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Case report / Приказ болесника

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Multidisciplinary treatment of massive trichobezoar-caused an acute gastric outlet obstruction

Мултидисциплинарно лечење циновског желудачног трихобезоара – ретког узрока акутне опструкције желуца

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SUMMARY

Introduction Trichobezoars presents a rare form of bezoar made of swallowed hair, with clinical manifestation of gastric or intestinal obstruction, gastric ulceration, bleeding, and perforation. It is predominantly found in emotionally disturbed or mentally retarded youngsters, who eating their own hair which is clinically known as trichophagia. Patients often deny eating their own hair which makes diagnosis difficult.

Case report We are presenting a case of acute gastric outlet obstruction caused by giant gastric trichobezoar made of a long thin hair, in a 20-year young female. Although patient had a long history of trichophagia, she didn't think her behavior was unusual and she has not been treated before. Followed initial diagnostic procedures, exploratory laparotomy was indicated. After anterior gastrotomy was performed, a massive stomach-shaped trichobezoar was removed. Postoperatively patient had a psychiatric consult exam. She recovered well and discharged without complications. She was referred for further psychiatric follow-up.

Conclusion Trichobezoars are non-digestible collections that usually accumulates in stomach and can extend to small bowel, causing mechanical injury such as hollow viscus obstruction. Patients with acute gastric obstruction caused by a giant trichobezoar require urgent removal of the trichobezoar, to preserve the stomach and avoid further, catastrophic consequences.

Keywords: trichobezoar; gastrotomy; trichophagia; emotional disturbance

САЖЕТАК

Увод Трихобезоари представљају редак облик безоара насталих од прогутаних длака, са клиничком манифестацијом опструкције желуца или црева, улцерације желуца, крварења и перфорације. Углавном се откривају код емоционално поремећених или ментално ретардираних младих, који једу сопствену косу, што је клинички познато као трихофагија. Пацијенти често поричу да једу сопствену косу што отежава дијагнозу.

Приказ случаја Представљамо случај акутне опструкције желуца изазване огромним желудачним трихобезоаром сачињеним од дугих танких длака, код 20-годишње младе жене. Иако је пацијенткиња имала дугу историју трихофагије, није мислила да је њено понашање необично и да раније није лечена. Након почетних дијагностичких процедура, индикована је експлоративна лапаротомија. Након учињене предње гастротомије, уклоњен је масивни трихобезоар у облику желуца. Постоперативно пацијенткиња је прегледана од стране психијатра. Опоравила се и отпуштена је без компликација. Упућена је на даље психијатријско праћење.

Закључак Трихобезоари су несварљиве колекције које се обично акумулирају у желуцу и могу се проширити до танког црева, изазивајући механичка оштећења, као што је опструкција шупљих органа. Пацијенти са акутном опструкцијом желуца изазваном циновским трихобезоаром захтевају хитно уклањање трихобезоара, како би се сачувао желудац и избегле даље, катастрофалне последице.

Кључне речи: трихобезоар; гастротомија; трихофагија; емоционални поремећаји

INTRODUCTION

Bezoars represent a undigested mass within the gastrointestinal tract which enter the stomach by swallowing non-absorbable materials [1,2]. It could increase in size passing further into the small intestine [1,2]. Trichobezoars are rare clinical entity, but they have been known for centuries [1–7].

They are consist of a bunch of hairs in the proximal gastrointestinal tract [1–7]. Hairs enter the stomach by compulsive swallowing, the disorder is called trichotillomania [7]. Trichotillomania is classified as an impulse control disorder, with prevalence estimate to 2% [3, 7]. Repetitive hair pulling may be unconsciously or unintentionally done [3]. The inability to control the impulse to pull one's hair out, is followed by an increase in tension which is replaced by a feeling of satisfaction after the act [8]. Trichophagia is hair swallowing occurs in up to 18% of patients with trichotillomania [1–8] Finally about 33% of patients with trichophagia develop stomach trichobezoars [1–8].

Trichobezoars are most often found in young women, in female children and adolescents with underlying behavioral disorder, but it can occasionally affect healthy adult [2, 4, 5]. Piles of hair create a ball in the stomach that grows slowly for years and is asymptomatic for a long time. Over time, first non-specific symptoms occur such as nausea, vomiting, abdominal pain, anorexia, early satiety, or weight loss. Trichobezoars can enlarge and creates a mechanical damage due to their presence in the lumen of the stomach and intestines. This can lead to serious complications such as gastric mucosal erosions, gastric ulceration, gastric outlet obstruction, pneumatosis, perforation and peritonitis [6, 9].

Trichobezoars are foreign bodies that must be removed. After visualization endoscopic extraction of large trichobezoars often fails. Complete removal of the massive trichobezoars and treatment of the intestinal damage caused by its presence is only possible surgically.

We are presenting a case of acute gastric outlet obstruction caused by giant gastric trichobezoar made of a long thin hair, in a 20-year young female. Consequences of her behavioral disorder in this young woman was conformed on the CT scan. After surgical removal of the massive trichobezoar, she recovered without adverse events. Patient will continue with psychiatric follow-up.

CASE REPORT

A 20-year-old female was referred to the surgical examination in Emergency Center (EC) with intermittent epigastric pain and vomiting, which started two weeks prior. Patient's medical history revealed loss of appetite and early satiety. She previous visit to the EC six months before because of the same symptoms when an abdominal ultrasound and upright abdominal radiography showed no specific findings. On general physical examination, abdominal palpation of epigastric region revealed smooth and hard intrabdominal curved lump approximately 10 cm in diameter. Differential diagnosis of gastric trichobezoar and gastric malignancy was made with further diagnostic tests. Abdominal ultrasound showed a large mass in the epigastric region with echogenic anterior margin. An abdominal computerized tomography (CT) scan showed a distended stomach with a clearly demarcated intraluminal mass extending from the gastric cardia to the duodenal bulb which was described as heterogeneous dense formation with interposed air inclusions and without postcontrast enhancement (Figures 1a, 1b, 1c). A gastric trichobezoar was diagnosed and gastroenterologist endoscopist was consulted.

Urgent esophagogastroduodenoscopy (EGDS) showed small amount of esophageal blood, foreign body regurgitation and stomach filled with hair and impacted food obstructing 80% of the gastric lumen with completely obstructing the antrum, disabling further propagation of the endoscope. Considering the size of the mass and the extent of bezoar on the CT scan it was thought that endoscopy would likely fail to remove the mass completely without large fragmentation and the risk for distal intestinal obstruction or gastric perforation. Thus, the patient was referred for emergency surgical treatment.

After adequate preoperative treatment by the anesthesiologist, a few hours after admission to the hospital she was operated. An exploratory laparotomy through an upper midline abdominal incision was performed. During exploration, smoothly contoured mass was

found occupying whole stomach and D1 of duodenum Exploration of other visceral organs was uneventful. A longitudinal 7 cm gastrotomy incision several cm above pylorus was made on the anterior surface of the corporal region of the stomach (Figures 2a, 2b, 2c). A massive trichobezoar of the stomach and duodenum was extracted (Figure 2b, 2c). The gastrotomy was closed in two layers and the abdomen was closed with drainage.

The patient made an excellent post-operative recovery with no complications. Postoperatively the patient was examined by a psychiatrist, this was her first psychiatric exam. According to anamnesis data, she started to bite the ends of her hair and then to pull it out at the root at the age of 13. She denied the existence of any significant stressors in both that period of her life, as well as presently. The diagnosis of trichotillomania was confirmed. During hospitalization oral intake was allowed on 5th post-op day. She was discharged from Surgery department on 7th post-op day in stable condition, with advice for psychotherapy and psychiatric follow-up to avoid recurrence.

This study was done in accordance with the institutional standards on Ethics.

DISCUSSION

Trichobezoars are foreign bodies, they are solid masses that cannot be digested. It occurs most often in young people, young women with behavioral disorders and retarded children [4–9]. Trichobezoars are formed when indigestible hairs are retained in the stomach, forming a ball, and instead of being expelled through the peristaltic wave, remains in the lumen, retaining parts of undigested food [1–7, 9]. Over time this solid mass “grows”, takes the shape of the stomach and extends distally into the duodenum and small intestine, create the Rapunzel syndrome [4, 10, 11, 12].

If the patient continues to eat the hair, the trichobezoar gets bigger and bigger, this process progresses slowly, and the symptoms are non-specific for years. Sometimes

fermentation of the bezoar give the patient's breath a putrid smell which could be a warning clinical sign [6].

Trichobezoars occurs most often in young people, young women with behavioral disorders and retarded children [3, 8].

Trichobezoars, with their mechanical effect on the mucosa of the lumen of the stomach and intestines, can cause minor or major and even catastrophic complications, such as organ perforation [3, 8, 10, 11, 12].

About 6% of all the bezoars presenting with symptoms of gastric outlet obstruction.

We presented a rare case of trichobezoar, which manifested itself as a surgical emergency due to acute gastric obstruction in a young woman who had been swallowing her own hair for years, without the idea of asking for a psychiatric support and without the idea that she could harm herself. She denied the existence of the psychology or external stressors. Considering her first contact with a psychiatrist in the context of the existing conditions this could be understood as an expression of less mature psychological defense mechanisms, as well as dissimulative behavior. In a situation where the first psychiatric examination is performed as part of the treatment of complications of the underlying disease, like in this case of trichotillomania, only detailed psychological and psychiatric exploration, the complete exploration patient's life, as well as examination outside the context of current events can provide sufficient insight into their mental status [3, 8].

The complications of gastric trichobezoar are different, it can rarely detach, and made satellites [11, 12]. Mirza et al. presented 17 cases of trichobezoars with different complications, there was one case that had a satellite small bowel trichobezoar in addition to the gastric trichobezoar [13]. Sometimes the signs of small bowel obstruction with intraluminal mass and the real cause of the intestinal obstruction can recognized late, which is one of the dangers of bezoars. Therefore, it is recommendation that in any case of gastric trichobezoar, during

abdominal exploration surgeon should looking for a secondary trichobezoar in small or large bowel.

Trichobezoars most often present with one of three clinical types: acute presentation due to complications made by massive lesions, asymptomatic or atypical picture in small or stable lesions, and incidental lesions. If there is no evidence about behavioral disorder, small trichobezoars can be asymptomatic for a long time, diagnosis is based on clinical suspicion and on the correct selection of diagnostic methods and radiological imaging. Depending on the size, location and mechanical injuries caused by trichobezoar the symptoms could be non-specific (dyspepsia, loss of appetite, nausea, weight loss) or dramatic [9–15]. Most often, trichobezoars are lesions located in the stomach, leading to increased pressure on the mucosa, erosion of the mucosa, malnutrition, gastric ulcerations, bleeding and even gastric perforation [14, 15]. Severe clinical finding is intestinal obstruction in about 25% of the patients and perforation and peritonitis in about 18% cases [7,13–19]. It is important to detect the existence of trichobezoar in time to avoid serious complications. In our case, the patient had an undetected trichobezoar for years, she did not consult a doctor until acute gastric obstruction occurred.

Treatment is multidisciplinary and includes a different specialist, psychiatrists, gastroenterologists, and surgeons in case of complications. Insight into the existence of trichophagia, indicates a detailed clinical, endoscopic, and radiological examination. The presence of hair in the stool and the clinical finding of a palpable mass in the abdomen, especially in the epigastrium, are specific clinical signs. Even abdominal ultrasound and plain radiograph are not sensitive in cases of large gastric trichobezoars, plain radiographs can show a gastric shaped opacified area, which corresponds with heterogenous intragastric mass on ultrasound.

The most sensitive in diagnostic algorithm is abdominal CT shows some distinctive features as well-defined non-attached intraluminal mass with air bubbles retained within the

interstices [1, 2, 15]. EGDS is presented as the gold standard because it serves both the diagnosis and the therapeutic evacuation of the trichobezoars but it is not suitable for all patients [16]. The therapeutic approach using EGDS is not suitable for all patients, as was the case with our patient because it was impossible to fragment the solid impacted gastric mass and there was a risk of unintentional stomach injury. Treatment is complete removal of the foreign body, endoscopically or surgically, by laparoscopic or open surgical approach [17–21]. We performed open midline laparotomy with gastrotomy following by complete bezoar extraction, without any complications. Some studies reported only 5% of successful endoscopic bezoars removals, successful rate of 75% for laparoscopic extraction and 99% successful for conventional laparotomy with gastrotomy [17–21]. Therefore, laparotomy is considered the treatment of choice. To completely eliminate the lesion from stomach or intestines, there is no clinical evidence of the drugs therapy effectiveness, such as enzyme therapy, and only mechanical removal of the lesion is considered successful [18].

Multidisciplinary treatment is mandatory, and in addition to mechanical removal of bezoars, is psychiatric support and treatment of trichomania as an impulse control disorder. To help the patient it is necessary to discover biopsychosocial specificities in the origin and development of the clinical characteristics of trichotillomania [3, 8]. The success of the treatment in this case can only be attributed to the good teamwork of the surgeon, gastroenterologist, psychiatrist who established the first contact with patient, the initial examination and initial diagnostic evaluation, and further psychiatric supervision. Treating trichotillomania is complex and includes pharmacotherapeutic, socio-therapeutic and psychotherapeutic modalities. In the treatment of these patients the most important results are provided by behavioral or behavioral-cognitive psychotherapy techniques [3, 8]. It is important to emphasize that inadequately treated trichomania is accompanied by relapses when trichophagia occurs, and therefore psychiatric follow-up is necessary [3, 8].

After surgical removal of trichobezoars, clinical and EGDS follow-up is advised during next 6, 12 and 24 months due to potential abdominal complications or early detection of recurrence of trichophagia with asymptomatic trichobezoars in the stomach or intestines [19, 20, 21].

This case emphasizes the importance of trichobezoars as a differential diagnosis in early diagnostic and therapeutic procedures in young females with non-specific upper abdominal complaints and an epigastric palpable mass. It is also important to understand that it is a complex clinical problem, primarily a disorder of impulse control which requires psychiatric treatment, while surgery or endoscopy only solve the consequences of this behavior disorder. In patients with trichobezoar and trichotillomania, serious complications may occur due to the presence of a foreign body in the stomach or intestines, and another problem is relapses of the disease. The experience with our patient shows that only the interaction of medical knowledge and skill from different medical disciplines can provide the best understanding and the most successful treatment of this disease.

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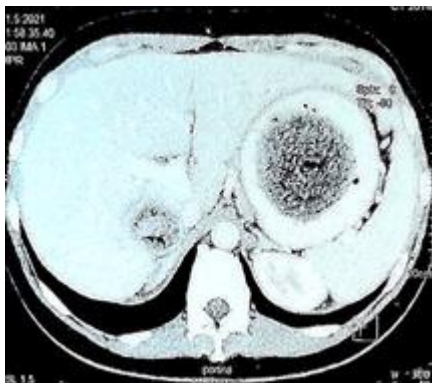
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Paper accepted

Figure 1a. Abdominal computed tomography scan – axial sections show a trichobezoar as non-enhancing intraluminal mass in stomach



Paper accepted

Figure 1b. Abdominal computed tomography scan – coronal section shows a trichobezoar as a non-enhancing intraluminal mass with trapped air bubbles in the interstices which is distending the stomach



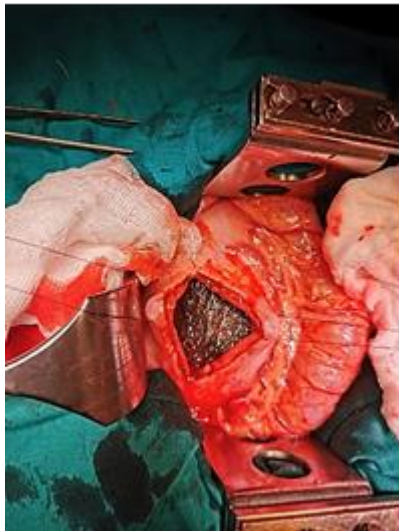
Paper accepted

Figure 1c. Abdominal computed tomography – axial sections that shows a trichobezoar as intraluminal mass in stomach and tail in duodenum



Paper accepted

Figure 2a. Gastrotomy, a massive compact foreign body completely occupies the lumen of the stomach



Paper accepted

Figure 2b. Intraoperative trichobezoar extraction following a gastrotomy



Paper accepted

Figure 2c. Trichobezoar forming a cast of the stomach



Paper accepted