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Case Report

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Uncommon Presentation of Giant Gastric Trichobezoar Extending into the Duodenum in a Patient Over 30: A Case Report of Rapunzel Syndrome

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ABSTRACT

Gastric bezoar, a foreign body mass resulting from the accumulation of indigestible material in the stomach, encompasses various types, among which trichobezoar is notable, frequently observed in young females with underlying psychiatric disorders. A trichobezoar consists of tangled hair. This condition is often observed in children who chew on their long hair, with the extension of a gastric trichobezoar into the duodenum or jejunum. Here, we present a case report of a 38-year-old female with gastric trichobezoar concomitant with a psychiatric disorder. The presentation of gastric bezoars tends to be insidious and nonspecific, often remaining asymptomatic for an extended period until reaching a size that triggers symptomatic manifestations. Endoscopy is the preferred diagnostic modality, enabling visualization of the bezoar and guiding treatment decisions. Therapeutic strategies are tailored based on type, size, and consistency; however, surgical intervention typically emerges as the preferred approach, often complemented by psychiatric interventions to prevent recurrence.

Keywords: Rapunzel Syndrome, Trichobezoars, Foreign Body Mass

Introduction

Bezoars are aggregations of non-digestible foreign material aggregated by gastric fluid in the stomach, which is typically a mass in the stomach and can extend into the small bowel; it refers to a mass that becomes trapped in the gastrointestinal system, typically in the stomach, and occasionally in the rectum, mainly discovered in psychiatric females [1-3].

Exactly bezoars can be classified based on their contents, and the four main types are **Phytobezoars** (non-digestible food materials, such as seeds and pits); and **Trichobezoars** (composed of hair. This is often associated with the habit of hair-pulling (trichotillomania and hair-eating "trichophagia"); **Lactobezoars**: (lactose). These are relatively rare and are associated with ingesting undiluted or concentrated formula feeds; **Pharmacobezoars**: (medications. This type can occur when certain medications form solid masses in the stomach that cannot be easily digested. Understanding the composition of a bezoar is crucial in determining the appropriate treatment approach [4].

The overall occurrence of bezoars in pediatric cases remains uncertain. In a 10-year study at a community hospital conducted between 1992 and 2002, 18 adults with bezoars were identified, estimating a hospitalization rate of 0.0125% or a prevalence of 0.6 per 100,000 annually [5]. In another study in Romania spanning from 1992 to 2011, 49 cases were reported over 20 years, accounting for 0.068% of all endoscopies conducted [6].

Delayed gastric emptying due to diabetes mellitus, mixed connective tissue disease, other systemic illnesses, or drugs

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increases the risk of gastric bezoar formation. Other predisposing factors include hypochlorhydria, diminished antral motility, and incomplete mastication; these factors are more common among older people, who are thus at higher risk of bezoar formation [7].

The optimal therapeutic approach for bezoars is a matter of debate due to the absence of randomized controlled trials comparing various options. In some cases, combination therapy may be necessary: **Chemical dissolution**, involving agents like cola and cellulase, is an option for patients with mild symptoms [8]. **Endoscopic removal** is recommended for patients with bezoars that do not respond to dissolution or those with moderate to severe symptoms due to large bezoars. If the initial diagnosis is made through endoscopy, removal can be attempted. Fragmentation techniques, such as forceps, wire snare, jet spray, argon plasma coagulation, or even laser may be employed to break up bezoars, facilitating their passage or extraction [9]. **Surgery** is reserved for cases where chemical dissolution and endoscopic intervention are not feasible or have failed. It is also considered for patients with complications or intestinal bezoars. Open surgery or laparotomy remains the primary treatment for large trichobezoars. However, postoperative complications such as perforation, pneumonia, bleeding, intussusception, wound infections, or unsightly scarring can occur [10].

Clinical Symptoms

Gastric bezoars are usually asymptomatic. When symptoms are present, the most common include postprandial fullness, abdominal pain, nausea, vomiting, anorexia, and weight loss.

Complications

Complications such as gastric ulceration leading to perforation and subsequent sepsis or peritonitis can ensue, with laboratory results possibly indicating anemia or vitamin B12 deficiency [11]. Rarely, bezoars cause serious complications, including Gastric outlet obstruction, Gastrointestinal bleeding secondary to ulceration, Ileus and intestinal obstruction, Perforation and peritonitis, and Intussusception. Over the years, the most frequently documented complications encompass gastric mucosal erosion, ulceration, and perforation in the stomach or small intestine, gastric outlet obstruction, intussusception, obstructive jaundice, protein-losing enteropathy, pancreatitis, and, in severe cases, mortality [12].

Trichobezoar

Trichobezoar are hair concretions that are uncommon and are typically discovered in young females with a psychiatric history, often exhibiting trichotillomania or trichophagia [13,14]. These individuals often deny consuming their hair. In addition, this condition is often observed in children who chew on their long hair by extending a gastric trichobezoar into the duodenum or jejunum. While infrequent, it is crucial not to overlook gastric trichobezoar as a potential diagnosis in females who present with vague epigastric pain. Timely intervention and treatment are essential to prevent complications associated with this condition [15].

Trichobezoars develop in approximately one-third of individuals with trichophagia. While extensively documented in surgical literature, their occurrence is less frequently reported in psychiatric literature [14]. Various factors increase the risk of bezoar formation, including conditions of gastric dysmotility such as diabetes mellitus, gastroparesis, previous gastric surgery, vagotomies, neurological disorders, peptic ulcer disease, gastric cancer, and hypothyroidism. Trichobezoar is a rare medical-surgical condition, with prevalence ranging between 0.4% and 0.6% in different case series [2,6].

Clinical manifestations of trichobezoars can range from serious symptoms, such as hematemesis or small bowel occlusion, to asymptomatic cases that are discovered incidentally through endoscopy [2,16]. The diagnosis and treatment approach are contingent on the observed clinical manifestations. Asymptomatic or minimally symptomatic forms are typically diagnosed and treated using endoscopy, which may involve removal or fragmentation, often assisted by Coca-Cola lavages. On the other hand, complicated cases are diagnosed through abdominal CT scans and usually necessitate surgical intervention [16].

Trichobezoars are frequently linked with psychiatric conditions such as trichotillomania (hair-pulling disorder) and trichophagia (compulsive hair-eating). This behavior is commonly observed in young and adolescent females. Among the various types of bezoars, gastric trichobezoar (GT) is the most prevalent variety in the stomach [17]. Detection of trichophagia and trichobezoar relies on effective screening for related behaviors, preventing potentially life-threatening conditions. Improved collaboration between medical and surgical specialties is vital, addressing therapeutic aspects and acknowledging the likelihood of trichophagia beyond typical clinical expectations [18-21].

Rapunzel Syndrome

Rapunzel syndrome derives its name from the character with long hair in the Grimm brothers' fairy tale. Its initial documentation in medical literature occurred in the West Indies by Duncan et al. in 1994 [22]. Trichotillomania, categorized under DSM-IV-TR 312.39, falls within the psychiatric classification of impulse control disorders [23]. Approximately 50% of all bezoars are gastric, with an incidence ranging from 0.4% to 1%, although this figure is likely underestimated.

This syndrome involves a hairball casting the stomach, with a tail extending beyond the pylorus into the small intestine or even up to the colon [24,25]. Patients afflicted with Rapunzel Syndrome may exhibit symptoms like persistent vomiting, hematemesis or coffee ground vomitus, inability to pass flatus, abdominal pain, and distension [24,25].

Case Report

A 38-year-old married female presented to the Accident and Emergency Department with chief complaints of diarrhea, postprandial vomiting, and frequent abdominal pain for the past three days. She had a history of recurrent similar episodes with an unknown duration.

Upon general physical examination, the patient, a well-appearing, thin-built cachectic with a Body Mass Index (BMI) of 17.30, displayed normal, stable baseline vital parameters. Abdominal assessment revealed tenderness and guarding, more pronounced on the right side, with a palpable mass at the epigastric region. There was difficulty in assessing the lump due to the patient's

irritability and short temper.

The patient was admitted to the surgical department as a case of acute abdomen with malnutrition. Clinically, the initial differential diagnosis was either Anorexia nervosa or gastric tumor.

Abdominal ultrasound indicated unremarkable findings. The Computed Tomography (CT) revealed a gastric mass, suggesting Rapunzel syndrome (Trichobezoar) (Figure 1). However, the patient's sister denied trichotillomania and trichophagia.

Figure 1: Abdominal CT of Rapunzel syndrome (Trichobezoar)

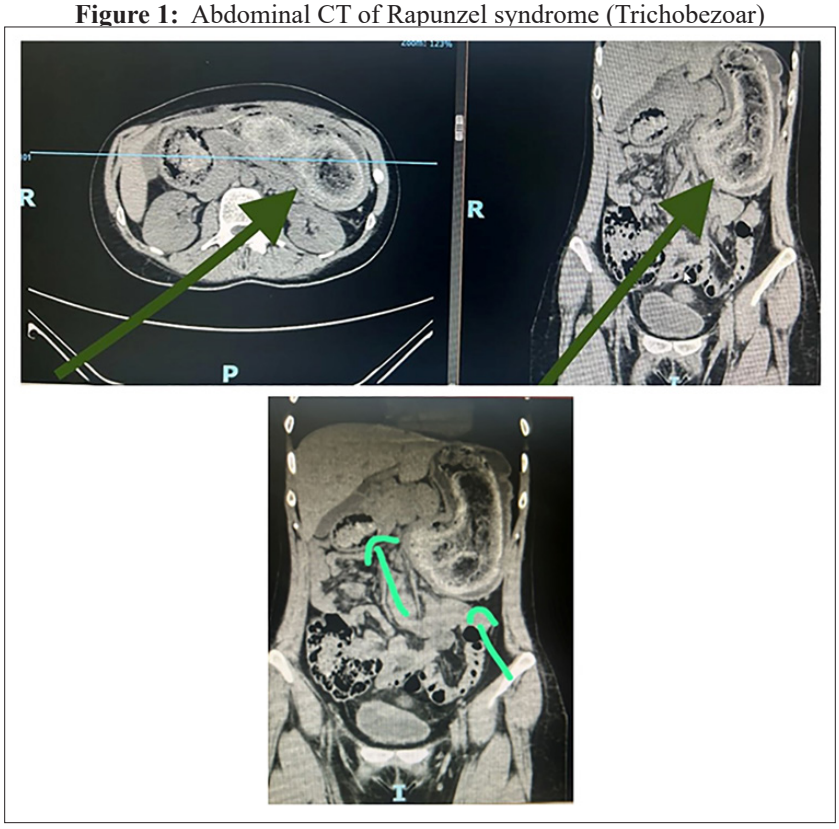

Figure 2: The RapoTrichobezoar

Considering the findings mentioned above, the decision was made to proceed with an attempt at endoscopic removal of the mass. However, the attempt was unsuccessful due to the large hair size and its extension to the 2nd part of the duodenum. A further decision was taken to perform surgery for exploratory laparotomy with a midline supraumbilical incision of 12 cm. Gastrostomy was performed, revealing a partially calcified mass extending to the 2nd part of the duodenum. The gastrotomy closure was done with ENDO-GIA in two layers and with mass closure to the anterior abdominal wall. Drainage was fixed at the surgical site.

The postoperative period was uneventful. The patient was monitored for an additional five days for further management and follow-up and discharged in good condition. The patient was scheduled for follow-up visits in the outpatient and psychiatric clinics.

Discussion

Trichobezoar is a rare gastrointestinal condition characterized by the abnormal accumulation of indigestible materials, such as fibers and hair, predominantly in the stomach compared to other

parts of the digestive tract [16,26]. The earliest recorded instance of trichobezoar dates back to 1854, when Swain described it during an autopsy [27]. Factors linked to the development of trichobezoar include female gender, mental retardation, excessive ingestion of food or non-food substances, and an underlying behavioral disorder associated with pica. In certain instances, the trichobezoar extends beyond the pylorus into the jejunum, ileum, or colon. This specific condition is referred to as Rapunzel syndrome, initially described by Vaughan et al. in 1968 [3,28].

When the bezoar extends from the stomach into the jejunum or beyond, it is referred to as "Rapunzel syndrome," identified in 1968 by Vaughan Jr. et al [11]. This extension increases the risk of severe complications, including gastric mucosal erosion, ulceration, and the potential for stomach or small intestine perforation. Alongside intussusception, unrecognized trichobezoars can result in complications such as obstructive jaundice, protein-losing enteropathy, pancreatitis, and even mortality. It is imperative to remain vigilant for perforation, a recognized complication of trichobezoars affecting the stomach or the intestine, and address potential complications promptly [29].

The presentation of trichobezoar is diverse, ranging from asymptomatic or chronically recurrent abdominal pain to sub-acute or acute gastric obstruction [13]. Gastric perforation, though rare, may also be present. Symptoms can include abdominal pain, early satiety, nausea/vomiting, intestinal obstruction, weight loss, erosive gastritis and esophagitis, and ulcerations leading to bleeding and/or perforation. In some cases, intussusceptions may occur [30]. The most common presenting sign is an abdominal mass [31]. In this specific case, there was also diarrhea, postprandial vomiting, and frequent abdominal pain with recurrent similar episodes. Tenderness and guarding were more pronounced on the right side, with a palpable mass in the epigastric region. There was difficulty in assessing the lump due to the patient's irritability and short temper. It is crucial to consider trichobezoar as a potential differential diagnosis in young females with a history of psychiatric illnesses, a tendency to ingest hair, or complaints of an epigastric mass, weight loss, and epigastric pain [3].

Endoscopy is typically the diagnostic method of choice, revealing the hair mass, often appearing black due to the denaturation of hair proteins by stomach acid. While endoscopy is effective in diagnosing, a CT scan is the most commonly used diagnostic tool in the literature. The CT scan typically shows an intraluminal ovoid heterogeneous mass with interspersed gas, a characteristic finding. The CT scan is beneficial for highlighting other locations beyond the stomach [32,33].

The enlargement of a trichobezoar hampers the blood supply to the stomach mucosa and portions of the intestine, resulting in ulceration and eventual perforation. Nirasawa et al. (2009), were the pioneers in reporting the laparoscopic removal of a trichobezoar. Laparotomy is the preferred treatment due to its high success rate, relatively low complication rate, and overall lower complexity [34]. Furthermore, the entire gastrointestinal tract can be swiftly assessed for additional masses. The development of GT (trichobezoar) is still not fully grasped. Hair strands, due to their smooth texture, successfully avoid peristaltic movements and become entangled in the crevices of the gastric mucosa. The accumulated hairball remains dormant, enabling the trichobezoar to grow in size and weight through ongoing, progressive hair ingestion. Ultimately, the trichobezoar assumes the shape of the stomach, usually consolidating into a singular, solid mass [35].

The prevalent symptoms include epigastric pain (70.2%), the presence of an epigastric mass (70%), nausea and vomiting (64%), hematemesis (61%), weight loss (38%), and occurrences of diarrhea and constipation (32%). A potential preoperative indication of trichobezoar may be discerned in patients exhibiting severe halitosis, patchy alopecia, a history of trichotillomania, and trichophagia. Additionally, associated complications of GT involve malabsorption issues such as protein-losing enteropathy, iron deficiency anemia, and megaloblastic anemia [35].

Valenciano et al. (2012), documented that perforation of either the stomach or intestine is the most common complication, occurring in 10.1% of cases, followed by intussusception (1.85%), pancreatitis (0.92%), and cholangitis (0.92%) [36].

The management of trichobezoars involves the removal of the mass, addressing complications, and preventing recurrence. Trichobezoars exhibit variability in terms of consistency, size, and location. While smaller trichobezoars can be endoscopically removed using a basket or direct suction, larger ones present challenges, with limited reports on their removal. Management options for trichobezoar include endoscopic removal, laparoscopic removal, or laparotomy removal. The optimal therapy seeks to balance minimal invasiveness with optimal efficacy. Endoscopic fragmentation or aspiration is the preferred alternative among non-invasive procedures and should be prioritized over surgical treatment, particularly in uncomplicated cases [33,35]. However, this approach has its limitations. The complete removal of all fragments may necessitate repeated introduction of the endoscope, posing risks such as esophagitis, pressure ulceration, and, in rare cases, esophageal perforation. In complicated cases where gastrointestinal perforation or small bowel occlusion has developed, surgical treatment may be warranted [35,37]. Moreover, fragments of a large trichobezoar may migrate after repeated manipulation through the pylorus, leading to intestinal obstruction further down the digestive tract. Gorter et al. (2010), in a retrospective review of 108 cases of trichobezoars, assessed the various treatments attempted in these cases [37]. The study noted that only 5% of attempted endoscopic removals were successful (small trichobezoars can respond to endoscopy removal with mechanical and laser fragmentation techniques and vigorous lavage). In comparison, 75% of attempted laparoscopic surgical extractions were successful. However, according to their findings, laparotomy with gastrotomy achieved a success rate of 99%, making it the preferred management choice [35].

It can be inferred that laparoscopy is considered inferior to laparotomy in treating trichobezoar. In preterm babies, medical treatments, such as enzyme therapy with acetylcysteine, cellulase, or papain, may be attempted in cases involving small bezoars, medicational bezoars, or gastric lactobezoars. Still, they are generally ineffective in larger bezoars or those with

complications. As a result, non-operative treatments, despite their appeal due to being less invasive, are rarely employed due to their high failure rates [3,33].

In this particular case, exploratory laparotomy with a midline supraumbilical incision was chosen due to an unsuccessful attempt at endoscopic removal of the mass due to the large size of the hair and its extension to the 2nd part of the duodenum. Although laparoscopy offers advantages in terms of cosmetic outcomes, reduced postoperative complications, and a shorter hospital stay, however in laparotomy, a meticulous examination of the entire gastrointestinal tract is more feasible, aiming to prevent secondary intestinal obstruction arising from the migration of residual bezoars. Laparotomy, considered the gold standard, is the most effective and commonly employed technique in medical literature.

Following the removal of a trichobezoar, the prognosis is generally favorable if successful psychiatric therapy effectively controls habitual trichophagia. Therefore, post-surgery psychiatric consultation plays a crucial role in managing trichophagia. Despite preventive measures, it is important to note that the recurrence rate of trichobezoars is 13.5%, highlighting the need for ongoing vigilance and follow-up care [26].

Conclusion

Gastric bezoars, while uncommon, can pose a potentially life-threatening risk if left untreated. Trichobezoars should not be overlooked as a potential differential diagnosis, especially in young females who present with an epigastric lump and chronic epigastric pain. It is advisable to conduct a psychiatric evaluation of the patient to delve into the underlying causes of the issue.

The diagnosis can be efficiently established through endoscopy, and CT scans are valuable in highlighting potential involvement in other locations. Various therapeutic approaches are available, and timely treatment is essential to prevent complications.

Beyond its surgical implications, Rapunzel syndrome demands a multifaceted approach encompassing psychological evaluation and emotional support. Collaboration between medical, surgical, and psychiatric specialties is crucial to address the underlying psychiatric factors and prevent recurrence in cases of trichobezoar.

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