

# Rapunzel syndrome with double trichobezoar. A case report

## Síndrome de Rapunzel con doble tricobezoar. Reporte de caso

Adrián Dersu Pliego-Ochoa,\* Eduardo Escalante-Ayuso,\*\* Edgar Rafael Mendez-Pérez\*

### Keywords:

Bezoars/pathology,  
bezoars/surgery,  
trichobezoar,  
intestinal obstruction/  
etiology, gastrotomy/  
methods,  
trichotillomania /  
complications.

### Palabras clave:

Bezoares/patología,  
bezoares/cirugía,  
tricobezoar,  
obstrucción  
intestinal/etiología,  
gastrotomía/métodos,  
tricotilomanía/  
complicaciones.

### ABSTRACT

**Introduction:** Rapunzel syndrome is a rare entity that consists of a gastric trichobezoar with extension to the duodenum. It is accompanied by symptoms of intestinal occlusion, and can produce complications such as perforation and peritonitis. **Case report:** A 15-year-old female with abdominal pain of two weeks, associated with nausea, emesis, early satiety, and decrease in the consistency of the stools. She referred trichotillomania and trichophagia habits of one year of evolution. On physical examination, a distended abdomen was found, with decreased peristalsis and no peritoneal irritation. Imaging studies showed a mass in the stomach and first portion of the duodenum, and air-fluid levels. Complete extraction was done by gastrotomy and enterotomy. Enteral feeding started at 72 hrs and the patient was discharged seven days after surgery. **Conclusion:** Bezoars can be resolved by non-surgical methods; however, surgical management is recommended with good results. Multidisciplinary management with this type of patients is necessary to prevent a recurrence.

### RESUMEN

**Introducción:** El síndrome de Rapunzel es una entidad no frecuente que consiste en un tricobezoar gástrico con extensión a duodeno acompañado de síntomas de oclusión intestinal, aumentando los riesgos de complicaciones como perforación y peritonitis. **Caso clínico:** Femenino de 15 años con cuadro clínico de dolor abdominal de dos semanas de evolución, asociado a náuseas, emesis, saciedad temprana y disminución en la consistencia de las evacuaciones. Refiere hábitos de tricotilomanía y tricofagia de un año de evolución. A la exploración física, abdomen distendido con peristalsis disminuida y sin datos de irritación peritoneal. En los estudios de imagen se observa una masa ocupativa en el estómago y primera porción del duodeno, así como niveles hidroaéreos. Se realiza manejo quirúrgico mediante laparotomía encontrando un tricobezoar gástrico, además de otro en yeyuno, efectuándose la extracción completa mediante gastrotomía y enterotomía. Se inició la dieta enteral a las 72 horas y es egresada del servicio al séptimo día del postoperatorio. **Conclusión:** Los bezoares se pueden resolver por métodos no quirúrgicos; sin embargo, se debe valorar el estado hemodinámico del paciente; se recomienda el manejo quirúrgico con buenos resultados. Es necesario un manejo multidisciplinario con este tipo de pacientes para prevenir su recurrencia.

\* Resident in General Surgery.

\*\* Attending Surgeon.

Servicio de Cirugía General, Hospital General "Dr. Agustín O'Horan", Mérida, Yucatán.

Received: 12/03/2018  
Accepted: 29/08/2018



### INTRODUCTION

A bezoar is a foreign body that forms in the gastric chamber after the ingestion of non-digestible substances such as paper, starch, resins, fibers, seeds, hair, and others. Likewise, they are classified according to their main composition, the trichobezoar (hair) being the most frequent, in 50% of the cases, followed by the phytobezoar (vegetable materials) in 40%.<sup>1</sup>

It is worth mentioning that trichobezoar appears in people with psychiatric and personality disorders mainly accompanied by trichotillomania and trichophagia. Intestinal occlusion as a mechanical complication of the bezoar is one of the causes of hospital admission in 4-10% of cases; it is also associated with complications such as ulcerations, perforation, and intussusception with a mortality of up to 30%.<sup>2</sup>

**How to cite:** Pliego-Ochoa AD, Escalante-Ayuso E, Mendez-Pérez ER. Rapunzel syndrome with double trichobezoar. A case report. Cir Gen. 2019; 41(3): 217-220.

Rapunzel's syndrome, a term coined in 1969 by Vaughan et al, is a rare and complex entity consisting of a gastric trichobezoar with extension to the duodenum and small intestine accompanied by symptoms of intestinal occlusion, and risk of complications such as perforation and peritonitis.<sup>3,4</sup>

Characteristic symptoms are abdominal pain in 70% of the cases, nausea and vomiting in 64%, weight loss in 38%, and alterations in consistency of the stools in 32% of the cases. Also, anorexia, dyspepsia, general malaise, weakness, and early satiety can be present. On physical examination areas of alopecia, and halitosis can be found, and a well-defined mobile abdominal mass can be palpated in the epigastrium in 88% of cases.<sup>5-9</sup>

### CLINICAL CASE

A 15-year-old female patient was admitted to the Emergency Department for two weeks of generalized colicky abdominal pain associated with nausea, multiple vomiting, early satiety, and decreased stool consistency. It should be noted that she referred trichotillomania and trichophagia habits of one year of evolution, as well as a depressive-anxiety disorder, medically treated.

The symptoms worsened, with an increase in the frequency of fecal vomitus and absence of stools. On physical examination, she was conscious, well oriented, afebrile, with normal vital signs. Her abdomen was distended, painful, and had a palpable plastron in the epigastrium with no signs of peritoneal irritation.

Simple abdominal radiographs showed signs of intestinal occlusion. A simple and



**Figure 1:** Gastric trichobezoar extraction.



**Figure 2:** Gastric trichobezoar.

contrasted CT scan showed a mass occupying the entire stomach and first portion of the duodenum, as well as air-fluid levels and dilation of intestinal loops.

Management was initiated placing a nasogastric tube for decompression, which rendered 200 ml of fecal liquid. Surgical management was done by gastrotomy (*Figure 1*). The trichobezoar occupied the entire gastric chamber and the first portion of the duodenum. After complete extraction (*Figure 2*) repair was done in two planes; with Connell's inverting suture for the internal plane and Lembert's sero-muscular suture for the external plane (*Figure 3*). Also, a transpyloric nasogastric tube was left. On exploration of the intestinal tract, a complete obstruction was found 40 cm distal from the angle of Treitz (*Figure 4*). An enterotomy was done, a second trichobezoar of approximately 5 × 3 cm (*Figure 5*) was extracted and the defect repaired in two planes with the Heineke-Mikulicz technique (*Figure 6*).

Postoperative evolution was favorable, with bowel movements present and no signs of infection.

### DISCUSSION

The diagnosis of this pathology is a challenge since only in 50% of the cases a history of trichophagia is obtained. A history of

psychiatric disorders, alopecia plaques, halitosis, and anemia with weight loss should lead to suspicion. Another important issue is the time required for a trichobezoar to form since ingested hair accumulates in the gastric folds and mixes with mucus, pepsin, bacteria, and hydrochloric acid with denaturation of proteins; a process that can take place over months or years.

Endoscopy is the study of choice for the diagnosis of a bezoar.<sup>10</sup> The contrasted CT scan has a sensitivity of 81% and a specificity



**Figure 3:** Gastric suture in two planes.



**Figure 4:** Location of the second trichobezoar.



**Figure 5:** Extraction of trichobezoar of jejunum.



**Figure 6:** Intestinal suture in two planes.

of 96%. Furthermore, it allows to differentiate the bezoar from a neoplasm, by showing its size, shape, and location.<sup>11</sup>

Gastric bezoars can be resolved by non-surgical methods (endoscopy, acetylcysteine, carbonated beverages, and cellulose, among others).<sup>12-14</sup> However, the size of the bezoar, the presence of associated complications and the hemodynamic status of the patient should be assessed. In intestinal bezoars, due to the high

percentage of failure of endoscopic treatments, surgical management is recommended with good results.

Multidisciplinary management is required to prevent a recurrence, which can be of up to 20% over a period of five to 20 years.<sup>15</sup>

## CONCLUSIONS

Multidisciplinary management and both clinical and psychiatric follow-up will always be necessary to prevent recurrence. Since our patient had a history of depression and anxiety, she received psychological support during her in-hospital stay and continued to be managed externally.

## REFERENCES

1. Parilli JC, Gómez TA, Rincon N, Berrios C. Trichobezoar: an unusual diagnosis. Report of 3 cases. *GEN*. 1995; 49: 40-41.
2. Yau KK, Siu WT, Law BK, Cheung HY, Ha JP, Li MK. Laparoscopic approach compared with conventional open approach for bezoar-induced small-bowel obstruction. *Arch Surg*. 2005; 140: 972-975.
3. Watt C, Harner J. Bezoars causing acute intestinal obstruction. *Ann Surg*. 1947; 126: 56-63.
4. Zamir D, Goldblum C, Linova L, Polychuck I, Reitblat T, Yoffe B. Phytobezoars and trichobezoars a 10-year experience. *J Clin Gastroenterol*. 2004; 38: 873-876.
5. Chaudhury S, John TR, Ghosh SR, Mishra GS. Recurrent trichobezoar in a case of the trichotillomania. *Indian J Psychiatry*. 2001; 43: 340-341.
6. Loja D, Alvizuri J, Vilca M, Sánchez M. Síndrome de Rapunzel: tricobezoar gastroduodenal. *An Fac Med*. 2003; 64: 71-77.
7. Chogle A, Bonilla S, Browne M, Madonna MB, Parsons W, Donaldson J, et al. Rapunzel syndrome: a rare cause of biliary obstruction. *J Pediatr Gastroenterol Nutr*. 2010; 51: 522-523.
8. Lopes LR, Oliveira PS, Pracucho EM, Camargo MA, De Souza CN, Andreollo NA. The Rapunzel syndrome: an unusual trichobezoar presentation. *Case Rep Med*. 2010; 2010: 841028.
9. Gonuguntla V, Joshi DD. Rapunzel syndrome: a comprehensive review of an unusual case of trichobezoar. *Clin Med Res*. 2009; 7: 99-102.
10. Ho TH, Koh DC. Small-bowel obstruction secondary to bezoar impaction: A diagnostic dilemma. *World J Surg*. 2007; 31: 1072-1078.
11. Ulsan F, Koç Z, Törer N. Small bowel obstructions secondary to bezoars. *Ulus Trauma Acil Cerrahi Derg*. 2007; 13: 217-221.
12. Palanivelu C, Rangarajan M, Senthilkumar R, Madankumar Mv. Trichobezoars in the stomach and ileum and their laparoscopy-assisted removal: a bizarre case. *Singapore Med J*. 2007; 48: e37-39.
13. Kan J, Huang TJ, Heish JS. Laparoscopy-assisted management of jejunal bezoar obstruction. *Surg Laparosc Endosc Percutan Tech*. 2005; 15: 297-298.
14. Lin Ch, Tung Ch, Peng Y, Chow W, Chang Ch, Hu W. Successful treatment with a combination of endoscopic injection and irrigation with coca cola for gastric bezoar-induced gastric outlet obstruction. *J Chin Med Assoc*. 2008; 71: 49-52.
15. Tiwary SK, Kumar S, Khanna AK. Recurrent Rapunzel syndrome. *Singapore Med J*. 2011; 52: 128-130.

### Correspondence:

**Adrián Dersu Pliego-Ochoa**

4<sup>th</sup> year Resident in General Surgery,  
"Dr. Agustín O'Horan" General Hospital,  
Av. Itzaes s/n & Av. Jacinto Canek,  
Col. Centro, 97000, Merida, Yucatan, Mexico.  
Phone: 01-999-930-3320, X- 45500, 45600.  
Cell: 99-9129-0385

**E-mail:** andersu.po@gmail.com

www.medigraphic.org.mx