Gastrointestinal trichobezoar causing atypical intussusception in a child: report of two cases

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Gastrointestinal trichobezoar causing atypical intussusception in a child: report of two cases

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Abstract:
Two cases of trichobezoar were reported in two girls aged 12 and 13 who developed intestinal intussusceptions secondary to incarceration of a trichobezoar in the intestinal lumen (gastroduodenal and ileal trichobezoars). The diagnosis was confirmed by sonography and tomodensitometry. Both patients underwent surgical extraction of the trichobezoar and intussusceptions reduction. The evolution was favorable in both cases.

Keywords: Trichobezoars, acute intussusception, surgical treatment
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Introduction
Trichobezoars are the result of the ingestion of indigestible substances in the gastrointestinal tract. This behavioral disorder is frequently noted among adolescents with mental disorders. The most common complication is an acute intestinal obstruction. The acute intestinal intussusception remains an occasional discovery rarely reported in the literature. We report two cases of acute intussusception secondary to gastroduodenal and ileal trichobezoars.

Case reports
Case 1
A 13-year-old female presented to the emergency room with periumbilical abdominal pain and vomiting lasting for 24 hours. The girl has a history of idiopathic thrombocytopenic purpura followed since the age of 2 years. The general examination was normal apart from a mucous cutaneous pallor. The abdominal examination allowed to palpate a fixed 10*12 cm, slightly sensitive periumbilical mass. The Laboratory tests showed an iron deficiency anemia in 6.4 g / dl and correct platelet levels. The abdomen x-ray detected a significant stercoral stasis without hydroaeric level.

An abdominal ultrasound was performed showing the presence of two small bowel intussusceptions, the first under the hepatic zone and the second at the left iliac fossa, measuring 6 and 4.5 cm respectively. Furthermore, there was a wall thickening in the intestinal without suffering signs.

The abdominal CT scan showed a gastric distension with an important heterogeneous content stasis: with a pseudo heterogeneous endoluminal mass poisoning the air with the presence of 3 intussusceptions: a 68 x 60 x 40 mm duodenojejunal, a 50 x 38 x 32 mm jejunojejunal and the last 105 x 60 x 55 mm ileoileal (Figure 1), respectively. Histopathology of biopsy from stomach and duodenum showed mucosal ulceration of mucosa with no inflammation. Baby
was discharged on day 10 of life on oral lansoprazole and breast feeding.

The diagnosis of intussusception secondary to gastroduodenal and ileal trichobezoar was retained. After a short preoperative preparation, filling and blood transfusion by packed cells, the patient was operated by laparotomy. The exploration noted the presence of three ileoileal intussusceptions (Figure 2).

The manual reduction of the intussusception was easy with good vitality of the bowel. The palpation of the small intestine showed the presence of a cord and hair balls (Figure 3 and 4).

Case 2
The second case is a 12-year-old female with an eating disorder for which she was followed from the age of 5 years with poor adherence. This girl presented to the emergency room in 2012 at the age of 10 for abdominal pain with vomiting. An upper gastrointestinal endoscopy was

Figure 1. Abdominal CT scan showing two tubes of ileo-ileal intussusception and heterogeneous gastric mass

Figure 2, 3 and 4. Ileo-ileal intussusception (2), palpation of an ileal hair cord (3), and palpation of a hair ball in the ileum (4).
performed revealing the presence of a large bezoar made of plant substances and hair extending from the upper part of the fundus to D2. The attempts to fragment and extract the foreign bodies endoscopically failed. The bezoar crashed at the cardia.

The patient was operated by laparotomy. She had an anterior gastrostomy that resulted in the extraction of a large hairball included in the stomach and reaching the duodenum. The follow ups were uneventful. The girl was referred to the child psychiatry consultation and not readmitted since.

She consulted again 3 years later for abdominal pain associated with a few episodes of vomiting. The patient interrogation revealed the persistence of the trichophagia.

The clinical examination was normal. The laboratory tests showed a 9.8 g/dl anemia deficiency.

The abdominal CT scan was performed showing the presence of two ileoileal intussusceptions measuring 15 mm and 20 mm with a thickening of the jejunal wall. The stomach and duodenum were distended by a dense heterogeneous mass.

The patient was operated by laparotomy. The exploration showed the presence of two ileoileal intussusceptions easily reduced. In addition there was a disparity between the dilated jejunum with an inflammatory thickened wall and the ileum which was of normal size (Figure 5). There was also a jejunal healed perforation (Figure 6).

An ileal solid content related to the hairball was palpated. The extraction of the latter and its extension in the small bowel (8 cm length) was made by an anterior gastrostomy (Figure 7).

The postoperative course was uneventful during the following 6 months.

**Discussion**

Trichobezoar is a rare disorder secondary to the accumulation of various substances within the gastrointestinal tract. Trichobezoars are usually confined to the stomach but rarely may dislodge and travel into the small intestine where they can produce obstruction and ileoileal intussusceptions. The most common sites of obstruction are the gastric outlet and duodenum. The Rapunzel syndrome occurs when gastrointestinal obstruction is produced by a trichobezoar with a long tail that extends to or beyond the ileocecal valve [1].

The clinical symptomatology is varied and non-specific abdominal pain, nausea and vomiting. But sometimes the diagnosis can be made in cases of complications [2].

Complications of intra small bowel trichobezoar are occlusions, jejunal perforation or acute pancreatitis secondary to obstruction of the ampulla of Vater by an extension of trichobezoar making Rapunzel syndrome [2, 3, 4].
The occurrence of the ileoileal intussusceptions is due to the mechanical bowel obstruction secondary to a long fixed trichobezoar and to the hyperactive peristalsis [4]. These intussusceptions are usually recorded as an incidental radiologic or preoperative finding.

The esophago-gastroduodenal endoscopy is the test of choice for confirming the diagnosis of intragastric trichobezoar. It enables viewing the tangled hair and it can have a therapeutic value for the endoscopic extraction of small trichobezoars [2, 5, 6].

Abdominal ultrasound allows the diagnosis in 25% of cases visualizing superficial hyperechoic curvilinear band with posterior shadowing. And in the case of ileoileal intussusceptions sonography shows doughnut or sandwich signs [5]. Computed Tomography has less importance in the diagnosis of trichobezoar. However it keeps a place in the diagnosis of small bowel-complication trichobezoar with good specificity and sensitivity [6].

Therapeutic management depends on the size of bezoar and the existence or absence of complications. The small bezoars can be eradicated by endoscopy. Surgical treatment is considered in case of gastrointestinal perforation, bleeding, very large bezoar not extractable by endoscopy, and in case the bezoar extended to the small intestine, as was the case of our patients. The aim is the extraction of gastric hairball by an anterior gastrostomy as well as the extraction of eventual extensions or fragments blocked away from the stomach through one or more enterotomies [6]. Recently laparoscopy has been proposed as an alternative to laparotomy [7].

Furthermore the psychiatric care is mandatory for those patients who suffer from trichophagia.

**Conclusion**

The bezoar is a rare condition, which requires multidisciplinary care to prevent complications and the risk of recurrence. It is a cause that must be taken into consideration in the case of diagnosing a grown up girl with intussusceptions.

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