Pediatr Pol 2019; 94 (6): 385–387
DOI: https://doi.org/10.5114/polp.2019.92955
Submitted: 25.10.2019; Accepted: 25.11.2019; Published: 31.12.2019

PEDIATRIA Polska
PISMO POLSKIEGO TOWARZYSTWA PEDIATRYCZNEGO
Polish Journal of PAEDIATRICS
OFFICIAL JOURNAL OF POLISH SOCIETY OF PAEDIATRICS

CASE REPORT

A giant bezoar in a child with cerebral palsy – could we prevent the disease?

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ABSTRACT

We present a case of giant bezoar in a 16-year-old boy with cerebral palsy. Endoscopy failed to remove the mass. Laparotomy revealed that the stomach, small intestine, and colon were full of cloth material. It was extracted by multiple enterotomies. A fifty-centimetre section of the small intestine was resected due to pressure necrosis. The bezoar mass in the stomach did not tightly fill its space. It was extracted by gastrotomy. Rising awareness among parents and healthcare professionals about the possible treatment of compulsive behaviours might prevent serious complications. We opt for laparotomy if oesophagoduodenoscopy fails to remove the bezoar entirely or in case of an unremarkable bezoar mass in the stomach associated with evident preoperative intestinal obstruction symptoms.

KEY WORDS:

intestinal obstruction, endoscopy, foreign bodies, compulsive behaviour, bezoars.

INTRODUCTION

There are fewer than 50 cases of Rapunzel syndrome reported in the literature [1–3]. The incidence of small bowel obstruction due to bezoars including food boluses is 0.3–6% [4]. Small bowel obstruction due to bezoar made of cotton threads is extremely rare [5]. In most cases, bezoars are present only in the stomach. Intestinal obstruction due to a bezoar in the intestine without a parent bezoar in the stomach is unusual. In case of trichobezoars the tail was reaching ileum, the cecum in some cases or transverse colon in one case.

CASE REPORT

A 16.5-year-old boy was referred to a paediatric surgical department because of an abdominal pain, vomiting, and constipation for three days. His medical history revealed cerebral palsy (CP) grade IV in Gross Motor Function Classification System (GMFCS), severe mental retardation (lack of logical contact), and aggressive behaviour. The deficits associated with CP included lower extremity weakness, developmental delay with gross and fine motor skills, and decreased flexibility. He presented little social interaction and had significant neuropsychomotor delay. He was nonverbal but did not have seizures. There had been observed uncontrolled ingestion of thread pieces for about four years. The patient's mother performed enemas and found some cotton threads in the loose stool. Palpable mass in the epigastric region and dehydration symptoms were found in the physical examination. Laboratory studies revealed moderately elevated white blood cell count ($18.99 \times 10^3/\mu l$) and C-reactive protein (67 mg/l), anaemia (haemoglobin level was 8.1 g/dl), hypokalaemia (3.4 mmol/l), and hyponatraemia (130 mmol/l). X-ray imaging with contrast revealed signs of intestinal obstruction as well as uncommon distribution of the contrast medium, possibly suggesting the pres-

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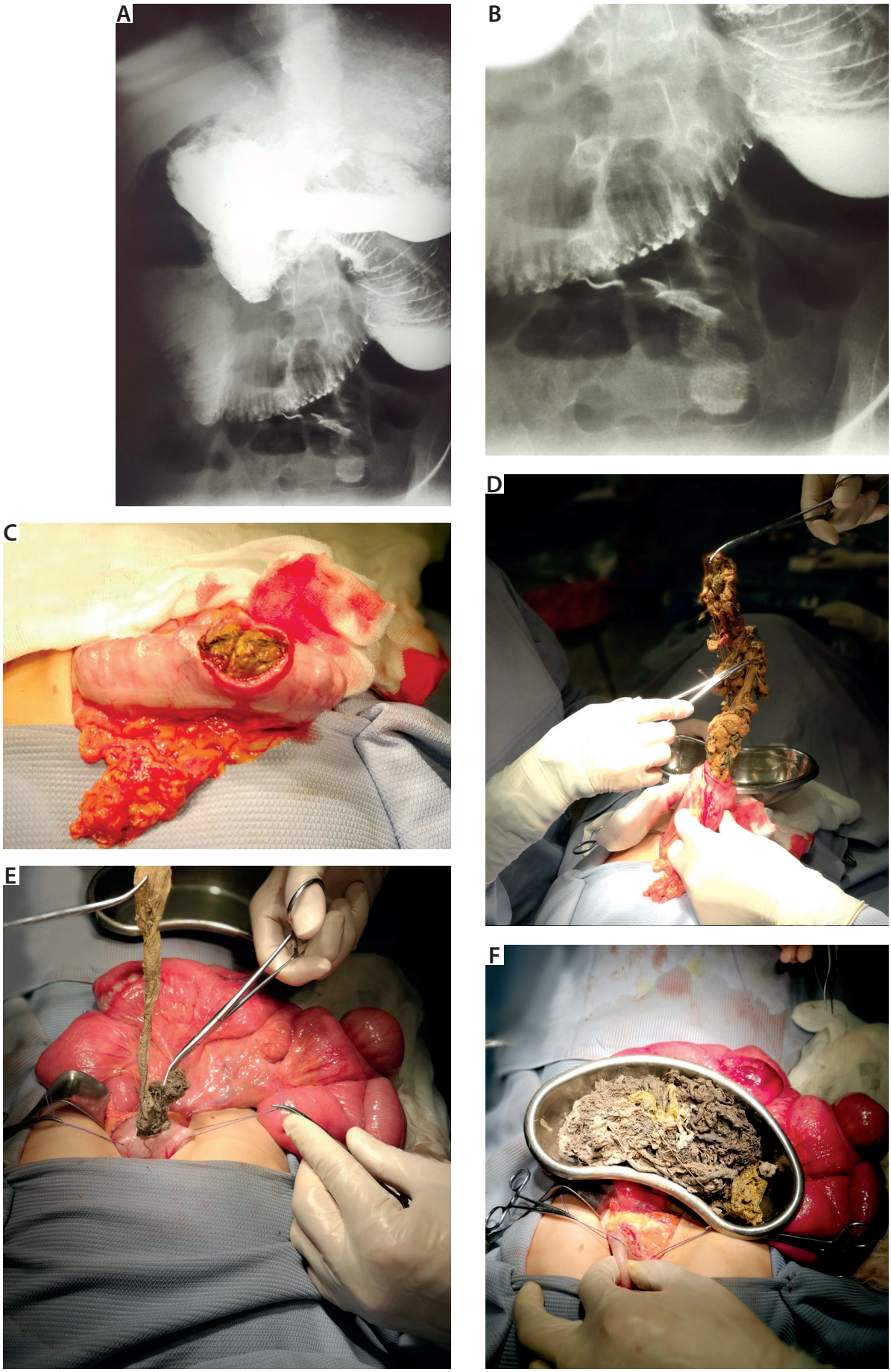

FIGURE 1. Diagnostic and intraoperative images. **A, B**) X-ray imaging with contrast, **C–F**) extraction of cloth material via enterotomies

ence of a foreign body (Fig. 1). The patient was qualified for an operative procedure. Esophagoscopy demonstrated masses of the fabric bezoar filling the lower part of the oesophagus. Laparotomy revealed that the stomach, small intestine, and colon were full of cloth material. It was extracted by enterotomies at the level of ileum, ascending, and transverse colon, respectively. The bezoar mass in the stomach did not tightly fill its space. It was extracted by gastrotomy (Fig. 1). A 50 cm section of the small intestine was resected due to pressure necrosis. Postoperatively, the patient was initially observed in the intensive care unit. By the 10th postoperative day a regular diet was tolerated; the wound healing process was uneventful. After psychiatric evaluation, pharmacotherapy with selective serotonin reuptake inhibitors was initiated, which resulted in a successful behavioural modification. There were no complications after eight months of follow-up.

DISCUSSION

In the literature, the described cases of cotton bezoars are limited to small and large intestine without affecting the stomach [1, 3, 4, 6]. Our case is thus an extremely rare variant of Rapunzel syndrome. The bezoar was made of cotton threads, it affected the intestine to the level of descending colon, and it was present in the stomach. However, the gastric part did not tightly fill the stomach to create a figure in its shape as is usually seen in the case of Rapunzel syndrome.

Oesophagoduodenoscopy is the investigation of choice for diagnostic confirmation. Endoscopic examination is also the preferred method of exploring the stomach for associated bezoar while managing a case of intestinal bezoar. Five per cent of gastric bezoars are multiple. On revealing a concomitant gastric bezoar, such an approach allows its endoscopic retrieval [4, 6]. In our case endoscopy failed to remove the mass. The ability to carefully examine the entire gastrointestinal tract for satellites in a short period of time and a high success rate are listed as the reasons for considering laparotomy as the treatment of choice in the case of multiple bezoars [4]. Alternative methods such as enzymatic degradation, pharmacotherapy, endoscopic fragmentation, and laparoscopy have been shown to be ineffective in many cases of Rapunzel syndrome [3].

Morbidity and mortality from intestinal obstruction are related to the diagnostic delays [7]. Pica is rarely volunteered by a patient or family. Although the parent was aware of the child's compulsive behaviour, the problem was not reported to the general physician. Good communication skills with the patient and his or her relatives are an essential point in eliciting a history of pica. Rapunzel syndrome is not a primary surgical condition, and a late relapse is possible [8, 9]. Long-term psychiatric follow-up supplemented with behavioural therapy is needed to avoid it. This cannot be achieved without a comprehensive home support network, with family or friends also monitoring treatment compliance at home. No currently approved treatment for pica renders selection of an appropriate therapeutic plan very difficult [3, 4]. In this regard, prevention of this condition is crucial.

CONCLUSIONS

Although the incidence of bezoars is low, it should always be kept in mind in cases of intestinal obstruction in young patients with psychiatric illness or mental retardation. Rising awareness among parents and healthcare professionals about the possible treatment of compulsive behaviours might prevent serious complications. We opt for a conversion to laparotomy if oesophagoduodenoscopy fails to remove the bezoar entirely or in the case of an unremarkable bezoar mass in the stomach associated with evident intestinal obstruction symptoms presented preoperatively. This therapeutic approach might be especially accurate in the case of cotton bezoars, which seem to affect intestine while omitting the stomach more often than trichobezoars.

DISCLOSURE

The authors declare no conflict of interest.

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