

Trichobezoar: An Unusual Cause for Pancreatitis in a Patient With Sickle Cell Anemia

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Pica is defined as the compulsive ingestion of particular food items or substances not fit as food or of no nutritional value. Organic causes of pica include iron deficiency, lead encephalopathy, pregnancy, and zinc deficiency.¹ Patients with pica have been shown to have decreased iron and zinc absorption compared with control subjects; subsequent zinc supplementation results in pica elimination.² The most common forms of pica include geophagia (ingestion of clay, sand, and dirt), pagophagia (ice), trichophagia (hair), and amylophagia (laundry starch).³ Pica occurs frequently in children with sickle cell disease, with one study reporting a prevalence of almost 34%.¹

A potentially devastating complication of pica is the development of a gastric bezoar, with consequent gastrointestinal sequelae such as gastric ulceration, bleeding and perforation, intussusception, and small bowel obstruction.⁴ Pancreatitis has been reported rarely. Trichobezoar, which results from the ingestion of hair or carpet fibers, is the most common bezoar in the pediatric age group.⁵ Patients commonly present with recurrent abdominal pain and a palpable abdominal mass.

Case Report

An 11-year-old African American boy presented with a 2- to 3-week history of abdominal pain, which became acutely worse on the day of hospital admission, when he also suffered a single episode of emesis. Pain was accentuated

by sitting, and for the previous 2–3 days had been accompanied by decreased oral intake and decreased frequency of bowel movements. The patient denied shortness of breath, fever, or diarrhea. Review of systems uncovered a 2.6-kg weight loss but was otherwise noncontributory, and he did not complain of symptoms related to sickle cell anemia. His past medical history was significant for hemoglobin SC disease, which had necessitated a single hospitalization for acute chest syndrome.

The patient's psychosocial history was significant for severe physical abuse and neglect as an infant, placement in multiple foster homes, and finally adoption at age 8. He had been known to eat hair and carpet since he was a toddler and had received outpatient counseling for poorly characterized "behavior problems" at home and school, but no specific interventions had targeted his hair pulling and eating.

On physical examination, the patient was noted to be cachectic, weighing 22 kg (less than the third percentile for the patient's age group). The patient had left upper quadrant tenderness, and a firm mass was palpable in the left epigastric area. Laboratory studies on admission demonstrated an elevated pancreatic amylase of 1,173 units (normal, 30–140 units), an elevated pancreatic lipase of 1,111 units (normal, 13–51 units), and an elevated bilirubin of 1.2 mg/dL (normal, 0.2–0.9 mg/dL). Nine days later, his serum zinc level measured 36 µg/dL (normal, 66–144 µg/dL).

Abdominal computed tomography (CT) (Figure 1) demonstrated an enlarged stomach containing a whorled mass composed of air admixed with moderately dense material, extending through the duodenum and into the proximal jejunum, consistent with a bezoar. The proximal small bowel wall appeared edematous. The pancreatic

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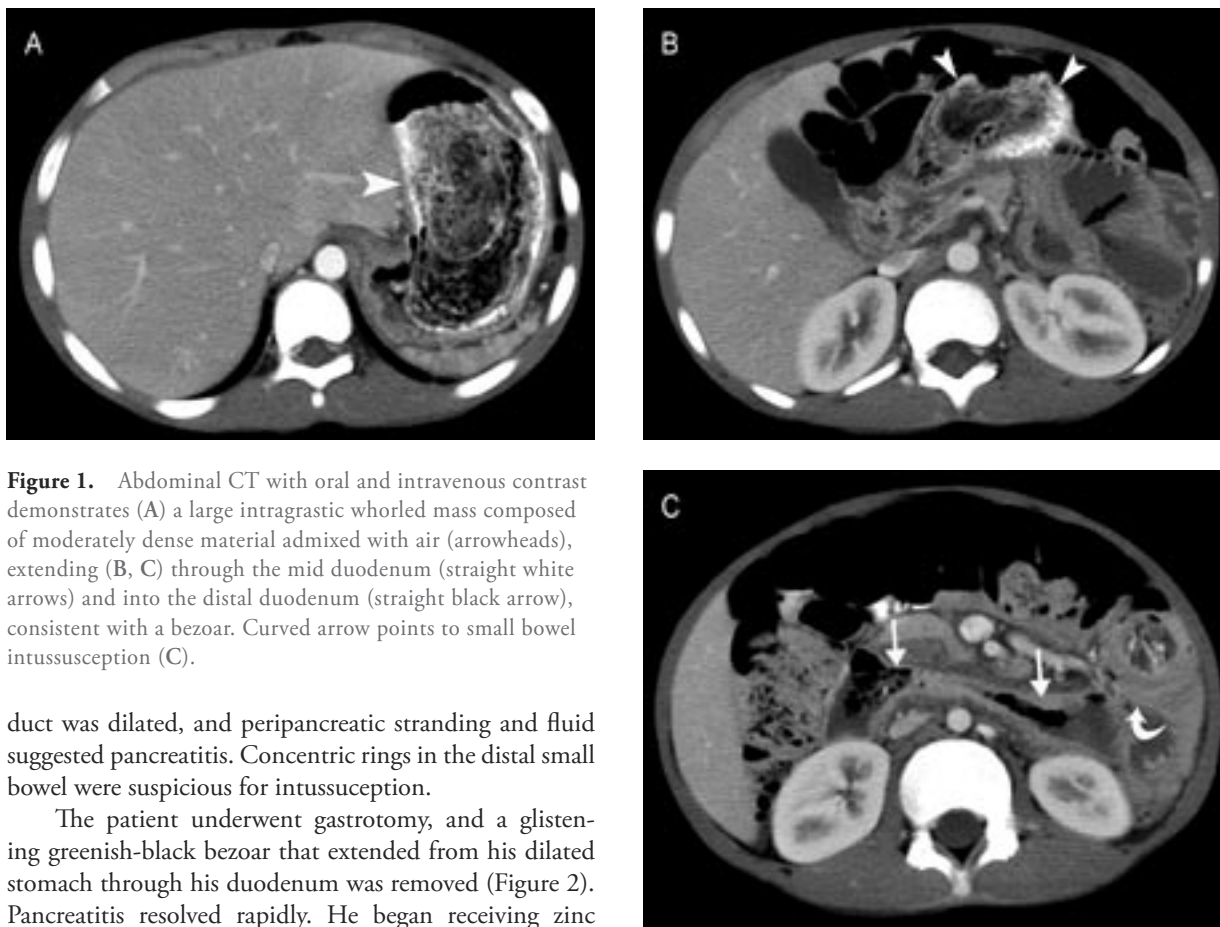


Figure 1. Abdominal CT with oral and intravenous contrast demonstrates (A) a large intragastric whorled mass composed of moderately dense material admixed with air (arrowheads), extending (B, C) through the mid duodenum (straight white arrows) and into the distal duodenum (straight black arrow), consistent with a bezoar. Curved arrow points to small bowel intussusception (C).

duct was dilated, and peripancreatic stranding and fluid suggested pancreatitis. Concentric rings in the distal small bowel were suspicious for intussusception.

The patient underwent gastrotomy, and a glistening greenish-black bezoar that extended from his dilated stomach through his duodenum was removed (Figure 2). Pancreatitis resolved rapidly. He began receiving zinc supplementation as well as psychotherapy for trichophagia. His hospital course was uneventful, and continued psychotherapy along with further zinc supplementation was recommended at discharge. A serum zinc level in the normal range was measured approximately 3 months after hospitalization and the pica had resolved.

Discussion

Pica is strikingly common but relatively poorly recognized in patients with sickle cell disease. Constituting persistent or compulsive ingestion of substances that have no nutritional content, it is often seen in small children and in pregnant women. It has been linked to both iron⁷ and zinc^{2,6,8} deficiencies, as well as to psychological disturbance.⁹ The term pica is derived from the Latin word for magpie, a bird known for its nondiscriminating appetite. A wide variety of substances are consumed, ranging from socks, newspapers, and sofa pillows to dirt and hair.

Pica has a very high prevalence in patients with sickle cell disease, occurring in 35% of those with hemoglobin SS and in 25% of those with hemoglobin SC.¹ It manifests at an older age in patients with sickle cell disease than in the general population. Some have found an association between pica and increased number of hospitalizations,³

suggesting an association with severity of disease, but this finding is not universal.¹

Patients with sickle cell disease who suffer from pica do measure at a significantly lower percentile for body height and weight for age, and their mean hemoglobin levels tend to be lower as well.¹ However, red blood cell indices do not suggest iron deficiency, as they are not hypochromic. These findings suggest that nutritional deficiency is more common in the group of sickle cell patients with pica. In the sickle cell population, zinc deficiency has been observed, probably resulting from the hyperzincuria that occurs due to repeated sickling, renal damage, and consequent decreased renal tubule zinc resorption.¹⁰ More than 50% of patients with sickle cell disease report abnormal eating patterns, and, of these, approximately 50% report pica.³ Although pica is found more often in older children than in the general population, among patients with sickle cell disease it is more common in younger age groups.³

Psychosocial stress may also contribute to the prevalence of pica in this patient population. Pica is theorized to represent an attempt at self-soothing for a patient population subject to repeated pain crises and hospitalizations.³ It can be postulated that the need for such a



Figure 2. Surgical specimen demonstrates a glistening greenish-black bezoar forming a cast of the stomach, with a typical tail constituting duodenal extension of the mass.

coping strategy would be even greater for a child whose developmental history also includes physical abuse and neglect. Reports in the literature suggest that in addition to correction of any measurable vitamin or mineral deficiency, psychotherapy may be helpful in decreasing pica behavior and has been successful at reducing hair-pulling compulsions like those of this patient. Careful evaluation for comorbid psychiatric disorders is warranted, and adjunctive psychopharmacologic treatment may be indicated in some cases.⁹

Bezoars are an unusual complication of pica, but when they occur they can be devastating, resulting in a 30% mortality rate if untreated. Although they arise in the stomach, trichobezoars, as in this patient, often extend into the duodenum and even into the small bowel. Fragments may separate from the body of the mass and migrate, resulting in more distal small bowel obstruction. Other common complications include gastric ulceration, bleeding, perforation, and intussusception.⁵ However, bezoars cause pancreatitis only rarely, with five cases reported in the literature. One involved a 59-year-old man, with a phytobezoar resulting from persimmon ingestion.¹¹ The other four were in young women, aged 15–29 years, three of whom suffered from trichophagia^{12–14}; the other ate athletic tube socks.¹⁵ This is the first reported case of pancreatitis complicating bezoar in a patient with sickle cell disease.

Postulated etiologies for pancreatitis include irritation from the adjacent bezoar causing edematous obstruction at the ampulla of Vater¹³ and bacterial overgrowth in the duodenum ascending the pancreatic duct.¹²

Readily diagnosed by ultrasound or computed tomography,¹⁶ bezoars can also be recognized at endoscopy. Trichobezoars of sufficient size require gastrotomy and removal, with possible additional incisions in the more distal bowel. As in this patient, pancreatitis resolves readily with removal of the offending object.^{11–14}

It is important to recognize bezoar, and its associated complications, as a possible etiology of abdominal pain in the patient with sickle cell anemia because people in this population group, which has a high incidence of pica, are at increased risk for development of this disorder. Some patients with sickle cell anemia will have splenomegaly, confounding the diagnosis. Their propensity to suffer from the often-underlying pica results from a variety of factors, including nutritional deficits (especially zinc deficiency) and intense psychosocial stresses. Although most patients with abdominal complications from bezoar will have the more common findings of gastrointestinal bleeding, obstruction, ulceration, and intussusception, bezoars are also an unusual cause of pancreatitis and should be considered in the sickle cell patient with pancreatitis.

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Review

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The sickling disorders are well known to have abdominal complications. The differential diagnosis for abdominal pain in this condition includes myriad possibilities that often present a diagnostic dilemma to the clinician.

Certainly, the most frequent etiology of abdominal pain in sickle cell disease is uncomplicated vaso-occlusion in intra-abdominal organs or musculoskeletal structures of the trunk, including the vertebral column. Referred pain from pulmonary or chest complications is also an important diagnostic consideration. Gallstones are well described in children with this disorder,¹ and though frequently asymptomatic, they can cause chronic pain. Patients can also present with acute pain if they develop cholecystitis. Gallstone pancreatitis is another common problem that presents with acute abdominal pain. Abdominal pain is often aggravated by severe constipation from the use of narcotics to relieve severe vaso-occlusive pain.

Pica is quite prevalent in children with sickle cell disease²⁻⁴ and has been associated with neuropsychological abnormalities. Pica is seen equally in males and females, and is generally more prevalent in younger children. Pica is common in tropical countries where cultural factors and nutrition may play a role.⁵ As stated by Stein-Wexler and colleagues,⁶ pica is classically associated with iron-deficiency anemia, lead encephalopathy, and zinc deficiency. The range of substances ingested is varied and includes paper, foam, clothing, and carpet fibers. In children with sickle cell anemia, pica is seen to be the most prevalent in the middle childhood years,² unlike in the general population. Iron deficiency does not appear to play a causal role in the pica of sickle cell disease, as these children do not exhibit microcytosis or hypochromia, reflecting adequate iron reutilization from their chronic hemolytic anemia.

Zinc deficiency occurs in various disease states such as malabsorption syndromes, hepatic cirrhosis, and chronic renal disease, as well as in subjects receiving total paren-

teral nutrition without zinc or following penicillamine therapy for Wilson disease and diabetes.⁵ The effects of decreased levels of zinc include delayed growth, hypogonadism, problems with dark adaptation, and immune deficiency.² Pica has been described in association with zinc deficiency.⁵ With the known occurrence of zinc deficiency in sickle cell disease, it is tempting to consider zinc deficiency as an etiology for the pica in this condition, but this particular association has not yet been published.

Stein-Wexler and colleagues⁶ describe trichobezoar as an additional and interesting cause of abdominal complications in children with sickle cell anemia. The case described is an 11-year-old boy with hemoglobin SC disease presenting with abdominal pain, constipation, weight loss, and a firm mass in the left upper quadrant of the abdomen. Laboratory investigations revealed acute pancreatitis, and imaging studies showed the presence of a whorled mass in the stomach resulting in pancreatic duct dilation. Management included surgical intervention, which resulted in the resolution of the pancreatitis, psychotherapy, and zinc supplementation.

This case successfully demonstrates the multifactorial etiology of pica, with the complex interaction of psychological stressors (psychosocial history as described in the case report) and malnutrition. Zinc deficiency may have aggravated the symptoms of pica in this child.

The case gives clinicians an additional reason to consider abdominal imaging for chronic or persistent abdominal pain, particularly when it is associated with pica. A seemingly "nonmedical" history of pica, which may be overlooked in patients presenting with pain, can lead to a potentially serious complication, as in this case description. Although gallstones are still the most common cause of pancreatitis in these children, trichobezoars should be considered in the differential diagnosis. This case also suggests the need for aggressive psychological intervention in older children with a significant history of pica to prevent the occurrence of this complication.

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