



Case report

Trichobezoar: A case report of a double gastric and ilial localization revealed by an occlusion

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ABSTRACT

Introduction and importance: A bezoar is an agglutination of ingested materials forming an indigestible and insoluble mass inside the gastrointestinal tract. Trichobezoars formed by ingested hair are rare. The stomach tends to be the typical location. In some cases there is an extension to the duodenum, jejunum or even to the colon and it is called Rapunzel syndrome.

Case presentation: We present a case of an occlusion due to double trichobezoars in 17 years old woman who underwent an extraction by enterotomy and gastrotomy.

Clinical discussion: Trichobezoar is a compact conglomeration of swallowed hair and makes up less than 6% of all bezoars. It appears in young women under 20 years of age who carry psychiatric or chronic metabolic diseases that can alter the behavioral balance. Physical examination is often poor in uncomplicated forms: forms without peritonitis or occlusion or hemorrhage or appendicitis or pancreatitis but it may reveal a well-limited, smooth, firm, and mobile epigastric mass. For uncomplicated forms or forms complicated by hemorrhage, the reference examination requested in the first line remains the esophagogastroduodenoscopy (EGD) which allows a dual role both diagnostic and therapeutic. Computed Tomography (CT) remains the preferred imaging modality requested urgently in case of an occlusion or peritonitis presentation. Endoscopic extraction represents the therapeutic approach for uncomplicated forms but surgical treatment remains the main therapeutic means of trichobezoar.

Conclusion: Trichobezoars are rare and present both diagnostic and therapeutic challenge. Laparotomy remains the most successful surgical treatment although laparoscopy currently appears to have promising results in expert hands. A post-operative psychological management is essential to correct psychobehavioral disorders of patients to prevent recurrence.

1. Introduction and importance

A bezoar is an agglutination of ingested materials forming an indigestible and insoluble mass inside the gastrointestinal tract. There are several types of Bezoars, namely: phytobezoars, lactobezoars, pharmacobezoars and trichobezoars [1,2]. Trichobezoars formed by ingested hair occurs in 5–10% of patients with trichophagy, which is present in only 1% of those with trichotillomania. It also occurs following the ingestion of artificial hair. This can alter the therapeutic management [3]. The stomach tends to be the typical location. In some cases there is an extension to the duodenum, jejunum or even to the colon and it is called Rapunzel syndrome [4]. The double localization of trichobezoars, which is the case of our patient, is considered for some authors as part of Rapunzel syndrome but remains a separate entity for others [4]. This

pathology is found especially in young adolescent women with a history of psychiatric disorders [1,2]. There are no specific semiological criteria referring to this disease and symptoms take so much time to appear which make the diagnosis more difficult. The clinical presentation varies depending on the site, size and whether or not a complication has occurred.

EGD is the gold standard for positive diagnosis and treatment of uncomplicated forms. The diagnosis of the complicated forms is established by medical imagery and their main treatment is based on surgery [2].

We report the case of a young 17-year-old patient who has a double localization of gastric trichobezoar and complicated ilial stenosis.

This case report is reported in line with SCARE guidelines [5].

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2. Case presentation

Seventeen years old patient, with congenital mental retardation, who had early postprandial vomiting for 3 years, was admitted for an occlusive syndrome with on examination: the patient was stable, afebrile. Abdominal examination found a distended tympanic abdomen with generalized abdominal tenderness. The examination of all abdominal hernial orifices were normal. In biology, she had a biological inflammatory syndrome: White blood cells (WBC) = 17,480 elmt/mm³; C-reactive protein (CRP) = 82.03 g / dl, hyponatremia at 111 mmol/l and hypokalemia at 3.22 mmol/l.

The abdominal CT showed a distension of the small bowel loops (maximum 52 mm) with an ileal loop containing a bezoar associated with gastric distension containing a voluminous bezoar (Figs. 1,2).

The patient underwent surgery with a median incision approach. In surgery, an acute intestinal occlusion was found upstream of a mobile intraluminal mass located 2 m from the ileo-caecal valve without signs of suffering (Fig. 3). The exploration of the rest of the digestive tract had shown a second mass that embraced the shape of the stomach (Fig. 4). The gesture consisted of extraction of the trichobezoar by enterotomy at 2 m of ileo-caecal valve associated with gastrotomy (Fig. 5). The post-operative course was normal and the patient was discharged on 10th post-operative day with a psychiatric consultation appointment to avoid any recurrence.

3. Clinical discussion

Trichobezoar is a compact conglomeration of swallowed hair and makes up less than 6% of all Bezoars [6,7]. It often occurs in children with mental retardation and it is seen as early as the age of 3 years [8]. It can also appear in young women under 20 years who carry psychiatric or chronic metabolic diseases that can alter the behavioral balance such as renal terminal insufficiency and celiac disease [9]. This mental

imbalance can be manifested by trichomania attitudes that are accompanied by trichophagy in 5–10% of cases and present with trichobezoar in 1% [3]. However the trichobezoar may be the result of an accumulation of artificial hair (dolls) or the combination of both human and artificial ones which can forbidden the endoscopic treatment.

There are no specific semiological criteria referring to this disease and symptoms take so much time to appear witch make the diagnosis more difficult.

Trichobezoar can manifest by abdominal pain especially epigastric, nausea, vomiting, transit disorders such as diarrhea or constipation, early satiety and asthenia with weight loss.

This pathology has several complications. The complicated forms are summed up in digestive hemorrhage due to ulcerations of the gastric wall, mechanical occlusion, digestive perforation witch can be responsible of peritonitis or sub-phrenic abscess or digestive fistula, acute pancreatitis, and appendicitis [10,11] (Table 1).

However, the discovery may be incidental during an etiological investigation carried out as part of an exploration for anemia or hypoproteidemia or growth retardation [12].

Physical examination is often poor in uncomplicated forms but may reveal a well-limited, smooth, firm, and mobile epigastric mass.

In our case, the 17-year-old patient with mental retardation presented an acute high bowel obstruction without signs of severity.

For additional studies requested to support the diagnosis, the indication differs according to the clinical presentation. For uncomplicated forms or forms complicated by hemorrhage, the reference examination requested in the first line remains the EGD which allows a dual role both diagnostic and therapeutic [2,3,9]. Diagnosis of complicated forms is based on urgently requested imaging. The abdominal X-ray without preparation is a non-specific examination that may show hydroaeric levels in order to confirm a clinical diagnosis of occlusion or an image of pneumoperitonium synonymous to gastro-intestinal perforation. Abdominal ultrasound, visualizing a superficial band, hyperechogenic,



Fig. 1. CT axial view showing the double location of bezoar: stomach (a), small bowel (b).

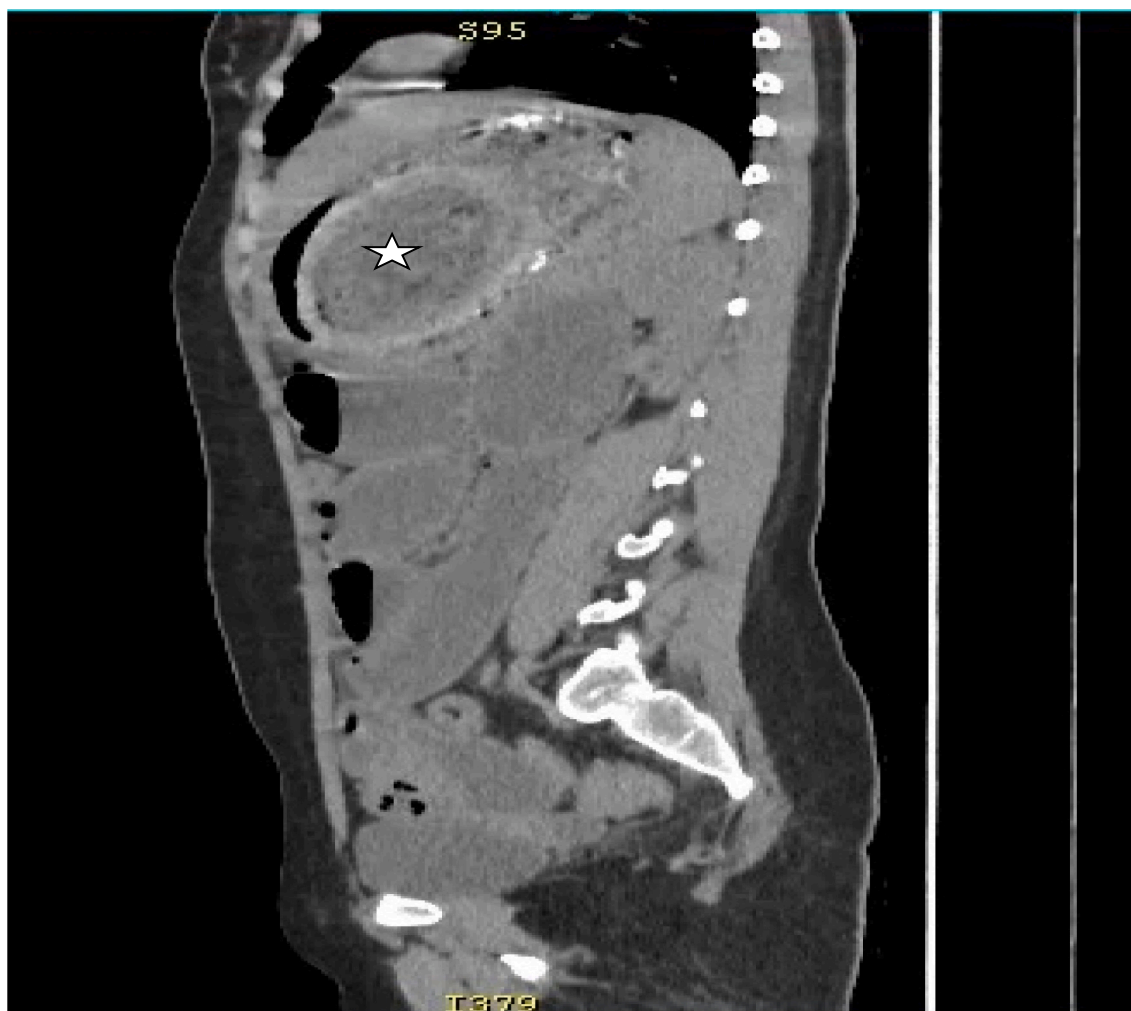


Fig. 2. CT coronal view showing a bezoar in the stomach as outlined by contrast (star).

curvilinear with a posterior shadow cone leads to correct diagnosis in 25% of cases [13].

Abdominal CT remains the preferred imaging modality requested urgently in case of an occlusion or peritonitis presentation. It allows positive diagnosis by showing a mass of variable, heterogeneous volume, occupying almost the entire gastric lumen and consisting of multiple concentric circles of different densities distributed in onion bulbs. Two pathognomonic and constant signs are the presence of tiny air bubbles dispersed within the mass and the absence of any attachment of the mass to the gastric wall [14]. The scanner also allows to specify the size of the obstacle, its location and its extension, and determines the signs of complications such as ischemia or perforation.

In our case, the abdominal X-ray without preparation, there were hydroaeric levels of the ilial type and the abdominal CT showed a distended stomach containing a voluminous bezoar as well as a distension of the small bowel loops (maximum 52 mm) with a transitional level corresponding to a bezoar blocked at the level of an ileal loop.

Multiple therapeutic modalities have been described in the literature, depending on the symptomatology, size, location and whether or not a complication is present. Thus for those of small size the medical treatment can be proposed namely the prescription of abundant drinks associated with transit accelerators [14].

Endoscopic extraction represents a second therapeutic approach and the first endoscopic ablation of a trichobezoar was attempted 20 years ago [2,3,9]. The latest review of the endoscopic treatment of trichobezoar published in 2020 and including 47 articles showed the success of

endoscopic treatment for 16 patients out of 52 patients (30.7%) [4]. The success of endoscopic treatment is conditioned by the size of the obstacle which must not exceed 7 cm of long axis and its weight which must not exceed 55 g, a preferred esophageal or gastric seat, the absence of Rapunzel syndrome and the experience of the treating physician [3]. This therapeutic modality should not be indicated in complicated forms or if the composition of trichobezoar suggests the presence of artificial hair whose electrocoagulation or the use of laser yag for fragmentation of large mass is not devoid of risk of silent burns of the digestive wall with the possibility of scab fall and subsequent perforation responsible for peritonitis [3].

Surgical treatment remains the main therapeutic means of trichobezoar. The incision differs on a case-by-case basis and we distinguish the broad median laparotomy which is the reference approach and allows treatment in 99% of cases, elective incisions and more recently the laparoscopic treatment with a success rate of 75% [2]. This treatment is indicated immediately in complicated cases, large masses, multiple locations or after endoscopic treatment failure. In fact, surgery allows the exploration of the entire digestive tract, the extraction of gastric trichobezoar through a gastrotomy, as well as the extraction of possible extensions (tail) or fragments blocked at a distance from the stomach through one or more enterotomies [1,14]. Our patient was operated by a broad median incision. In fact, we have chosen surgical treatment for three principles reasons; first, the huge size of the trichobezoar, then the rapunzel syndrome specially the second location on the small bowel and finally the occlusion which make endoscopic treatment almost



Fig. 3. surgical extraction of the ileal trichobezoar.



Fig. 4. A stomach-shaped trichobezoar.



Fig. 5. trichobezoars of the stomach and ileum.

Table 1

Clinical symptoms of trichobezoar [4,10].

Clinical features	Percentage
Abdominal pain and vomiting	88%
Abdominal distension	47%
Epigastric mass	41%
Severe malnutrition	23%
Occlusion	23%
Peritonitis	12%
Gastrointestinal hemorrhage	6%
Appendicitis, pancreatitis	–

impossible and even dangerous. The exploration showed a double gastric and small bowel localization. A huge trichobezoar that hugs the gastric wall and another smaller responsible for an occlusion at the level of a slender loop located two meters from the ileocecal valve.

This pathology is endowed with a high morbidity and postoperative mortality of up to 30% or the need to stem psychological disorders in order to prevent recurrence, the indicators of possible recurrence are anemia, weight loss, post-prandial vomiting and the appearance of abdominal mass, and thus the importance of family-psychotherapist collaboration for early detection to avoid significant mortality.

4. Conclusion

Trichobezoars are rare and present both a diagnostic and therapeutic challenge. Clinical symptomatology depends on the size of the trichobezoar and whether or not complications are present.

EGD remains the reference examination for uncomplicated and early forms. It plays a role not only in diagnostic but also therapeutic. For complicated forms, multiple localisations and large masses, laparotomy remains the most successful surgical treatment although laparoscopy currently appears to have promising results in expert hands. A post-operative psychologic management is essential to correct psychobehavioural disorders of patients to prevent recurrence.

Ethical approval

It is exemption from ethical approval because it is an observation report.

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Declaration of competing interest

No conflict of interest.

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