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CASE REPORT

Bezoar - Case report of a preschool children with Rapunzel Syndrome

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Abstract

Case report of a 2 year and 11 months-old patient with giant bezoar extending to the duodenum. The case becomes interesting given the fact the patient's age, which on literature reports do not cite its occurrence in a so young patient. We discuss also options for diagnosis and treatment. The benzoar has occupied the distance between the stomach to the duodenum.

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INTRODUCTION

Historical records of bezoars in ruminants date to the pre-Christian era and are shrouded by folklore and religious beliefs[1-3]. In humans, bezoars have been described since the eighteenth century, and it is defined as the accumulation of undigested materials that form foreign bodies in the stomach and duodenum[1-3].

Trichobezoar is the bezoar most commonly found in pediatrics. It is usually associated with trichotillomania, which is a compulsion to pull out one's hair, and trichophagia, which is the habit of eating hair[4]. It is known that these disorders are common in pediatric patients and should be carefully addressed by health professionals because the latter may be the first professionals to have contact with these patientes[4].

This study reports the case of a preschool patient with bezoar composed primarily of hair.

CASE REPORT

A.L.S.V., a female patient aged 2 years and 11 months, lives in Araxá, state of Minas Gerais, Brazil, and was taken to the Pediatrics Urgent Care (PUC) at the Clinics Hospital of the Federal University of Triângulo Mineir, Uberaba, Minas Gerais, because of the presence of a large mass in the abdomen on ultrasound (USG) examination. The mother reported that her child presented with sporadic fever for nine days, with a peak temperature of 38.5°C, associated with vomiting containing mucus. The patient also presented with hyporexia and epigastric pain.

Ultrasound imaging showed an echogenic mass of 33 mm in diameter in the medial line above the umbilical scar, and abdominal X-ray revealed opacification of the gastric bubble and part of the transverse colon. Upper gastrointestinal endoscopy showed that the bezoar occupied much of the gastric body and fundus, reached the duodenum, and its removal was not possible.

Surgical intervention via laparotomy was required to remove the foreign body. This material was composed of hair, plastic, and food remains (Figure 1). Follow-up was done up to eight months after surgery, and the patient discontinued follow-up after this period.

COMMENTS

Bezoars were initially described in humans in 1779 by Baudamant and were found by chance during a necropsy. In 1883, the first surgery for the removal of a trichobezoar was performed by Schonborn[1-3,5].

Bezoars are rare and its incidence is higher in older populations. Phytobezoars are the most common type[1,3]. In pediatrics, the incidence of phytobezoars increases during adolescence, and their presence is usually associated with

Figure 1. Bezoar composed of hair, plastic, and food remains.

psychological disorders that lead to trichophagia, which makes trichobezoar the most common type of bezoar in this age group[2]. The bezoars that reach the stomach and extend to part of the duodenum are even more rare, and this unusual form, known as Rapunzel syndrome, was observed in the case reported above[2].

The symptoms may vary from mild abdominal discomfort to manifestations of obstructive syndrome, including vomiting, anorexia, and weight loss. The latter is common, and the severity of this condition varies according to the size of the clusters in the gastrointestinal tract. Other complications include perforations, intussusception, disabsortive anemia, and gastritis[1-3,5].

Diagnosis usually involves the correlation between symptoms and imaging tests. Phase contrast X-ray and ultrasound imaging of the abdomen can be useful in the diagnosis and are easily accessible[3]. Upper gastrointestinal endoscopy (UGIE) is able to visualize and identify the components of bezoar and even remove them depending on the case[3].

Treatment is classical, and a surgical approach is considered in cases in which bulky masses do not respond to initial therapy or significant complications may occur[1-3,5]. UGIE

can be used to remove small- to medium-size bezoars, depending on the degree of fragmentation[5]. Surgical procedures such as gastrostomy and enterotomy can be performed, and parts of the intestine may need to be resected depending on the complication[5].

After the removal of bezoars—particularly trichobezoars—trichotillomania and trichophagia need to be monitored. The patient should be monitored by a multidisciplinary team and, if necessary, medications should be introduced to control anxiety[4].

No previous studies have reported cases of patients with bezoar at such a young age. Despite their low incidence, it is important to consider the presence of bezoars when examining pediatric patients because the case presented here indicates that bezoars are not found simply in adolescents and adults.

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