



Giant Gastric Trichobezoar and Rapunzel Syndrome: Case Report in a Hospital in the South of Huila- Colombia



Felipe Andrés Beltrán Torres¹, Rafael Enrique Gómez Celedón¹, Mario German Orozco Sandoval^{2*}, Johann Fernando Orozco Castro², Laura Cristina Fantino Ibarra³, Stefany Mejía Buesaquillo³ and Xsara Camila Rodríguez Cerón³

¹Department of Anesthesiology and General Surgery, Hospital Departmental San Antonio de Pitalito, Colombia

²Anesthesiologist, San Antonio de Pitalito Hospital Huila, Colombia

³Internal Medicine Physician, San Antonio de Pitalito Hospital Huila, Colombia

Received: December 07, 2021; Published: December 13, 2021

*Corresponding author: Mario German Orozco Sandoval, Anesthesiologist, San Antonio de Pitalito Hospital Huila, Colombia

Keywords: Trichobezoar; Trichophagia; Gastric; Trichotillomania

Introduction

Bezoars correspond to conglomerates of non-digestible materials, in the case of hair trichobezoars that, when mixed with gastric secretion and food debris, compact forming masses of progressive growth either in the stomach, small bowel or even the colon. They occur mostly in young women and are closely related to trichotillomania and trichophagia. The clinical spectrum ranges from asymptomatic cases, vague and nonspecific symptoms, to complications that can be lethal. The fundamental therapeutic principle is based on the total removal of the foreign body, for which endoscopic and surgical techniques have been described, including minimally invasive ones. It should be taken into account that the mental health approach cannot be left aside, not only of the patient but also of the psychosocial environment.

In our environment, it is considered a rare entity, its incidence being unknown, with few publications limited to case reports and literature reviews. Therefore, and with the aim of improving the available information about this pathology, we present a case of successful extraction of a giant trichobezoar in a 19-year-old girl with a history of trichophagia and psychiatric disorders.

Description of the Clinical Case

This is a 19-year-old female patient, single, housemaker, with mild cognitive deficit and trichotillomania since the age of 4 years, with no other relevant medical, toxic-allergic or

surgical history. She was admitted to the institution for clinical symptoms of several months of evolution, consisting of abdominal pain and distension, associated with unquantified weight loss and occasional emetic episodes. When questioned about trichotillomania or trichophagia, the mother confirmed these behaviors since she was 4 years old. On physical examination, the patient is in poor muscular and nutritional condition, BMI 17 kg/m², the abdomen is slightly distended, painful on deep palpation in the left hypochondrium with a sensation of a hard, well-defined mass, positive peristalsis, without signs of peritoneal irritation. Paraclinical tests showed moderate anemia of normal volumes, mild leukocytosis with left shift and thrombocytosis, without coagulation disorder.

Taking into account the history and clinical findings, the presence of a trichobezoar was considered as the main diagnostic suspicion, so an esophagogastroduodenoscopy was requested, which reported a foreign body occupying the entire gastric cavity, consisting of mucus, food debris and hair, thus confirming the diagnosis. Due to the size of the trichobezoar it is not possible to perform endoscopic extraction, opting for surgical extraction by laparotomy. The patient is taken to the surgical procedure, supraumbilical midline incision with systematic exploration of the peritoneal cavity, identifying 100% gastric occupation by a mass of pasty consistency, oblique gastrotomy is performed, trichobezoar is identified, complete stripping is performed as a single piece,

measuring 30 by 10 cm, which molded the entire gastric cavity and had a tail extending into the small bowel of approximately 50 cm (Figure 1), in addition to ulcer in the gastric base, covered by fibrin. Gastrographic and abdominal wall closure were performed with conventional technique (Figure 2); immediate postoperative

multimodal analgesia given by nonsteroidal anti-inflammatory (Diclofenac 50mg IV), local anesthetic (Bupivacaine 50mg SC) administered by the surgeon at the operative site and opioid (Morphine 3mg IV).



Figure 1: Extracted trichobezoar.



Figure 2: Gastrorrhaphy after foreign body extraction.

In the immediate postoperative period, she was evaluated by the psychology service due to episodes of anxiety, who documented that the patient had an adequate family support network and socioeconomic support; however, a cognitive alteration was perceived, with sensory perception and attention within normality. During hospital stay, control paraclinics reported anemia grade III

WHO (Hb 7.4 GR/L), however without clinical signs of low output, so it was decided to reserve the patient and a hemogram control was indicated, which did not show acute anemia. The patient presented a satisfactory postoperative evolution, discharged on the sixth postoperative day, with multidisciplinary management by general surgery, psychology and psychiatry.

The pathology report indicates macroscopic findings of an elongated nodular formation formed by numerous black hairy structures, measuring 32x12x10 cm and weighing 1,428 grams, corresponding to TRICOBEOBAR.

Discussion

Bezoars are defined as conglomerates or concretions of poorly or non-digestible material within the gastrointestinal tract, resulting from accidental or intentional ingestion of these substances [1]. They are typically named according to their constituent material and have been categorized into five main groups: phytobezoar (vegetables, fruits), trichobezoar (hair), lactobezoar (dairy products), pharmacobezoar (drugs), and foreign body [2]; being the phytobezoar the most common [1-3]. For many centuries, bezoars have been found in the digestive tract of humans and animals, and although they are most commonly found in the stomach, they can lodge in any part of the gastrointestinal tract [3]. According to the literature the first case of trichobezoar was described by Baudomant in 1779 and the first surgical removal was performed by Schnbern [4] and since the beginning of its descriptions it has been associated as in the case of our patient to psychiatric disorders such as trichotillomania (hair pulling out) and trichophagia (hair ingestion). One third of patients with trichotillomania present trichophagia, and of the latter only 1% develop complications that justify surgical management [5].

Its incidence is not well known; up to 2019, 120 cases have been reported in the medical literature; according to the Colombian Journal of Gastroenterology, the disease is more prevalent in the female population and its peak age is between 5 and 23 years old [6,7]. In patients with trichotillomania, a 0.6 to 1.6% prevalence and a 14% recurrence rate have been described [8]. There is a complication of trichobezoars [9], in which a distal extension is formed, reaching the jejunum and even the ileocecal valve, causing an obstructive phenomenon, called Rapunzel syndrome [10], There has been controversy as to the true diagnostic criteria of this entity, so it has been accepted that the characteristics that would qualify these cases as Rapunzel syndrome would be: trichobezoar with tail; extension of the tail at least up to the jejunum; symptoms suggestive of obstruction [11]. The case presented here complies with the characteristics of Rapunzel Syndrome.

The clinical symptoms are variable, ranging from asymptomatic or with the classic triad that corresponds to the majority of cases and is presented in this report: weight loss, abdominal pain and palpation of abdominal mass; it can also be associated with early satiety and halitosis. There have also been reports of aberrant presentations such as acute abdomen, which may include gastrointestinal bleeding, gastric ulcer, pancreatitis, cholangitis, obstructive jaundice, appendicitis, intestinal intussusception, perforation or obstruction [12-14]. Diagnosis involves clinical suspicion, and documentation of the bezoar by

diagnostic imaging or digestive endoscopy. Contrast tomography is the radiological method of election, with a sensitivity of 90% and a specificity of 60%. In addition to identifying the cause, it establishes the presence of complications and allows surgical planning [15]; although CT is the gold standard for the diagnosis and choice of surgical strategy, the EVD report, already indicated clear signs of obstruction of the intestinal lumen, so the use of this resource was not considered.

Risk factors include dental deficiencies, mental retardation, previous gastric surgery and gastric motility disorders [9]. In the coexistence of personality disorders such as delusions, mood disorders or cognitive deficits [16], as in the case of our patient, trichobezoar becomes a preventable pathology through follow-up and therapy by services such as psychology and/or psychiatry, depending on the need. However, getting to this behavior can be difficult because patients usually deny data on trichotillomania or trichophagia and arrive at the emergency department with symptoms compatible with intestinal obstruction in most cases, so their diagnosis is usually delayed.

Treatment depends on the size, location and associated pathologies. Different therapeutic alternatives have been described, one of them is chemical solutions, which have not shown effectiveness in trichobezoars [17]. The endoscopic technique which consists of mechanical fragmentation using lithotripters, forceps for biopsy, polypectomy loops, argon, electrocoagulation devices, laser, even dormie baskets [18-20]. Despite all these options, trichobezoars are often not amenable to endoscopic management and require surgical management.

Laparoscopic surgery is an option in selected cases of trichobezoars with the additional benefits of the technique [21].

Laparoscopic extraction of giant trichobezoars should be undertaken with great caution due to the risk of contamination of the abdominal cavity and major complications [22,23]. Two cases of cooperative surgery using both endoscopic and laparoscopic approaches have also been described [24,25], which was not a therapeutic choice in our patient due to the size of the foreign body.

The open surgical technique continues to be the most widely used, consisting of performing the enterotomy with complete extraction of the bezoar, and additionally, according to each case, defining the need for intestinal resections, anastomosis, ostomies, etc. As in our center, laparotomy is still used as the laparotomy of choice, given the possibility of resolution of all cases, the relatively low rate of complications, the ability to completely evaluate the gastrointestinal tract, and the shorter surgical time.

Conclusion

Gastric trichobezoar is an uncommon pathology, which can become complicated causing from intestinal obstruction to gastric perforation. Early diagnosis and the appropriate treatment

strategy are essential to reduce complications, remembering that concomitant management with the psychology service and, if necessary, with psychiatry, should always be included among the pillars of treatment. Therefore, it is important to remember that the definitive treatment of trichobezoar is not only focused on its removal either endoscopically, laparoscopically or openly, it should be centered on a multidisciplinary management, with the objective of preventing recurrences.

References

- Paschos K A, Chatzigeorgiadis A (2019) Pathophysiological and clinical aspects of the diagnosis and treatment of bezoars. *Annals of Gastroenterology* 32(3): 224-232.
- Hall, Joshua D, Shami Vanessa M (2006) Rapunzel's Syndrome: Gastric Bezoars and Endoscopic Management. *Gastrointest Endoscopy Clin N Am* 16(1): 111-119.
- Iwamuro M, Okada H, Matsueda K, Inaba T, Kusumoto C, et al. (2015) Review of the diagnosis and management of gastrointestinal Bezoars. *World J Gastrointest Endosc* 7(4): 336-345.
- Delgado Duatis GY, Guillen G (2015) Trichobezoars: An unusual cause of intestinal obstruction. *Anales de pediatria (Barcelona, España: 2003)* 83(4): 289-290.
- Riveros Vega, Javier Humberto, Gómez Zuleta, Martín Alonso, Estarita, et al. (2020) Rapunzel Syndrome: Myth or Reality. *Revista colombiana de Gastroenterología* 35(2): 207-211.
- Pinilla R O, Vicente M L, González M, Vicente A A, Pinilla M E (2016) Tricobezoar gástrico, revisión de la bibliografía y reporte de un caso. *Revista Colombiana de Cirugía* 31(1): 44-49.
- Abril L, Olivera M, Garrido J (2016) Tricobezoar. Reporte de caso. *Revista Med Bogota Tomó* 24(2).
- Vasquez-Ciriaco S, de Oca-Duran, Edgar Montes (2011) Síndrome de Rapunzel: tricobezoar gastroduodenal Caso clínico y revisión de la literatura. *Evidencia Médica e Investigación en Salud* 4(3): 104-108.
- Corona Cruz J F, Sánchez Lozada R, Gracida Mancilla N I, Palomeque López A, Vega Chavaje G R (2005) Intestinal tricobezoar as a cause of small bowel consumption. *Gaceta medica de Mexico* 141(5): 417-419.
- Corona Cruz J F, Sánchez-Lozada R, Gracida Mancilla N I, Palomeque López A, Vega Chavaje G R (2005) Intestinal tricobezoar as a cause of small bowel consumption. *Gaceta medica de Mexico* 141(5): 417-419.
- Faria AP, Silva IZ, Santos A, Avilla SGA, Silveria AE (2000) The Rapunzel syndrome - a case report: trichobezoar as a cause of intestinal perforation. *J Pediatría (Rio J)* 76(1): p83-86.
- Pérez E, Ramón Sántana J, García G, Mesa J, Ramón Hernández J, et al. (2005) Perforación gástrica en adulto por tricobezoar (síndrome de Rapunzel). *Cirugía española* 78(4): 268-270.
- Dalshaug G B, Wainer S, Hollaar G L (1999) The Rapunzel syndrome (trichobezoar) causing atypical intussusception in a child: a case report. *Journal of Pediatric Surgery* 34(3): 479-480.
- Barrows A, Vachon T, Campin RC, Ignacio RC (2015) Trichobezoars detected and treated based on plain radiography. *Mil Med* 180(10).
- Perales Cheleon F (2016) Tricobezoar gástro duodenal recurrente. *Rev Cuerpo méd* 9(2): 124-128.
- Fernández López, A J, González Valverde FM, Rodenas-Moncada J y Albarracín Marín Blazquez A (2014) Trichophagia and Trichobezoar. *Cirugía Española (edición inglesa)* 92(6): 448-449.
- Wang PY, Wang X, Zhang L, Hai-Fei Li, Liang Chen, et al. (2015) Bezoar-induced small bowel obstruction: Clinical characteristics and diagnostic value of multi-slice spiral computed tomography. *World J Gastroenterol* 21(33): 9774-9784.
- Ugenti I, Travaglio E, Lagouvardou E, Caputi Iambrenghi O, Martines G (2017) Successful endoscopic treatment of gastric phytobezoar: a case report. *Int J Surg Case Rep* 37: 45-47.
- Mao Y, Qiu H, Liu Q, Zhongsheng Lu, Kaichun Fan, et al. (2014) Endoscopic lithotripsy for gastric bezoars by Nd: YAG laser-ignited mini-explosive technique. *Lasers Med Sci* 29(3): 1237-1240.
- Senturk O, Hulagu S, Celebi A, A E Duman, G Dindar, et al. (2014) A new technique for endoscopic treatment of gastric phytobezoars: fragmentation using guidewire. *Acta Gastroenterol Belg* 77(4): 389-392.
- Javed A, Agarwal AK (2013) A modified minimally invasive technique for the surgical management of large trichobezoars. *J Minim Access Surg* 9(1): 42-44.
- Sharma D, Srivastava M, Babu R, Anand R, Rohtagi A, et al. (2010) Laparoscopic treatment of gastric bezoar. *JSLS* 14(2): 263-267.
- Ulukent SC, Ozgun YM, Şahbaz NA (2016) A modified technique for the laparoscopic management of large gastric bezoars. *Saudi Med* 37(9): 1022-1024.
- Kurosu T, Tanabe S, Hasegawa R, Takafumi Yano, Takuya Wada, et al. (2018) A giant trichobezoar extracted by laparoscopic and endoscopic cooperative surgery (LECS). *Endosc Int Open* 6(12): E1413-E1416.
- Gorter R R, Kneepkens C M F, Mattens E C J L, Aronson D C, Heij H A (2010) Management of trichobezoar: Case report and literature review. *Pediatr Surg Int* 26(5): 457-463.



This work is licensed under Creative Commons Attribution 4.0 License
DOI: [10.19080/OAJS.2021.13.555869](https://doi.org/10.19080/OAJS.2021.13.555869)

**Your next submission with Juniper Publishers
will reach you the below assets**

- Quality Editorial service
- Swift Peer Review
- Reprints availability
- E-prints Service
- Manuscript Podcast for convenient understanding
- Global attainment for your research
- Manuscript accessibility in different formats
(Pdf, E-pub, Full Text, Audio)
- Unceasing customer service

Track the below URL for one-step submission
<https://juniperpublishers.com/online-submission.php>