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Case Report

Lessons Learned From a Rare Type of Gastric Bezoar

E Lacombe[1]*, E Kadz[1], B Des-Marez[2], L Dedeken[3], A Salame[4], K Khelif[5] and P-Q Lê[1]

[1]Pediatric department, Etterbeek-Ixelles Hospital, Brussels, Belgium

[2]Gastroenterology department, Etterbeek-Ixelles Hospital, Brussels, Belgium

[3]Hemato-oncology department, Hôpital Universitaire des Enfants Reine Fabiola, Brussels, Belgium

[4]Gastroenterology department, Hôpital Universitaire des Enfants Reine Fabiola, Brussels, Belgium

[5]Surgery department, Hôpital Universitaire des Enfants Reine Fabiola, Brussels, Belgium

***Address for Correspondence:** E Lacombe, Pediatric department, Etterbeek-Ixelles Hospital, Brussels, Belgium, E-mail: l_manoli@hotmail.com

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Background

A bezoar is a compact mass stuck in the alimentary canal. The condition is more commonly seen in female adolescents and in institutionalized children with neurological or psychiatric disorders. Bezoars' classification is based upon their composition. Phytobezoars (vegetable aggregates), trichobezoars (undigested hairs), and pharmacobezoars, are the most common types. More rarely, a large variety of other miscellaneous materials can be found such as paper, tissue, vinyl gloves, stones, chewing-gums, candies and even sponges [1,2].

There is generally a large delay in the diagnosis of gastro-intestinal bezoars. Therefore, the true incidence in the pediatric population cannot be known accurately and is probably underestimated. If no treatment is administered, the morbidity and mortality rate, mainly due to gastric bleeding and perforation remains high [3].

Case Presentation

A 12-year-old boy was admitted to our pediatric emergency unit for hyperemesis and abdominal pain localized at the right-upper-quadrant, lasting for a period of 12 hours. He had not eaten nor drunk since the beginning of the symptoms. There was no fever, no diarrhea or other signs of infection.

His medical history included multiple episodes of acute otitis media that led, in time, to partial deafness and slight neuro developmental delay. He was therefore institutionalized in a special education school. He also suffered from attention deficit and hyperactivity disorder, for which a daily treatment of methylfenidate hydrochloride was started three years ago.

Physical examination showed pallor, ill appearance associated with abdominal pain. There were no acute abdominal signs and hemodynamic parameters were normal. After initiation of intravenous fluid therapy and administration of alizapride hydrochloride at the emergency department, the patient was hospitalized for further investigations.

Investigations

Initial hematological blood test showed a hemoglobin level of 14.2g/dl, leukocytosis at 7300 cells/µl with a neutrophilic count of 6205 cells/µl and platelet count of 373000/µl. The C-reactive protein was negative (<0.3mg/l). Electrolytes and other biochemical tests were normal. No pathogens were isolated in the blood, urine and stool cultures. Serologies were also negative. Abdominal radiography showed a normal gastric gas pattern, homogenous colonic distribution of feces and gas and no signs of digestive perforation.

Abdominal ultrasonography (US) was normal.

During the first days of hospital stay, the abdominal pain fluctuated, following an 'on-off' pattern. The symptoms and the general state of the patient improved during the initial phase of hospitalization, when fasting was initiated. This is the reason why further investigations were not performed initially. The symptoms worsened when feeding was attempted on day six and day nine. When present, the abdominal pain was regularly followed by food vomiting, which was refractory to intravenous alizapride hydrochloride.

Treatment

At day eleven, a light-sedation-upper endoscopy, realized by an adult gastroenterologist, revealed a foreign body obstructing the duodenal bulb. The mass was difficult to fragmentate and therefore could not be aspirated through the esophagus (Figure 1). A second conservative endoscopic approach was tried on day twelve, using this time, a total anesthesia. After a long and laborious procedure, only partial removal of the foreign material could be performed. A third unsuccessful attempt was realized on day fourteen by a pediatric gastroenterologist, which resulted into an iatrogenic esophageal hematoma with longitudinal ulceration (Figure 2). Finally, a supra-umbilical incision followed by a five-centimeter antrotomy permitted the surgical removal of the foreign body. This later had the rubbery texture and macroscopic appearance of a chewing gum aggregate and measured 3.5x5.3x4.2 cm (Figure 3). No complications where noted during the post-operative course and during follow-up. Further interrogation of the family revealed that the patient had swallowed a big amount of candies and chewing gums during a Halloween party, two days before the onset of symptoms. It appeared also that he had had previous episodes of gum ingestion in the past.

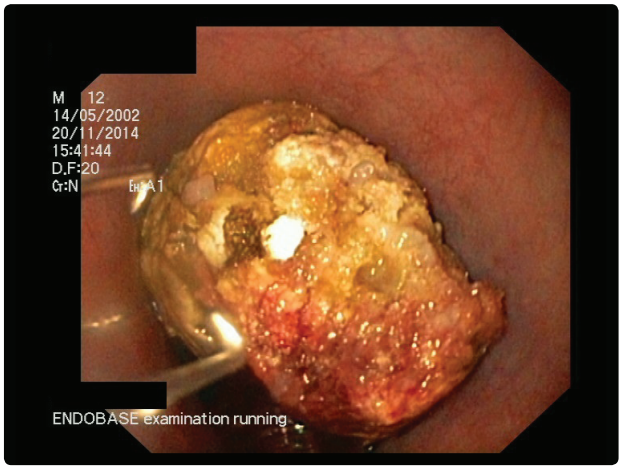

Figure 1

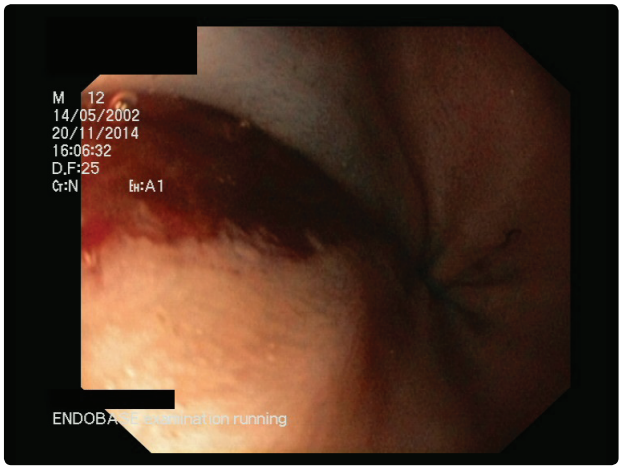

Figure 2

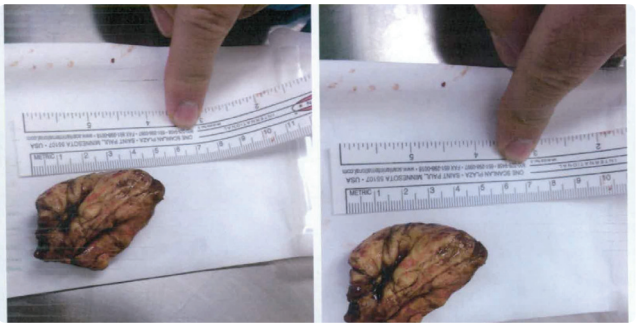

Figure 3

Discussion

According to the DSM-IV classification, Pica syndrome is a disorder characterized by the persistent eating of non-nutritive substances for a period of at least one month. There is a 25% incidence of Pica syndrome among the developmental delayed population [4] and it is recognized as one of the most dangerous forms of self-injurious behavior exhibited by this group of people, which can lead, in time, into death and other life-threatening events [5]. Some authors have suggested that it may be the consequence of parental neglect and deprivation in early years [6].

Our patient presented slight neuro developmental impairment and was institutionalized in a special education school. In addition, after further questioning of the parents, it appeared that it was not the first episode of excessive gum ingestion. In view of this and given the present diagnosis, it can be hypothesized that he suffered from Pica syndrome, at that time.

Bezoars can mimic many other gastro-intestinal diseases and represent a real diagnostic challenge, especially in the pediatric population. The most common complaint reported, in the emergency department, is abdominal pain [7,8]. Other frequent clinical signs and symptoms are nausea and post-prandial vomiting, early satiety, epigastric discomfort, anorexia, weight loss, hematemesis, diarrhea or constipation [2,7]. It is important to emphasize the fact that our patient presented with symptoms of vomiting undigested food after his meals and improved clinically when fasting was initiated.

Thus bezoars are easily missed and especially in mentally impaired patients [9,10]. Therefore, diagnosis should be evoked when institutionalized children with developmental disabilities, or with previously known medical history of Pica, present in the emergency department with epigastric or abdominal pain, post-prandial hyperemesis, or other acute abdominal symptoms.

A wide range of complications due to bezoars have been reported including gastric ulcers, protein-losing enteropathy, steatorrhea, vitamin deficiencies, pancreatitis, intussuseption [7,11,12]. Gastro-intestinal obstruction, perforation, as well as gastric bleeding, are mainly responsible for the mortality rate of up to 30%, if no treatment is administered [3].

There is generally a large delay in the diagnosis of gastro-intestinal bezoars. In most of the documented cases no clinical suspicion is reported before carrying out radiological or surgical intervention [2]. On abdominal radiographs, the bezoar appears as a mottled heterogenous mass that may be mistaken for a food-filled stomach.

The administration of barium contrast identifies the presence of a mobile concretion. On abdominal US, it appears as a band of increased echogenicity with loss of posterior echoes. Abdominopelvic computed tomography examination reveals an intraluminal heterogenous aggregate with entrapped air. Endoscopy permits direct visualization and determination of the bezoar's composition, and therefore is useful in establishing treatment modalities Park, et al. [2] showed that endoscopy is the most effective approach for gastric bezoars diagnosis (diagnostic ratio 88.1%), followed by barium study (85.2%). Abdominopelvic computed tomography seemed to have less diagnostic value and US did not show any utility [8].

According to published studies, surgery used to be the initial treatment modality for all gastro-intestinal bezoars [8]. However, after the first endoscopic bezoar removal, performed by Mc Kechnie [13], this new technique has been realized more frequently and with increasing success [7,8,12]. Endoscopic fragmentation of the bezoar can nowadays be performed using a wide range of instruments including water jet, mechanical lithotripter, large polypectomy snares, electrosurgical knife, drilling, or laser destruction. The fragments are then cleared with a large channel endoscope or a dormia basket [8,12]. The reported success rate of endoscopic treatment for bezoars has increased in recent studies, from 71.5 % to 100 % [8].

Due to the low prevalence rate, there is still little evidence and no standardized approach according to the treatment modalities of gastrointestinal bezoars in children. This latter generally depends upon the size, the location and the associated symptoms. Chemical dissolution is commonly attempted for patients presenting small bezoars and mild symptoms. The mechanism of bezoar dissolution by cola has not yet been well explained, but its mucolytic effect and its acid pH are thought to be important elements. For patients with moderate symptoms or presenting large bezoars resistant to chemical dissolution, a conservative endoscopic approach is considered to be the first therapeutic choice. Surgery is generally reserved when endoscopic fragmentation cannot be performed, fails, or for patients presenting severe complications [7,11,8].

Interestingly, some authors have mentioned cases of spontaneous bezoar resolution without any specific treatment. This raises the possibility of considering, in some specific situations, an expectative attitude with regular follow up, as first therapeutic choice. Further studies must be conducted to identify patients for whom this spontaneous dissolution is likely to be expected [7,14].

Therapy for gastric bezoars has also to be tailored to the nature of the concretion. Phytobezoars are, because of their composition, relatively soft and easy to dissolve. Thus, administration of cola is considered to be, for these conglomerates, the first therapeutic choice[15,16]. On the contrary, the hairs of a trichobezoar are firmly matted together and form a solid mass [12]. In that case, chemical degradation, pharmacotherapy and endoscopic fragmentation have been reported to be ineffective. Indeed Gorter, et al. [17] reviewed in their study, 40-reported trichobezoar cases for which endoscopic removal was attempted. The procedure was unsuccessful in 38 of those cases and open surgery had to be achieved. Therefore, the authors recommended surgical approach as first-line treatment for these kind of aggregates [17]. Surgery has also been recommended as initial treatment of bezoars composed of vinyl gloves [1,18]. These examples stress the fact that surgery still has higher success rates than endoscopy and remains necessary for some specific types of concretions [8].

Concerning the particular case of chewing-gum bezoars, there is, to our knowledge, very little information concerning the specific clinical hazards associated with the ingestion of large amount of chewing gum and their treatment. A literature research using the keywords 'chewing-gum' and 'bezoar' and with limits activated ('humans' and 'ages' from 'birth to 18 years'), was performed using PubMed, in April 2016. The literature associated with pediatric chewing-gum bezoars exclusively deals with case reports. Only six of them were found, of which five were relevant. Most of these documents report low-situated-large-bowel bezoars, which led to fecal impaction, and were easily treated manually or with enemas [19,20]. Interestingly, Rimar and his team, report their experience of successful endoscopic treatment of a gastric chewing-gum bezoar found in an 18-year-old woman, who was in the habit of swallowing at least five wafers of chewing gum per day. However, neither the size, nor the details of the procedure are related [21]. On the contrary, Michel, et al. [22] report an unsuccessful endoscopic attempt, during the treatment of a large gastric bezoar composed of both hair and chewing-gums, in a 16-year-old patient. This later had to be removed surgically[22].

Consequently, and according to the above mentioned examples, it appears that no standardized therapeutic approach is defined for this indication.

This present case confirms the complexity of endoscopic resolution regarding large gastric-chewing-gum bezoars. Because of their elastomeric and therefore rubbery texture, these aggregates can be laborious or impossible to split by endoscopy and multiple session therapy can easily lead to procedural complications. Indeed, despite constant technological improvement, endoscopic procedures can still lead to various iatrogenic complications including perforation, laceration, hematoma, and ulceration of the gastric wall, as well as small-bowel obstruction caused by the downstream migration of the fragmented bezoar [,8,10,11]. These complications occur especially during the treatment of large bezoars, that, such as in our case, tend to require multiple endoscopic sessions [8,11].

As mentioned before, similar endoscopic limitations, linked to the nature of the concretion, have already been observed for tricho and vinyl glove-bezoars, for which an initial surgical approach is generally recommended [1,17,18]. In addition to that, endoscopic procedures are known to be time consuming, technically challenging, and require highly trained professionals, with great experience in a wide range of endoscopic techniques [11]. Therefore, it appears that an iterative endoscopic therapy may not be recommended in the particular case of large gastric chewing gum bezoars and surgery should be considered rapidly after a first unsuccessful endoscopic attempt realized by a highly trained endoscopist.

Conclusion

This is a rare case report of large gastric-chewing-gum bezoar formation in a 12-year-old boy. Bezoars' symptoms are unspecific and the condition can therefore be easily missed. If no treatment is administered, morbidity and mortality rates remain high.

The present case shows that a large chewing-gum bezoar may be very difficult to split or manipulate endoscopically, also considering that such a procedure can easily lead to iatrogenic complications. As a result, an iterative endoscopic therapy may not be recommended.

Surgery should rather be considered after a first unsuccessful endoscopic attempt.

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