



Case report

Huge gastric trichobezoar caused by involuntary nocturnal trichophagia and trichotillomania in a female adolescent

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ABSTRACT

Introduction and importance: Gastric trichobezoar consist of the accumulation of chewed hair in the stomach and can extend to other structures of the digestive tract. The relevance of this entity is that it affects the process of nutrition and digestion, and depending on the size and distension of the digestive tract structures, it can cause perforation, infections, cholangitis and obstructive jaundice, intussusception and pancreatitis.

Case presentation: We present the case of a 15-year-old female patient came to the emergency department with a clinical picture of 8 h of evolution, abundant vomiting and lack of appetite for more than 15 days. Physical examination revealed a palpable mass in the epigastrium and left hypochondrium, without pain. Elective laparotomy was performed with findings of a mass of hard consistency and well organized with gastric shape, with hairs intertwined throughout its thickness, weighing 385 g.

Clinical discussion: Currently, there are gaps in the evidence on the best approach to this condition, although both laparoscopic and endoscopic techniques and open surgery have been used, with satisfactory results. However, for the definitive solution of the primary cause, the surgical team must investigate further to avoid recurrence and greater burden of disease in the future.

Conclusion: Gastric trichobezoar may be caused by involuntary nocturnal trichotillomania. Trichobezoar can seriously compromise the nutritional and physical condition of the affected person. The surgical team can inquire about causes of psychological or psychiatric origin, which may explain the development of trichobezoar, giving an answer to the primary cause and avoiding recurrence.

1. Introduction

Gastric trichobezoar, or Rapunzel's syndrome, consist of the accumulation of chewed hair in the stomach and can extend to other structures of the digestive tract [1]. The population that most frequently presents with this condition are young women with psychiatric antecedents, who suffer from trichotillomania or trichophagia [2]. However, there are no precise statistical data on the incidence of gastric trichobezoar in this risk group. In general, it is estimated that the incidence of trichobezoar is around 0.5 % [3,4]. The manifestations caused will be related to the location and extension of the trichobezoar, so they are

unspecific, giving great relevance to the psychiatric history as a plausible risk factor [2,4].

The relevance of this entity is that it affects the process of nutrition and digestion, and depending on the size and distension of the digestive tract structures, it can cause perforation, infections (mainly mycosis and parasitosis), cholangitis and obstructive jaundice, intussusception and pancreatitis [5,6]. Therefore, it is a pathological entity that can seriously compromise the integrity, quality of life, functional capacity and life of the affected person [7,8]. At present, there is no specific solid evidence to manage this condition, so it is solved depending on the experience and tools of the surgical team [8]. However, another point to highlight is the

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fact that in the absence of psychiatric history and lack of information, the surgical diagnosis can identify psychological or psychiatric factors not previously investigated, which can definitively solve the primary cause, preventing the recurrence of trichobezoar in the future. Case reports and case series have reported that trichophagia is a behavior that is strongly associated with trichotillomania [2–5]. However, involuntary nocturnal trichotillomania is more complex to diagnose and treat, and may be referred almost exclusively by an observant family member.

Based on the above, the aim of this study is to report a case of gastric trichobezoar caused by involuntary nocturnal trichotillomania in a female adolescent, discussing the relevance of the surgical team in resolving questions beyond the surgical disease. This case report followed the SCARE guidelines for its realization [9].

2. Presentation of case

A 15-year-old female patient came to the emergency department with copious vomiting for approximately 8 h and lack of appetite for more than 15 days, who received two transfusions in the last two months due to severe anemia with no definite cause. Presented as a relevant antecedent, trichotillomania since childhood. Abdominal exam revealed a palpable mass in the epigastrium and left hypochondrium, without pain. Paraclinical tests were performed with no abnormal findings. Based on the above, the general surgery department suspected gastric trichobezoar, and requested endoscopy of the upper digestive tract, finding a large elongated mass, composed of hair, extending from the fundus to the duodenum, with its most distal part crossing the pylorus, which was found to be open. This confirmed the diagnostic suspicion.

Days later, elective laparotomy was performed without complications (Fig. 1), with findings of a mass of hard consistency and well organized with gastric shape, with hairs intertwined throughout its thickness, weighing 385 g and measuring 17 cm long × 12 cm wide, shaped like a gastric structure (Figs. 2–3). During the postoperative recovery phase, the patient and her relative were questioned about the cause of the trichobezoar, the mother reported involuntary nocturnal trichotillomania, but not trichophagia. Involuntary trichotillomania could be accompanied by involuntary trichophagia, without anyone noticing, and this will trigger trichobezoar. The patient associated this behavior to a sexual abuse trauma suffered by a family member in her childhood, which was never followed up or treated by psychology or psychiatry. The patient evolved satisfactorily, was discharged and referred for management by a multidisciplinary mental health team.

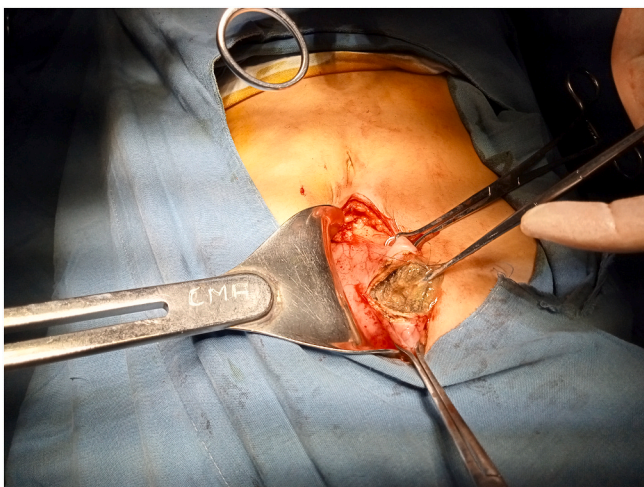


Fig. 1. Intraoperative photograph showing laparotomy and opening of the stomach.

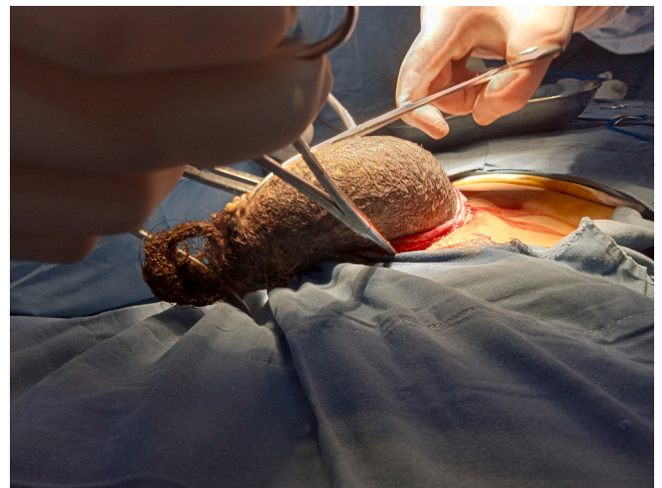


Fig. 2. Intraoperative photograph showing removal of gastric trichobezoar.



Fig. 3. Photograph showing the size of the surgical specimen.

3. Discussion

Among the current gaps in the management of gastric trichobezoar is the choice of surgical technique. Mazzei et al. [10] discuss the considerations to be evaluated when choosing between endoscopy, laparotomy or laparoscopy. The authors argue that small-sized bezoars can be removed endoscopically without difficulty. Laparotomy, performed in our case, may be the technique of choice if the hospital does not have an experienced laparoscopic team or does not have the necessary tools and resources. Likewise, in case of perforation or other complication that requires a more extensive repair [10]. Laparoscopy can be used in stable, low-risk cases. Javed & Agarwal [11] designed about 10 years ago a

minimally invasive technique for the management of large trichobezoar, which consists of a laparoscopic-assisted technique with temporary gastrocutaneopexy, which only requires a single port with a camera and an incision of maximum 5 cm. The same authors report that the average duration of surgery is 45 min, with minimal blood loss and very good functional and aesthetic results at 6 months [11]. Recently, the description of another technique, called combined dual knife-electric snare, was published [12]. It consists of the partial destruction of the trichobezoar using a cauterizing device via endoscopy, taking great care to avoid injuring the stomach mucosa, and then removing the residues [13]. It is really a minimally invasive, reproducible technique that needs further study and dissemination [12]. In our case, laparotomy was performed because of the lack of equipment for laparoscopy.

Among the complications described, which can seriously compromise the life of this group of patients, is peritonitis, either bacterial or fungal, facilitated by the contamination of the hair that is digested, especially in patients with psychiatric antecedents who have poor hygienic habits. Sotoudeh et al. [13] reported the case of an infant who developed fungal peritonitis with fungal balls due to gastric trichobezoar, which presented septic clinical complications, and required a prolonged treatment regimen and reintervention [13]. Those cases where there is some comorbidity, especially at the gastrointestinal level, are a little more complex to treat due to the degree of malnutrition and surgical nutrition process, which allows a complete recovery [14,15].

But a very interesting point to highlight and discuss in this case, is that beyond the approach of the surgical condition, to investigate in greater depth allows that from the surgical approach, a problem of psychiatric or psychological origin can be solved. This makes it clear that surgery is also a discipline that must consider and participate in the integral management of an extra-surgical condition with surgical implications. Obinwa et al. [16] share this view, mentioning that gastric trichobezoar itself is not a primary surgical condition, but that it can trigger a surgical complication. Thus, when surgical manifestations occur, both questions and answers can be derived from surgery [16]. Although this vision is analytical, philosophical and reflective from medical-social education and ethics, in the practice of care, it has an impact on the burden of surgical diseases, health costs, quality of life, and improvement of outcomes. For example, if the surgical team makes an appropriate referral to a multidisciplinary team that can treat the primary cause that triggered the trichophagia and thus the trichobezoar, it decreases or negates the risk of another trichobezoar occurring in the future, necessitating another surgical intervention.

This case allowed the surgical team to uncover an unidentified and untreated case of abuse in an adolescent that could finally be managed and possibly resolved. Then, the impact that surgery could have, from the objectives of global surgery, on the identification and solution of neuropsychiatric disorders in the surgical practice should be taken into account. Finally, the patient was satisfied with the approach she received.

4. Conclusion

Gastric trichobezoar may be caused by involuntary nocturnal trichotillomania. Trichobezoar can seriously compromise the nutritional and physical condition of the affected person. In case of absence of information or psychiatric history, the surgical team can inquire about causes of psychological or psychiatric origin, which may explain the development of trichobezoar, giving an answer to the primary cause and avoiding the recurrence of this entity in the future. There are still gaps in the evidence on the technique of choice in the management of this condition, depending on comorbidities, costs or other variables.

Consent written

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the

written consent is available for review by the Editor-in-Chief of this journal on request.

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All authors equally contributed to the analysis and writing of the manuscript.

Declaration of competing interest

None.

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