

Rapunzel Syndrome: Gastroduodenal Tricobezoar. A Case Report

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Abstract

Rapunzel Syndrome is an unusual form of gastric trichobezoar with extension to the intestine, it is composed of different materials such as: hair, food waste, paper, starch, among others. We report the case of a 19-year-old woman, single, originally from the province of Chimborazo (Alausí) in Ecuador, who apparently presented for 2 months symptoms of nausea that reaches vomiting of food content on several occasions, early satiety, as well as intermittent, colicky abdominal pain. On palpation, a mass of hard consistency, regular painless and smooth edges, mobile approximately 30x15cm, occupying the epigastrium and mesogastric, was evidenced. Trichobezoar that was surgically removed due to its size was documented; and then your successful recovery.

Keywords: Bezoars; Trichotillomania; Gastrointestinal Diseases

Introduction

Foreign bodies in the digestive tract have been a concern since ancient times [1]. When foreign bodies remain in the digestive tract for a long time and resist the action of gastric juices, they form concretions called bezoars, a term derived from the Arabic words badzher, from Turkish panzehir, from Persian padzhar which means anti poison or antidote [2]. In the 12th century medicinal properties were attributed to them and they were used to treat epilepsy, dysentery, plague and leprosy [3]. Gastric bezoars are rare, cause nonspecific symptoms, and are usually found incidentally in patients who undergo gastrointestinal endoscopy or imaging tests [4]. A gastric bezoar is defined as a foreign body as a result of the accumulation of non-deferrable ingested material such as: hairs, date residues, papers, starch, resins, lacquers, among others [5].

Bezoars are classified according to their composition [6]:

- Phytobezoar: Composed of plant matter, it is the most common type of bezoar.
- Trichobezoar: composed of hair
- Farmacobezoar: composed of ingested drugs
- Others: Compounds of substances such as paper, silk, lacquer, fungi, cement, vinyl gloves.

- **Pathogeny:** Gastric bezoars are the result of the ingestion of indigestible material with an alteration of the stomach mechanism and the migratory interdigestive motor complex [7]. The underlying cause of bezoars was thought to be delayed gastric emptying, today studies have shown that it can occur in patients who have normal or accelerated gastric emptying, with the composition of ingested material being important in pathogenesis [8]. Trichobezoars develop in a patient with tricotillomania and trichophagia. It begins with retained hairs in the gastric folds; the hair is then denatured with gastric acid, turns black after being oxidized and combines with food to form a trapped mass, to later be colonized by bacteria resulting in halitosis.
- **Epidemiology and Risk Factors:** Gastric bezoars are rare with an incidence of 0.3% in upper endoscopy. Most trichobezoars are seen in women in their 20s and are often associated with psychiatric disorders [9]. There are risk factors that predispose them such as gastric dysmotility, gastric outlet obstruction, use of anticholinergic agents and opiates.
- **Clinical Manifestations and Complications:** Affected patients are asymptomatic for many years and the onset of symptoms is insidious. The most common symptoms include abdominal pain, nausea, vomiting, early satiety, anorexia, and weight loss. On physical examination, nothing relevant stands out except for an abdominal mass or halitosis; patients with trichobezoars may have irregular areas of alopecia [10]. Bezoars are rarely associated with gastrointestinal complications including gastrointestinal perforation, peritonitis, enteropathies, steatorrhea, pancreatitis, obstructive jaundice, appendicitis, constipation, and intestinal pneumatosis [11] [12].
- **Diagnosis:** Upper endoscopy is necessary to establish the diagnosis of a gastric bezoar and obtain samples to determine its composition.
- **Treatment:** The treatment must be adapted to the composition and the underlying pathophysiological process. Optimal management is controversial in the absence of studies comparing the different treatments.
- **Chemical dissolution:** Involves the administration of an agent to degrade the gastric bezoar, there are several agents used for chemical dissolution such as: Coca-Cola, cellulose, papain, acetylcysteine. However, there are no trials comparing these agents [13].
- **Endoscopic extraction:** Involves fragmenting the bezoar with a jet of water, direct aspiration through an endoscope channel, forceps, or hatches. The fragments can then be cleaned with the endoscope using a large-bore nasogastric tube, or allowed to pass through the gastrointestinal tract [13].
- **Surgery:** Is reserved for patients with large bezoars that fail to achieve chemical dissolution and endoscopic therapy and for patients with complications such as obstruction and significant bleeding [14].
- **Prevention of recurrences:** Up to 20 percent of patients with bezoars have a recurrence. To prevent recurrence, the patient should be encouraged to increase water intake, modify diet, chew better food, and psychiatric evaluation [15].

Presentation of the case

19-year-old female patient, originally from the province of Chimborazo (Alausí), single, indigenous race. Personal pathological history the patient has been diagnosed with iron deficiency anemia for 13 months, for which she receives iron treatment for two months. For two months she began symptoms of nausea that reached vomiting of nutritional content, on several occasions, early satiety, as well as intermittent and colic abdominal pain. The physical examination revealed blood pressure: 110/60 mm Hg, heart rate: 85 per minute, respiratory rate: 17 per minute, temperature: 36.5°C, nutritional status: poor, skin and mucous membranes: hydrated, with slight pale-

ness in addition to presenting halitosis. Abdomen: soft, depressible, not painful, a mass of hard consistency, regular painless and smooth edges, mobile of approximately 30x15cm that occupies the epigastrium and nasogastric, is evident on palpation, air-fluid noises present.

Complementary exams: Hb 8.0 Hcto 28.7, VCM 59, MCHC 26, PLATELETS 376,000. Leukocytes 4,500,000, N 55% L 35%, Creatinine 0.5, normal liver profile, TP 12, TTP 24 CEA <3.2 Ca 125 = 25, Ca 19.9 = 4.9, B 2 microglobulin 1.4, alpha fetus protein 2.8 Abdomen ultrasound. - The study shows a probable tumor mass, teratoma or peritoneal liposarcoma Pelvic Abdomen CT. - Conclusion: Gastric Bezoar; normal rest.

Upper endoscopy: Gastric trichobezoar with intestinal extension, polypoid lesions in the incisura, superficial ulcers in the gastric body.

Evolution: A laparotomy, gastrostomy and extraction was made. A 30x15 cm long trichobezoar were obtained without any complications, after 5 days postoperatively she was discharged in good condition.

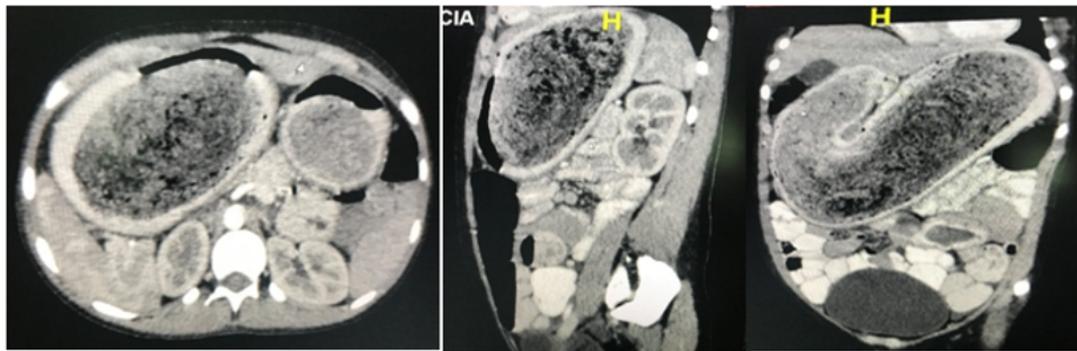


Figure 1: Data from the patient's medical history. General Surgery Service of "Hospital General Ambato.

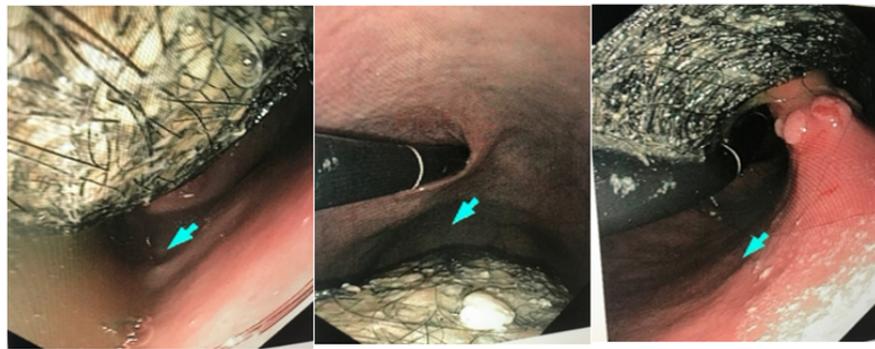


Figure 2: Data from the patient's medical history. General Surgery Service of "Hospital General Ambato.

Discussion

Several types of bezoar have been described in the literature, trichobezoars are accompanied by trocotillomania and trichophagia, the causes that lead to it are not known; there is talk of psychological, psychiatric, metabolic disorders, micronutrient deficiency, including educational level and unhealthiness and the status of poverty, in which the patient develops in his social environment.

It is also indicated that they are more frequent in women, but it may be due to their gender since they usually have long hair, which would facilitate the formation of voluminous trichobezoars in the gastric body.

Bezoar treatment depends on its size, and its location in the digestive tract, it also depends on professional experience with chemical solvents and many cases lead to unnecessary surgeries, even putting the patient's life at risk.

Conclusion

- There is a low prevalence or underreporting of this pathology, both in developed and developing countries, such as our country.
- The handling of this type of bezoar should be protocolized, which makes it easier for medical personnel to provide the best care to the patient.
- The patients that present this pathology, after being treated for their emergency; They must comply with comprehensive and multidisciplinary care, especially their psychological-psychiatric, nutritional assessment, complementing their family environment in which they may be immersed, poverty, dysfunctional homes that are triggering factors.
- Both the patient and her family environment should monitor the case to avoid recurrences of this pathology in the same patient.

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