Case Report

Gastric trichobezoars in children: A clinical case report

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Abstract

Trichobezoarele gastrice la copii: prezentare de caz clinic

Trichobezoarul reprezintă o concrețiune a părului ingerat depistat la un anumit nivel în tractul digestiv, care apare cel mai frecvent la pacienții cu tulburări psihice, care mestecă și înghit propriul păr (trichotilomanie și trichofagie).

Existând riscul dezvoltării unor potențiale complicații grave, se impune necesitatea unui diagnostic promt și rapid, care va determina alegerea unei opțiuni adecvate de tratament.

Autorii prezintă un caz clinic de trichobezoar gastric diagnosticat la o pacientă în vârstă de 13 ani cu trichotilomanie. Diagnosticul a fost stabilit în baza examenului radiologic, inclusiv tranzitul baritat și tomografia computerizată, examenul endoscopic punând în evidență o masă cenușie de dimensiuni impresionante, de culoare neagră, localizată în stomac.

Cazul dat a fost rezolvat prin îndepărtarea chirurgicală a unui trichobezoar uriaș de dimensiuni 20 cm x 6 cm. Postoperator, pacienta a fost supusă unui tratament de recuperare cu realimentare treptată, beneficind și de consiliere pisihiatrică.

Autorii conchid, că în pofida faptului că trichobezoarele de dimensiuni mici pot fi extrase endoscopic după un tratament conservativ preventiv, tratamentul de elecție în trichobezoarele de dimensiuni majore este cel chirurgical, în perioada postoperatorie pacienții necesitând un tratament de recuperare treptată și o evaluare psihoemotivă cu scop de a preveni reapariția acestora.

Cuvinte cheie: trichobezoar, sindromul Rapunzel, ocluzie intestinală, tratament chirurgical

Abstract

Trichobezoar is a concretion of ingested hair found at a certain level in the digestive tract, which occurs most commonly in patients with mental disorders, who chew and swallow their own hair (trichotillomania and trihophagia).

Given the risk of developing potential serious complications, the need for a prompt and rapid diagnosis is required, which will determine the choice of an appropriate treatment option.

The authors present a clinical case of gastric trichobezoar diagnosed in a patient 13 years of age with trichotillomalania. The diagnosis was established on the basis of the radiological examination, including barite transit and computer tomography, the endoscopic examination highlighting a gray mass of impressive size located in the stomach.

This case was solved by the surgical removal of a huge trichobezoar of dimensions 20 cm x 6 cm. Postoperatively, the patient underwent a recovery treatment with gradual refeeding, benefiting from psychiatric counseling.

The authors conclude that despite the fact that small trichobesores may be endoscopically removed after a preventive conservative treatment, the treatment of choice in the trichobezores of major dimensions is the surgical one, in the postoperative period the patients needing a gradual recovery treatment and a psycho-emotional evaluation, in order to prevent their recurrence.

Keywords: Trichobezoar, Rapunzel syndrome, intestinal obstruction, surgical treatment

Background

The bezoar is a tumor formation containing swallowed substances, which are most commonly located in the stomach and small intestine. Based on their components, they are classified into phytobezoars that are made up of undigested fruit and vegetable fibers, trichobezoar -made up of hair and mixed bezoars consisting of gauze, paper, fungi, etc. [4, 12].

The most common type of bezoars are the trichobezoars mostly found in children with mental disorders. Specialized literature has described only a few cases of trichobezoars among children. The clinical picture onset is usually asymptomatic (eventually being detected during a routine investigation), whereas the clinical signs may develop in complicated cases. The most common complications involve intestinal obstruction (Rapunzel syndrome), intussusception, acute appendicitis, intestinal perforation and hemorrhage [1, 5]. The diagnosis is often challenging requiring differential diagnosis. Most cases of trichobezoars are treated surgically, whereas in Rapunzel syndrome, the treatment of the underlying disease is necessary to prevent recurrences. The bezoar disease ("morbus bezoaris") remains a rare pathology, which has so far been little studied by specialists. Thus, this is a major reason for a delayed diagnosis once the severe complications have already developed. Further basic investigations will include abdominal ultrasonography and radiography, fibrogastroduodenoscopy. However, CT (computed tomography) and NMR (nuclear magnetic resonance) are required for a more accurate topical diagnosis [2, 12].

Trichobezoars are commonly treated with spasmolytic, analgesic drugs and vegetable oil, which can be effectively in small trichobezoars. Endoscopic attempts may be performed to retrieve the trichobezoars via fibrogastroscopy. In case of huge trichobezoars and

their complications (perforation or hemorrhage), a surgical intervention is required [2, 7, 11].

Throughout the last five years, 4 children from the Department of Pediatric Surgery were diagnosed with GI trichobezoars (2 children with gastric trichobezoars, of which one child exhibited a huge type and was operated on, whereas another child had a small-sized trichobezoar and self-induced vomiting). 2 children revealed small bowel trichobezoars, who underwent a conservative therapy. In order to alert the importance of an early diagnosis and treatment, we would like to report a clinical case of a 10-year-old girl with gastric trichobezoar, who showed specific symptoms and was hospitalized in our clinic for a period of several months.

Clinical case report

A 13-year-old girl was found to have intra-abdominal tumor formation, following an additional investigation for recurrent abdominal algic syndrome. Over the last month, the patient often experienced upper abdominal pain, vomiting, loss of appetite and nausea. During a detailed discussion, her parents stated that the child has a habit of pulling and eating hair, resulting in considerable weight loss within the last year. The clinical examination revealed a mobile, hard-elastic and slightly tender to touch tumor mass within the mesogastric region with a size of 20 x 6 cm and without signs of peritoneal irritation.

The barium swallow showed an enlarged, dilated and non-homogeneously opaque stomach with partial gastric emptying and contrast medium retention at intervals up to 12 hours after the ingestion (fig. 1).

Fibrogastroscopy revealed a mass of hair particles that occupied the entire gastric lumen and extended to the pylorus. The CT detected the presence of a gastric trichobezoar formation (fig. 2).

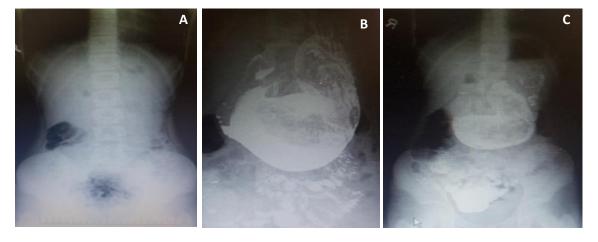


Fig.1. Abdominal X-ray: A - on an empty stomach; B - more than one hour after barium medium swallow; C - over 12-hour interval



Fig. 2. The abdominal CT detected a homogeneous mass within the stomach, which occupied almost the entire lumen with signs of partial intestinal occlusion.

The preoperative children were administered symptomatic and parenteral nutrition therapy. Following an improvement in general condition, the children were subjected to a surgical intervention under general anesthesia via endotracheal intubation. The upper midline laparotomy and the inspection of the abdominal cavity revealed an enlarged stomach as well as collapsed small bowel loops ("hungry gut"). The laparotomy and the gastrostomy were made along the longitudinal axis, which revealed a giant hair mass, occupying almost the entire gastric lumen with a short tail of hair extending to the pylorus and some hairs present in the duodenum). The gastric trichobezoar was removed and the gastric incision was sutured.

The extracted trichobezoar was examined. It resembled the shape of the stomach, having dimensions of 20x6x2 cm and weighing 150gr (fig. 3).



Fig. 3. Intraoperative piece. Giant trichobezoar.

The postoperative course was favorable, without complications. After the improvement of the child's overall condition and removal of the sutures, a psychologist and psychiatrist consulted the child in order to undergo a specific treatment. Subsequently, the child's condition has improved, followed by an increase in appetite and weight gain, as well as missing abdominal algic syndrome. The child was discharged 14 days postoperatively, having a good health condition.

Discussions

Trichobezoars particularly occur in girls with long hair, most commonly in teenagers [5, 8]. It commonly occurs due to the hair that is trapped in the gastric mucosal folds. Swallowed hairs gather and form a large hair bolus covered with gastric mucosa, which impairs the gastric peristalsis. The trichobezoar then gets too large to leave the stomach and that might result in gastric atony [2, 4, 11].

Rapunzel syndrome is a specific form of trichobezoar [1, 9, 10]. There have been used a number of methods and criteria to specify the diagnosis of Rapunzel syndrome. The researchers have found the following identical characteristics for Rapunzel syndrome: gastric trichobezoar of any size with a tail extending through the pylorus into the small intestine, the presence of symptoms suggestive of intestinal occlusion (a partial or a complete one) and mental impairment: trichophagia – compulsive eating of hair and trichotillomania [8, 10].

There are no pathognomonic or characteristic signs for bezoars. The most common clinical features include anorexia, weight loss, vomiting syndrome, recurrent abdominal pain, and detection of abdominal "pseudotumors" [2, 5]. The long-term evolution of the disease might lead to various complications, such as anemia, erosive gastritis, intestinal obstruction, malabsorption syndrome, intussusception and appendicitis [3, 10, 11].

The diagnosis is based on the patient's history taking, physical examination and imaging studies. The diagnostic standard includes: abdominal ultrasonography, fibrogastroscopy and computed tomography, as well as upper GT X-ray [2, 4, 5, 12]. The major treatment approach of bezoars is to remove the tumor mass and prevent its recurrence. The treatment of choice depends on the bezoar's consistency, size and location, thus it can be removed conservatively, by endoscopy or by a surgical intervention [1, 4, 7]. The treatment of bezoars can be initiated by using vegetable oil and spasmolytic drugs. In case of trichobezoars, a combined therapy of pancreatic enzymes and papain syrup (from Papaya fruit) can be administered [5, 6].

An attempt of endoscopic extraction via a rigid fibrogastroscopy may be used in small and medium sized bezoars; however, it is associated with various complications such as iatrogenic esophageal perforation and hemorrhage [2, 4]. At present, the emergence of mininvasive surgery enabled the use of the laparoscopic

techniques for extraction of small and medium bezoars. Laparoscopy might be used to remove gastric trichobezoars if these are not large and can be fragmented by using special endoscopic bags. Nevertheless, it proves ineffective when extracting large-sized trichobezoars (> 20 cm). Thus, the surgical intervention is the treatment of choice in complicated cases [2, 3, 7, 11].

Conclusions

1. Small-sized trichobezoars might be extracted by fibrogastroscopy following a conservative and fragmentary therapy.

- 2. The endoscopic extraction of gastric bezoars may allow avoiding surgery or might be an alternative treatment to surgical intervention.
- The treatment of choice in giant trichobezoars is the surgical intervention via the upper laparotomy and gastrostomy. A psychologist should council children postoperatively in order to avoid recurrences.

REFERENCES

- Eryilmaz R., Sahin M., Alimoglu O., Yildiz M.K. A case of Rapunzel syndrome. Ulus Travma Acil Cerrahi Derg. 2004; 10:260-3
- 2. Holcomb III G.H., Murphy J.P., Ostile D.J. Ashcraft's Pediatric Surgery, Sixth Ed. 2014.
- 3. Jensen A.R., Trankiem C.T., Steve Lebovitch S. et al. Gastric outlet obstruction secondary to a large trichobezoar. JPS. 2005; 40:8.
- 4. Lee J. Bezoars and foreign bodies of the stomach. Gastrointest. Endosc. 1996; 6: 605-19.
- 5. Naik S., Gupta V., Naik S. et al., Rapunzel syndrome reviewed and redefined. Dig. Surg. 2007; 24(3):157-61.
- 6. Nichol P.F., Rollins M.D., Muratore C.S. Fundamentals of Pediatric Surgery. Second Ed. 2017.
- 7. Prasad A.J., Rizvon K.M., Angus G. et al. A giant trichobezoar presenting as an abdominal mass. Gastrointest Endosc. 2011; 73(5):1052-4.
- 8. Santos T., Nuno M., Joao A. et al. Trichophagia and trichobezoar: Case report. Clin. Pract. Epidemiol. Ment. Health. 2012; 8:43-5.
- Singla S.L., Rattan K.N., Kaushik N., Pandit S.K. Rapunzel syndrome: A case report. Am. J. Gastroenterol. 1999; 94(7):1970-1.
- 10. Zent R.M., Cothren C.C., Moore E.E. et al. Gastric trichobezoar and Rapunzel syndrome. Am. Coll. Surg. 2004; 990.
- 11. Веселый С.В., Сопов Г.А., Латышов К.В. и др. Инородные тела желудочно-кишечного тракта у ребенка на фоне частичной обтурационной кишечной непроходимости. Дет. Хирургия. 2012; 1:50-1.
- 12. Гиткевич А.Э., Шмаков А.П., Зуев Н.Н., Зуев Н.И. Трихобезоар желудка и тонкой кишки у ребенка. Дет. Хирургия. 2010; 1:48-9.