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Case report

A rare case of acute large bowel obstruction due to colonic bezoars associated with pica in a pediatric patient: A case report

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ABSTRACT

Introduction: Colonic bezoars are undigested or partially digested materials that accumulate in the form of a mass in the colon. There are occasional reports of constipation and intestinal obstruction due to geophagia or pica. Bezoars are due to pica, which is commonly associated with iron deficiency anemia and psychiatry disorders. Case presentation: A nine-year old female child presented with complaint of abdominal pain, vomiting and constipation for few months, and lately with features anemia and acute intestinal obstruction. She had history of pica. After evaluation she was treated surgically in two stages. In the first stage exploratory laparotomy with evacuation of lithobezoars with transverse colostomy was done, which was followed by colostomy reversal in the second stage. As a part of multidisciplinary treatment, psychiatric counseling and nutritional support were provided.

Discussion: Colonic lithobezoars presenting with features of chronic constipation and complicated with large bowel obstruction is a rare finding. Diagnostic imaging modalities such as X-ray and CT scan play an important role in confirming the diagnosis. Initial management involves conservative measures for uncomplicated cases with surgical intervention as a definitive option for complications like obstruction or perforation.

Conclusion: This case highlights the importance of considering pica with bezoars in children presenting with anemia and features of bowel obstruction. In such case multidisciplinary support and definitive medical and surgical management is done.

1. Introduction

The gastrointestinal bezoars are collections of undigested or partially digested materials that accumulate as a mass in the tract. It has a rare incidence of less than 1% in the general population. The bezoars are classified based on their composition, which includes trichobezoars (hair), phytobezoars (vegetables/fruit fibers), lactobezoars (milk protein), lithobezoars (stone or soil-like materials), etc. The cases of bezoars due to materials like cotton, seeds, medications, etc. can occur rarely [1]. Bezoars are usually asymptomatic and are frequently found in stomach. However, it can sometime migrate to and lodge within the small intestine or colon and can present with complications like intestinal obstruction or perforation [2]. The common symptoms include nausea, vomiting, abdominal discomfort or pain, and abdominal distention [3]. Though rare, colonic bezoars present with symptoms such as constipation, painful bowel movements, straining during defecation, feeding difficulties and weight loss [4]. Lithobezoars are frequently associated with pica, especially in children. Pica has been reported to be associated with iron deficiency anemia in up to half of patients and also with mental retardation, autism and behavioral disorders [5]. Colonic lithobezoars presenting with obstructive symptoms is uncommon. The management ranges from a multidisciplinary approach initially with the conservative approach to surgical interventions [2]. We present a case of a female child with habit of pica presented with the features of chronic constipation complicated with acute intestinal obstruction. This case report has been reported in line with the SCARE 2023 criteria [7].

2. Case presentation

A nine-year-old female presented with the complaint of abdominal discomfort, with occasional dull aching pain on the lower part, and abdominal distention on and off for the last three months. She had history of few episodes of non-projectile watery vomiting with low stool volume occurring over one week. She was referred from a local center to our tertiary center. At presentation in our center, she had not passed

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stool for 48 h and had progressive increase in the severity of her abdominal pain, distension as well as the frequency of vomiting. Her family was unaware of her habit of eating soil, which she had been doing for over a year. She had good appetite despite her weight loss. There was no known medical or psychiatric comorbidity. On physical examination, she appeared thin-built with body mass index (BMI) of 14 kg/m² and had pallor with mild dehydration. Vitals showed mild tachypnea and mild tachycardia. Per abdominal examination demonstrated moderately distended abdomen with more fullness and tenderness over the lower part of her abdomen, and a palpable hard mass could be felt at left iliac fossa and hypogastric region. Per rectal examination showed a slightly spacious rectum but soft to firm mass could be felt, and blackish stool was smeared on the gloved finger.

Her initial laboratory reports obtained from the other center revealed hemoglobin (Hb) level 5.1 g/dl, total leukocyte count (TLC) $6300/\text{mm}^3$ and platelets count $187,000/\text{mm}^3$. Two units of packed red blood cell (PRBC) and one unit of whole blood (WB) were transfused before the referral. Investigations done at our center showed Hb level 10.3 g/dl, TLC $3900/\text{mm}^3$, platelets count $122,000/\text{mm}^3$, hematocrit 33.4%, mean corpuscular volume (MCV) 76.8, mean corpuscular hemoglobin (MCH) 23.6, mean corpuscular hemoglobin concentration (MCHC) 30.8 and red cell distribution width (RDW) 22. The iron profile showed serum iron level 59 µg/dl, total iron binding capacity (TIBC) 494 µg/dl, transferrin saturation 12.06%, and serum ferritin level 29.5 ng/ml with no typical findings of iron deficiency anemia (IDA). Peripheral blood smear (PBS) findings showed slight microcytic picture with no pencil or tear drop cells.

The abdominal radiograph showed distended proximal large bowel with radiopaque shadows in distended descending and sigmoid colons (Fig. 1). Contrast enhanced computed tomography (CECT) of abdomen revealed grossly dilated sigmoid colon with intraluminal filling of large amount of heterogeneous contents with mottled appearance secondary to mass trapped within the dilated segment expanding over a length of 28 cm with maximum width measuring 14 cm; the intraluminal fecal contents in the distal-most part of the involved segment showed whorled appearance, proximal and distal ends of the involved segment showing patent lumen with no evidence of narrowing; no evidence of extraluminal mass lesions or volvulus, multiple mildly enlarged mesenteric lymph nodes noted largest one measuring 11×7.5 mm, likely reactive. This suggested features of acute colonic obstruction with lithobezoars (Fig. 2).

After the evaluation, provisional diagnosis of partial large bowel obstruction secondary to colonic bezoars with pica was made. Patient was initially admitted to monitoring ward and managed conservatively. Patient was kept nil per os (NPO) and glycerin enema was given. Blood transfusion was performed. However, her condition deteriorated with gradual increased pain and distension over 24 h; the nasogastric tube placed in-situ had drained around 400 ml of bilious putrid collection. These clinical features and CT imaging findings were evaluated and emergency surgery was planned. Exploratory laparotomy with evacuation of rectosigmoid bezoars with diversion transverse colostomy was performed. The operative findings include large amount of rectosigmoid fecaloma (lithobezoars) with megacolon, grossly dilated descending and sigmoid colon, multiple mesenteric lymphadenopathy (Figs. 3,4). Colostomy was planned after informed consent because of her poor nutritional status, severe anemia, and grossly distended colon. Serial biopsy taken from rectosigmoid junction and sigmoid colon revealed presence of ganglion cells; mesenteric lymph node biopsy showed



Fig. 1. Abdominal X-ray, Erect and Supine Views: showing grossly distended proximal large bowel [green arrow] and opaque intraluminal mass (lithobezoars) within descending and sigmoid colon [yellow arrow]. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

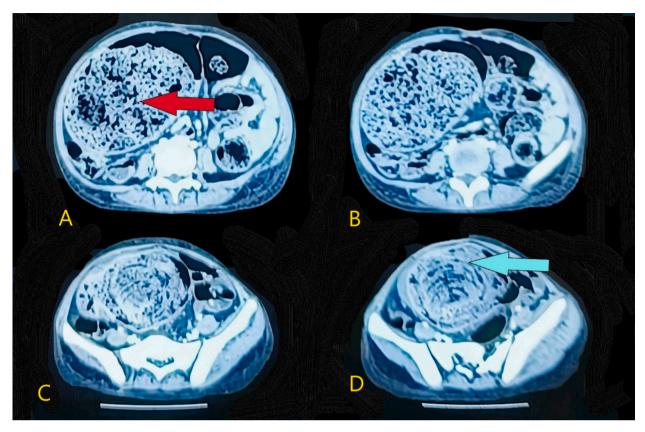


Fig. 2. Computed Tomography (CT) scan of abdomen, serial axial images(A-D) showing large intraluminal mass (lithobezoar) within grossly distended sigmoid colon extending to the right side of abdomen [red arrow] and intraluminal mass (lithobezoar) within the rectum [blue arrow]. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

reactive follicular hyperplasia; and colostomy site biopsy showed colitis with presence of ganglion cells. The postoperative stay was uneventful. She was monitored in Surgical Intensive care unit (SICU) for 2 days and later in general ward. She was allowed a liquid-to-soft diet by the second post-operative day (POD). Colostomy was functional after POD1. Psychiatric consultation and counseling was done to the patient and her family regarding her eating habit. Nutritional consultation was also done involving the dietary department. Work up for anemia was continued. Colostomy care was taught well before she was discharged on the sixth POD. Her follow up visit was uneventful with gradual weight gain and correction of anemia. Review psychiatric evaluation showed improvement of her eating habit. Pica was completely resolved. Colostomy reversal was performed after 2 months following the appropriate weight gain and correction of anemia. The length of hospital stay on her second surgery was five days. The postoperative period was uneventful. She was then kept in follow-up for the next visit after 3 months.

3. Discussion

Lithobezoars are the uncommon cause of chronic constipation and intestinal obstruction. Colonic lithobezoars presenting with obstructive symptom is a rare finding and only few cases are reported in medical literature [5]. It is a rare disorder causing mechanical intestinal obstruction in the pediatric age group [5,7]. Colonic lithobezoars are mostly associated with history of pica. Pica occurs most commonly in children, in patients with learning disabilities and in pregnancy. The etiology is poorly understood [8]. The American Psychiatrist Association, Diagnostic and Statistical Manual of Mental Disorders, fifth Edition (DSM -V) defines pica as eating nonnutritive non-food substance over a period of at least one month. The individual must be at least two years of age and should be inappropriate to the developmental level, without

being socially normative, and excluding culturally acceptable behavior [9]. Risk factors for developing pica include stress, nutritional deficiency, pregnancy, low socioeconomic status, child neglect, epilepsy, underlying mental disorder, or familial psychopathology [10]. There are different forms of pica that include eating hair (trichophagia), ice (pagophagia), starch and talcum powder (amylophagia), as well as eating soil and clay (geophagia). The prevalence of geophagia is difficult to estimate because patients only seek medical care in case of severe symptoms or complications. The potential complications include parasitic and fungal infections, metabolic abnormalities, heavy metal poisoning, electrolyte and other elemental deficiencies [11]. Pica is associated to iron deficiency, zinc deficiency, mental retardation and developmental problems [12]. When associated with iron deficiency, it is believed to be a symptom of the deficiency rather than its cause [11].

Clinically, children with pica often present with signs and symptoms of mechanical bowel obstruction. A palpable abdominal mass is occasionally found. On rectal examination, the presence of the "colonic crunch sign" increases the suspicion of obstruction due to bezoar. Hard material like stone can be palpated and removed during rectal examination. Radio-imaging studies are the mainstay for the diagnosis in the early stages. A plain abdominal X-ray demonstrates various-sized opacities, the sign referred to as "corn on the cub" [13]. In intestinal obstruction, a CT scan is the most effective method to determine the accurate treatment modality along the detection of co-existing multiple bezoars, level of obstruction, intestinal ischemia, perforation, and other potential intestinal diseases. The most frequently seen CT findings of bezoars include the appearance of a round or ovoid or a long sausageshaped mass containing mottled gas at the obstructed site [14]. Kuang et al. reported that the bezoars causing bowel obstruction were frequently phytobezoar type, with larger volume measured on CT images i.e. the major diameter of the bezoar was a possible risk factor [15].



Fig. 3. Intraoperative image: showing large distended mobile sigmoid colon with evacuation of lithobezoar through an enterotomy site (covered with gauze piece).

Contrast enhanced CT scan has a sensitivity of 90% and a specificity of 57% in recognizing bezoars, which is considered the gold standard in the diagnosis of bezoars and bowel obstruction. The CT scan is valuable in terms of differential diagnosis and helps to decide for a conservative or a surgical/endoscopic treatment [16]. If prompt diagnosis and timely treatment are not carried out, the condition may lead to significant morbidity and mortality [17].

The treatment of colonic bezoars depends on the type, localization and size of geophagia ingested and the clinical presentation of patient. In cases of small-size bezoars, it can be managed conservatively with laxatives and enemas. In contrast, large-size bezoars can be removed by anal dilatation and rectal wash under GA, either digital or endoscopic removal as reported by Senol M et al. [6]. However, in delayed presentation or when conservative management fails, surgical intervention is required to relieve obstruction or manage perforation [11]. The endoscopic treatment for bezoars is gaining popularity because of the increased success rate, however, surgical intervention is necessary when there is a failure of nonsurgical treatment [2]. Conventional or laparoscopic surgical approaches can be performed. The most frequently employed procedure is to remove the lithobezoars with enterotomy, sometimes requiring resection and anastomosis of bowel [17]. Therefore lithobezoars should be considered in the evaluation of young children displaying pain abdomen or indications of bowel obstruction.

In our case study, the patient had a long history of pica or geophagia and presented with chronic constipation and anemia, complicated with the features of partial large bowel obstruction. She was initially managed at the local health center a few times before, where symptomatic treatment was only done for her abdominal discomfort and anemia, and the history of pica was not disclosed before the presentation

to our center. The patient's history of eating soil with chronic constipation had contributed to the gradual formation of the lithobezoars leading to intraluminal obstruction of colon. Though she was anemic, her Iron profile did not show typical feature of iron deficiency anemia (IDA). This scenario may be due to her post-transfusion status. The actual cause of anemia was under further evaluation. With initial attempt of conservative management and its failure, we managed the case surgically: exploratory laparotomy with evacuation of colonic bezoars with transverse colostomy, owing to the delayed presentation with features of bowel obstruction. Additionally child psychiatric counseling and nutrition consultation was done for the behavioral problem of pica and malnutrition respectively. This highlights that multidisciplinary approach is crucial for the management of bezoars with pica.

4. Conclusions

Colonic lithobezoars are rare conditions that should be excluded in children who present with features of chronic constipation and/or intestinal obstruction. The features of anemia, weight loss, abdominal mass, under-nutrition and parental concern should prompt early evaluation. The management of colonic bezoars includes a multidisciplinary approach with nutritional support, psychiatric evaluation and counseling, and definitive medical and/or surgical intervention. Moreover adequate follow up is important for good outcome of treatment and prevent recurrence.

Author contribution

Sushil Gyawali and Priyanka Sah: Study concept, design, acquisition,

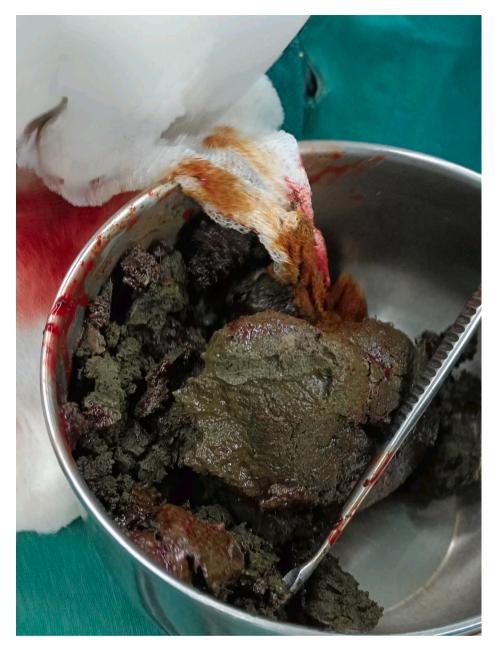


Fig. 4. Intraoperative image: showing the evacuated content (intraluminal lithobezoars) in a container.

interpretation of the data, writing the draft, review and editing, validation.

Mridul Prasad Joshi and Anuj Kayastha: Acquisition, interpretation of the data, critical review for important intellectual content, validation.

Ramana Rajkarnikar: concept, review critically for important intellectual content, validation, supervision of the work.

All the authors provide final approval of the version to be published and are accountable for all aspects of the work ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Consent

Written informed consent was obtained from the patient's parents/ legal guardian for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This is the case report; and doesn't require an ethical approval for the case report in our institution, hence was not obtained.

Guarantor

Sushil Gyawali.

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