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Gastrointestinal trichobezoars presenting as abdominal pain and obstruction - Rapunzel syndrome

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Abstract

Presentation

A 14-year-old female presented to the emergency department with a five-day history of abdominal pain and vomiting. Amylase was raised on admission and she was managed conservatively for presumed acute pancreatitis. Her condition did not improve and she deteriorated a number of days into admission and developed bilious vomiting**.**

Diagnosis

CT abdomen-pelvis showed trichobezoars in the stomach and duodenum.

Treatment

She underwent emergent laparotomy and manual removal of bezoars in a tertiary centre followed by multi-disciplinary input post-operatively.

Discussion

Atypical abdominal pain in the paediatric population not responding to initial treatment warrants review of diagnosis and further investigation with consideration for early tertiary service input.

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Introduction

Bezoar is a rare entity, and the vast majority of published literature on the topic is from case reports. We present the case of a teenage girl presenting with abdominal pain progressing to intermittent intestinal obstruction with bilious vomiting as presentation of gastric and duodenal trichobezoar.

Case Report

A 14-year-old female presented to the emergency department with a five-day history of right upper quadrant pain and vomiting. There was no significant medical history, no previous hospitalisations or regular medications. She was developmentally appropriate for age. She was seen by the surgical service and acute abdomen was excluded and was referred to the paediatric service for further assessment. On examination of the abdomen, she was markedly tender in the right upper quadrant.

Initial laboratory investigations revealed elevated amylase (248), with no other abnormalities. She had severe abdominal pain requiring both regular simple and opioid analgesia. PFA appeared normal. On abdominal ultrasound the pancreas was inflamed and there was free fluid in the pelvis. Management of presumed acute pancreatitis (AP) was discussed with the gastroenterology service with advice to treat with intravenous fluids, analgesia and diet as tolerated.

Inflammatory markers rose on day two of admission and she was commenced on broad spectrum intravenous antibiotics to cover for infection secondary to AP. On day three of admission, she had multiple episodes of bilious vomiting accompanied by worsening abdominal pain. She was reviewed by the surgical team and was made NPO, commenced on intravenous antiemetics and PPI, and nasogastric tube was inserted on free drainage. Given lack of response to the presumed appropriate therapy and clinical deterioration, she underwent CT abdomen and pelvis. This showed bezoars within the stomach and duodenum. Her case was discussed with a tertiary paediatric surgery service and she was transferred for further management.

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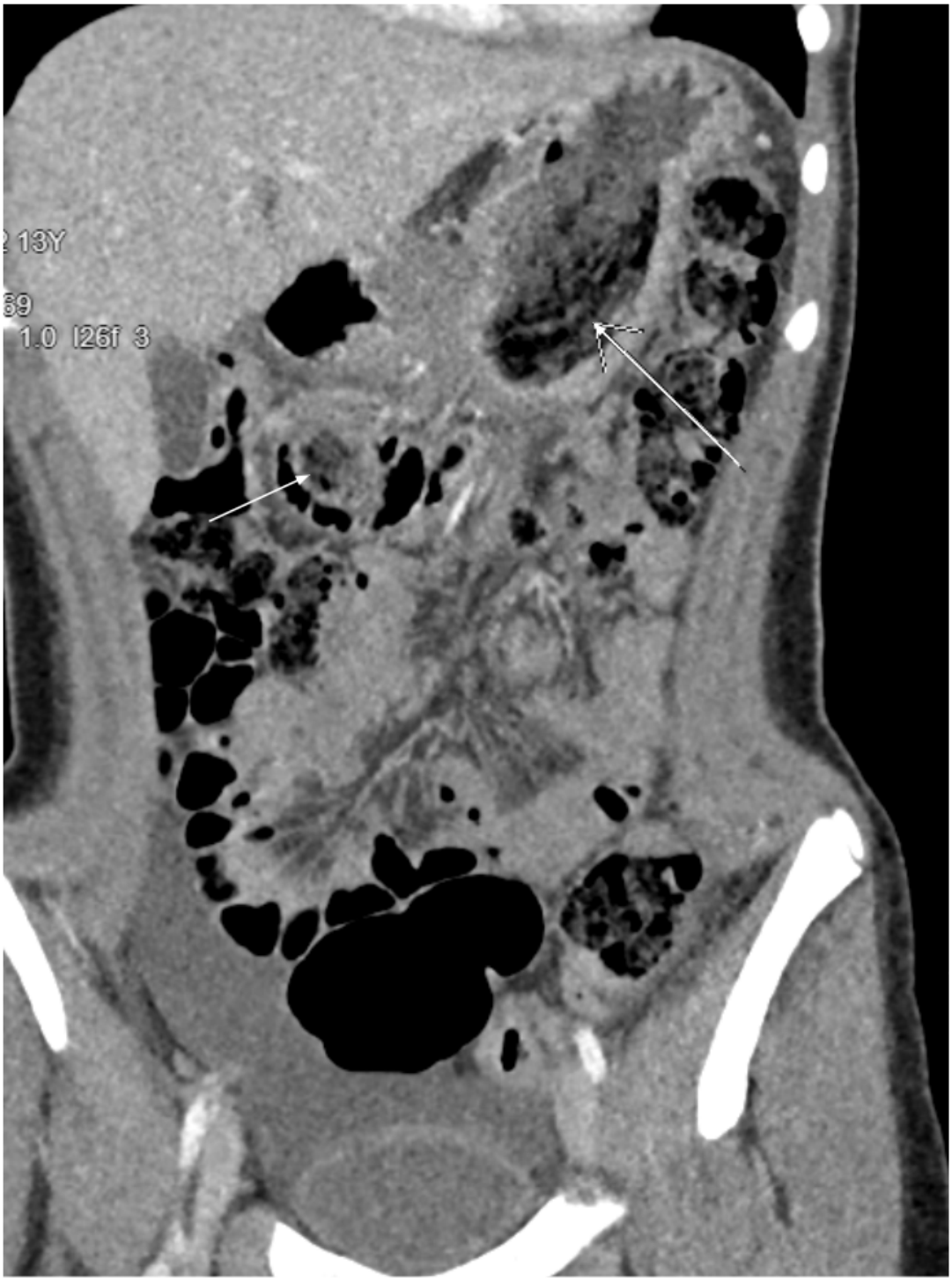

Figure 1: *White arrows indicate bezoar in stomach and 3rd part of duodenum on CT Cor MPR. Image courtesy of Dr Manana Pienaar.*

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She revealed a distant history of trichotillomania and trichophagia. She underwent emergent exploratory laparotomy. The stomach was opened and one bezoar was removed, the jejunum was opened and the second bezoar was removed. Post-operatively pain was managed by PCA, nutrition delivered via total parenteral nutrition and intravenous antibiotics and fluids were continued.

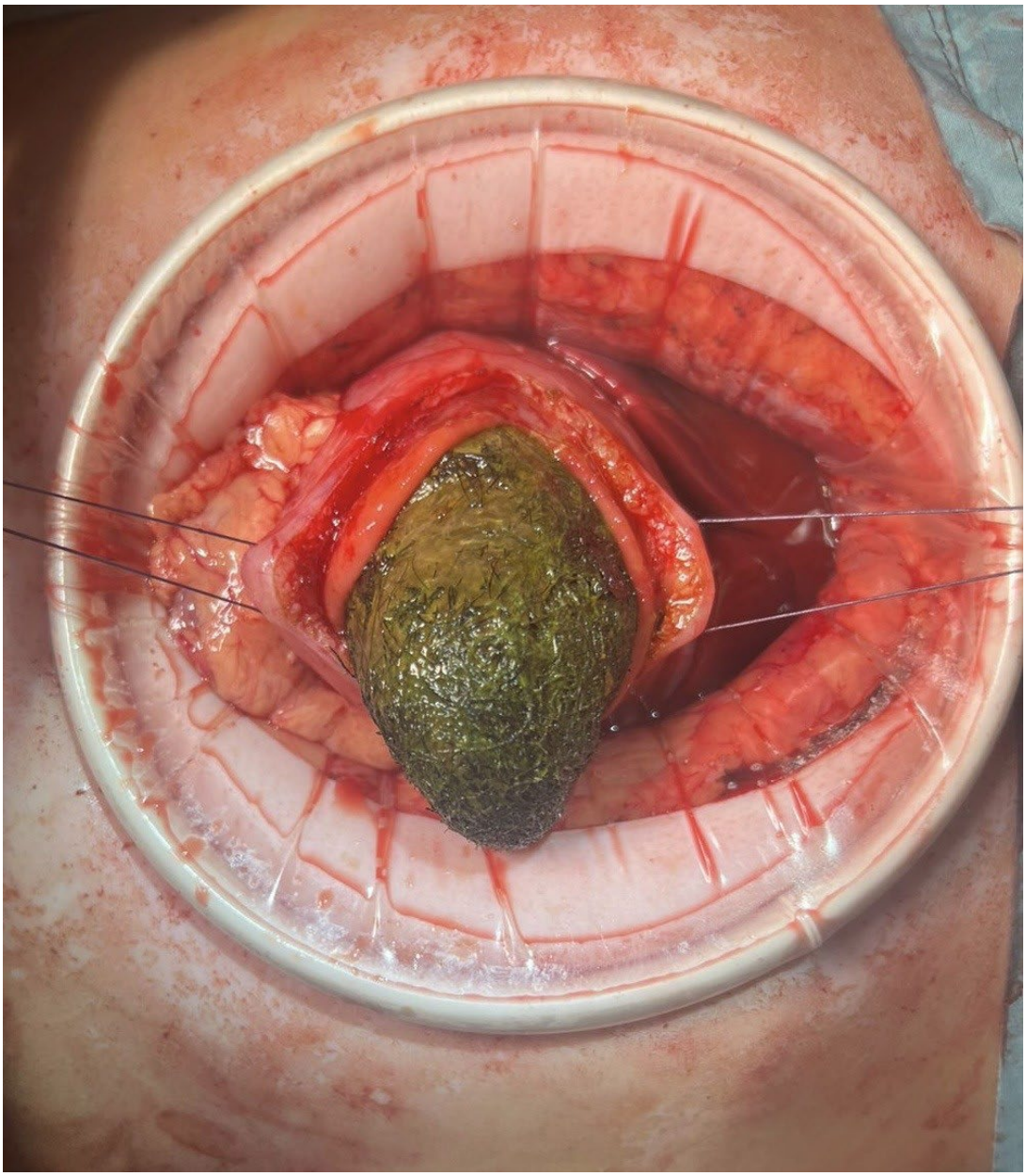

***Figure 2**: Intra-operative findings of bezoar in stomach during laparotomy. Image courtesy of Prof Israel Fernandez-Pineda.*

During her 8-day inpatient stay she received multidisciplinary input and had a normal video fluoroscopy. Psychology review was declined. She was discharged home on oral PPIs with surgical outpatient review.

Discussion

Abdominal pain is a common and heterogenous paediatric presenting complaint. We present a rare case. Trichobezoar is the most frequently encountered bezoar and is associated with trichotillomania and trichophagia.[3] 90% of bezoars are found in adolescent girls.[3] Trichobezoar can remain asymptomatic for years until it increases in size to the point of obstruction.[4] 'Rapunzel Syndrome' is characterised by the extension of a trichobezoar from the stomach to the intestine or beyond.[3-7] Abdominal pain, vomiting and nausea are common presenting symptoms.[3-5]

AP is defined as at least two of; abdominal pain, serum amylase +/- lipase three times the ULN and imaging consistent with the condition.[8] On abdominal US the pancreas appears diffusely enlarged and hypoechoic.[9] Our patient fulfilled these criteria. She was commenced on appropriate management as per BSPGHAN[8] guided by gastroenterology. Patients who are not improving after 48-72 hours should undergo CT or MR imaging to assess for complications.[10] CT outruled AP and revealed an alternative diagnosis. There are cases in the literature of bezoar obstructing the ampulla of Vater or compressing the common bile duct leading to pancreatitis. [4-7]

In children with abdominal pain unresponsive to initial treatment or with clinical deterioration, it is of value to consider further imaging early to delineate the cause.

Declarations of Conflicts of Interest:
None declared.

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