Trichobezoar with small bowel obstruction in children: Two cases report

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ABSTRACT

A trichobezoar is a mass of cumulated hair within the gastrointestinal tract. Stomach is the common site of occurrence. Intestinal obstruction due to trichobezoar is extremely rare. The authors report two cases of a trichobezoar obstructing the terminal ileum in one and the jejunum in another.

Key words: Small bowel obstruction, surgery, trichobezoar

INTRODUCTION

Trichobezoar usually occurs in patients with trichotillomania history, it is characterized by a compulsive behavioral disorder of pulling one's hair, combined with trichophagia that consists of ingesting that hair[1-3]; it typically occur in the stomach[2-5] and rarely affects the small intestine. It is also a cause of small bowel obstruction.[1,3,4]

In this report, we describe two cases of an atypical localization of a trichobezoar in a 9 and 15-year-old girls, who presented with small bowel obstructions. A review of the clinical presentation, radiographic findings, and surgical management is performed.

CASE REPORTS

Case 1

A 9-year-old girl had a paroxysmal abdominal pain for 4 months; she was presented to the emergency department with various symptoms consisting of acute intestinal obstruction, distension, vomiting, and a complete constipation during the last 7 days. The clinical examination revealed an anxious patient. The abdomen was tender associated to peritoneal signs. The laboratory assessment was found a marked leucocytosis of 15,000/cm^3 with normal blood chemistry. The X-ray of the abdomen showed multiple air-fluid levels without any gas under the diaphragm [Figure 1]. The ultrasound of the abdomen did not reveal any anomaly. Laparotomy was performed through medline incision of <3 cm, which revealed significantly distended small bowel loop with a bowel mass in the ileum measuring 5 × 3 × 3 cm^3 situated at 20 cm from the ileocaecal junction [Figure 2]. Enterotomy was performed and found an unclean smelling mass of hair threads in the form of a bunch, the mass was removed and the enterotomy was sutured using separate points [Figure 3]. There was neither bezoar in the stomach nor duodenum and proximal jejunum. The postsurgery follow was favorable. The patient was referred for psychiatric treatment.

Case 2

A 15-year-old girl without any particular history, who presented with abdominal pain and fecaloid vomiting during the previous 15 days. At admission in the hospital, the swollen abdomen was diffusely sensitive to palpation. The blood counting showed leucocytes at 18,700 elements/μL, and hemoglobin at 10.2 g/dL demonstrating a profile of an anemic patient with a functional renal deficiency. The X-ray of the abdomen showed tow air fluid levels [Figure 4], while the ultrasound examination did not reveal any abnormality. The upper gastrointestinal (GI) contrast radiography found a well-delimited trichobezoar at jejunal level respecting the intestinal wall [Figure 5]. The laparotomy demonstrated trichobezoar in the jejunum located at approximately 15 cm of the Treitz angle measuring 8 × 4 × 4 cm^3 [Figure 6], without any parent bezoar in the stomach. The enterotomy was performed [Figure 7] and the mass was removed; finally the enterotomy was sutured using separate points [Figure 8]. The patient was discharged with no recurrence of the symptoms. She

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Figure 1: The X-ray of the abdomen showed multiple air fluid levels without any gas under the diaphragm



Figure 2: Distended small bowel loops with a bowel mass in the ileum measuring 5 × 3 × 3 cm3 about 20 cm from the ileocaecal junction



Figure 3: Trichobezoar removed surgically

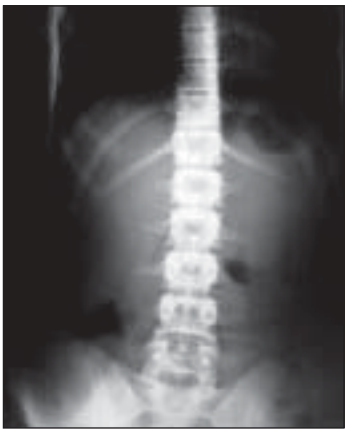


Figure 4: X-ray of the abdomen showed tow air fluid levels



Figure 5: The upper gastrointestinal (GI) contrast radiography found a well-delimited image at the jejunal level, respecting the intestinal wall



Figure 6: A bowel mass in the jejunum, located at approximately 15 cm of the Treitz angle, measuring (8 x 4x 4 cm)

was referred to the psychiatry department for etiological treatment.

DISCUSSION

Trichobezoar (hairball) is a complication of trichotillomania. It consists of recurrent hair pulling, and subsequent trichophagia or mouthing of the hair.[1,4,6,7] Bezoars are foreign bodies in the lumen of the digestive tract. The lumen size increases in time by the accumulation of ingested nonabsorbable food or fibers.[3-6] Baudamant reported the first case of a human trichobezoar in 1779. In 1896, Stelzner described the first correct preoperative diagnosis of trichobezoar on



Figure 7: The enterotomy found a trichobezoar

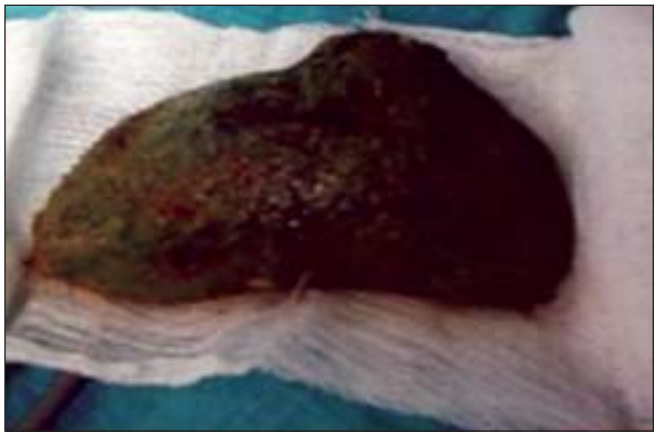


Figure 8: Trichobezoar removed surgically

the basis of physical examination.[3] Bakey and Ochsner reviewed published literature and showed that 171 cases of trichobezoar, 119 cases of phytobezoar, and 13 cases of other concretions were reported.[3,5] Schonborn performed the first surgical removal of a trichobezoar in 1883.[3]

The bezoar is mostly caused by the presence of indigestible substance in the lumen. Some substances encourage stickiness and concrete formations.[2,4,8] Bezoar occurs mainly in young women, who chew and swallow their hair (trichobezoar), vegetable fibers (phytobezoar), persimmon fibers (diospyrobezoar), or semi-liquid masse of drugs (pharmacobezoar).[2,3,5] During the time, these substances are retained by mucus and become enmeshed; this yields a mass having the shape of the stomach localization where they are usually found.[2,4] These substances attend large size due to the chronicity and delayed investigation of the affection. The age of occurrence of bezoars has been reported to range between 1 and 56 years old; the most dominant fraction of age is comprised between 15 and 20 years old with 90% of females.[1,3,4] About 10% of patients have shown psychiatric abnormalities or mental retardation.[2] Although about 1 of 2000 children suffer from trichotillomania, trichophagia is rarely seen, and a bezoar does not occur in all children with trichophagia.[3,6] Bezoars mostly originate at the level of the stomach,[1-4] it is probably related to high fat diet causing unspecific symptoms like epigastric pain, dyspepsia, and postprandial fullness. The stomach is not able to exteriorize hair and other substance out of the lumen because the friction surface is not sufficient for propulsion by peristalsis. The bezoars might also occur with GI bleeding (6%) and intestinal obstruction, or perforation (10%).[2] The term "Rapunzel syndrome" was assigned to trichobezoars extending continuously through the entire length of the small intestine as a tail and was first described by Vaughan *et al*. in 1968.[3,5]

Primary small bowel bezoars without any associated gastric bezoar are uncommon.[1,3,4] Small bowel obstructions caused by bezoars, which rise within small bowel diverticula were reported. A decade later, a neonatal resection was performed for intestinal atresia. Recently, Carmon *et al*.[4] described two cases of small bowel obstruction from bezoar formation in dilated jejunum. Reduced intestinal mortality is the most quoted factor in the intestinal bezoar formation. This last one is usually caused by a portion of the gastric trichobezoar which became detached to cause small or large bowel obstruction. In rare cases, it is caused by the trichobezoar itself such is the case for our patient. The most common sites of obstruction are the gastric outlet, or duodenum. Obstructions of distal parts of the small bowel or the large bowel are extremely rare.[3] Biological investigations might show anemia. It is well demonstrated that iron deficiency anemia is rather a result and not a cause of trichophagy; this was established since the iron level was normal in most patients with trichotillomania and trichophagy. The examination of the hair content in stool would establish the diagnosis, but usually it is not done.[3]

Various imaging modalities have been recommended for detection of bezoars.[3,5] The imaging findings are helpful in diagnosing trichobezoar. The conventional radiography shows a masse of opaque soft tissue in a swollen stomach.[1,3,5] A calcified rim may delineate the edge of the bezoar.[1,3] The ultrasonography shows a typical curvilinear trichobezoar with bright echogenic band, this does not allow transmitting the ultrasound waves which generate a shadow over the left upper quadrant. The high echogenicity of hair and the presence of multiple acoustic interfaces created by trapped air and food limits the ultrasonography of the trichobezoars.[1,3,8]

Both, the contrast radiography and the endoscopy of the upper GI tract are the diagnostic procedures of choice for establishing the diagnosis.[3,8] The upper GI contrast radiography confirms the existence of the trichobezoar and might detect other complications such as gastric ulcers. In addition, the upper endoscopy is definitively the diagnostic support for trichobezoar; it might be used for endoscopic retrieval of proximal small trichobezoars.[3] The computed tomography (CT-scan) is the most useful diagnostic tool in patients with bezoars because it reveals the localization of the bowel obstruction; it shows also a well-defined intralumina mass of the bezoar in the transitional zone of the obstruction. A mottled gas pattern in the mass is reported characterizing the bezoar, and it is supposed to be air bubbles retained within the bezoar.[8,9]

Recently, researchers have recommended magnetic resonance imaging (MRI) for the evaluation of small-bowel disease. Fast imaging techniques coupled with advantages of breath holding improved MRI visualization of bezoars. Therefore, MRI is found to be better support for determining both the site and the cause of small-bowel obstructions. MRI shows the bezoar as a mass in the small bowel containing mottled and confluent low signal intensities on both T1- and T2-weighted MR images.[9]

The treatment consists of removing the mass by a single enterotomy or resection of the bowel if not feasible.[2,4] Duncan *et al*. recommended bezoar extraction by multiple enterotomies in the Rapunzel syndrome. DeBakey and Oschner reported a surgical mortality of 10.4%.[2,5] It is mandatory to perform a thorough exploration of all the small intestine and the stomach searching for retained bezoars.

The endoscopic examination is the preferred method of investigating the stomach when available; this allows searching for an associated bezoar while managing an intestinal bezoar. This exploration may reveal concomitant gastric bezoar which may be retrieved using endoscopy or gastrotomy approaches.[2] Escamilla *et al*. reported 23 cases of associated gastric bezoars extracted by gastrotomy over 87 cases of intestinal bezoars.[2] The psychiatric follow-up is essential to prevent recurrences.[1]

CONCLUSION

Trichobezoar is a rare clinical entity. Stomach is the common site of occurrence. The intestinal obstruction due to trichobezoar is extremely rare. Various imaging modalities have been recommended for revealing the pathology. The treatment consists of removing the mass using a single enterotomy.

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