particles. Similar to the function as described above, the free mobility of the mass in the belly lumen may be visible in



supine and prone views. CT helps in the better configuration of the bezoar and in appropriately identifying its location. Furthermore, CT helps in better characterisation and differentiates it from other pathologies that would be difficult to identify on simple radiography or ultrasound. [11]

The treatment of huge bezoars and concretions is largely surgical. Gastric bezoars can be retrieved endoscopically or via gastrotomy, endorsed bezoar extraction by way of multiple enterotomies in cases of Rapunzel syndrome. [12] It is far obligatory to conduct a thorough examination of the rest of the small intestine and stomach to detect bezoars in the remaining gut and stomach. Other methods like the modified needle-knife (bezotome) and a modified lithotripter (bezotripter) have been successfully used. The affected person's psychological problems additionally need to be treated. [13]

Conclusion

It is crucial to make an early diagnosis of Rapunzel syndrome to avoid unnecessary investigations and confusion with other potential diagnoses.

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