

Childhood Bezoars: A Case Series

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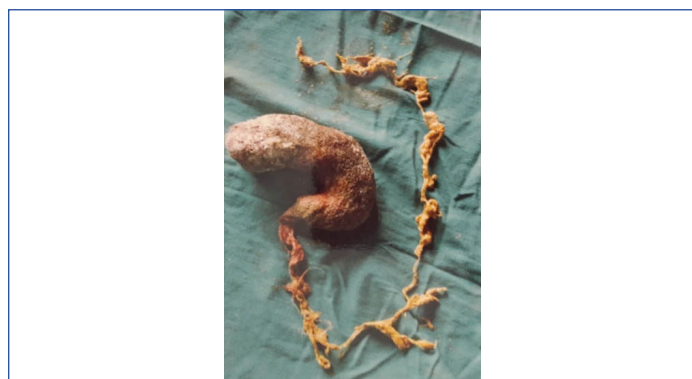
ABSTRACT

Bezoars are the rarest causes of upper abdominal masses and upper gastric outlet obstruction in children and are most commonly seen in females of adolescent age. They can present with the common symptoms of abdominal mass or abdominal pain but may lead to serious complications as well. They must be diagnosed and managed early despite their rarity so as to prevent complications like obstruction, perforation and bleeding. This article is reporting our 25 years experience with abdominal bezoars with variable presentation in our tertiary care centre.

Bezoars can be classified according to their components into phytobezoars (vegetable matter), trichobezoars (hair), and pharmacobezoar (medications), lactobezoars (milk). Trichobezoars are usually associated with trichotillomania, characterised by compulsive pulling of hair followed by eating of the same (trichophagia). This condition was first described by Baudamant in 1779 [1]. They are mostly found in stomach, but can also extend into the duodenum and jejunum, a condition known as Rapunzel syndrome, the rarest type of bezoar. This was first described in the literature by Vaughan ED et al., [2].

CASE SERIES

During the study period of 25 years from 1991 to 2016, we had 5 gastric bezoars, 3 intestinal bezoars and 3 rapunzels [Table/Fig-1]. These are described in [Table/Fig-2]. All intestinal bezoars were present in males. Two of them were trichobezoars and one was a phytobezoar. One child with intestinal trichobezoar presented with intestinal perforation and peritonitis, expired in the post-operative period due to sepsis and disseminated intravascular coagulopathy. The most common mode of presentation of gastric bezoars was abdominal lump with upper gastric outlet obstruction, while the intestinal bezoars presented with acute intestinal obstruction. All of them presented with the common symptoms of weight loss, loss of appetite and history of trichotillomania and trichophagia, loss of scalp hair since last 3-4 years. The symptoms were associated with psychiatric illness characterised by depression and anxiety only in 2 out of 11 cases. The diagnosis was made with the help of X-ray abdomen, abdominal ultrasound and Computerized Tomography (CT) scan of abdomen to confirm the diagnosis [Table/Fig-3]. Barium meal follow through was done in cases of lower gastrointestinal obstruction.



[Table/Fig-1]: Rapunzel causing ileal obstruction in a four-year-old male child.

Keywords: Phytobezoar, Rapunzel syndrome, Trichobezoar

All the cases were managed with exploratory laparotomy. The gastric bezoars were removed through gastrotomy [Table/Fig-4]. The intestinal bezoars were removed by enterotomy and resection and end to end anastomosis, as there was associated perforation in one case and unhealthy looking gut in others. Feeds were started on post-operative day 3 in most of the cases after the return of bowel sounds and passage of stools. All the cases were referred for psychiatry follow-up to avoid further recurrence.

DISCUSSION

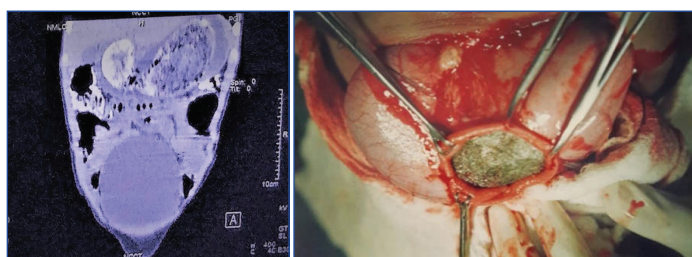
The origin of the word 'Bezoar' stems from a Persian word 'Badzehr' with literal meaning of 'antidote'. It was regarded as a stone capable of curing any kind of poison, which was later found to be a misconception. It was called a stony concretion in late 15th century, found in the gastrointestinal tracts of certain animals. The hair in the stomach escapes digestion and gets entrapped in the gastric mucosal folds, escaping peristalsis as well. They get coated by gastric mucus and get denatured by gastric acid resulting in their black and shiny appearance. The continued ingestion of hair leads to their accumulation in the stomach, with the bezoar assuming the oblong shape of the stomach [3]. Due to large capacity of the stomach, it takes long time of about 3-4 years for the bezoar to assume sufficiently large size to present as abdominal lump or obstruction. The complications include gastric outlet obstruction [4], gastric ulcer and gastric perforation [5], intestinal obstruction, peritonitis [6,7], intussusception, pancreatitis, obstructive jaundice/ cholangitis and death [8].

They may extend further to a variable extent into the jejunum, resulting in Rapunzel syndrome. There, the tip may get adhered to the jejunal wall at solitary or multiple sites, or it may lie free. They may primarily form in the intestine or get detached from the gastric bezoar to lodge into the intestine and cause obstructive symptoms [2]. The clinical manifestations depend on the site and size of the bezoar.

Contrary to the available literature, with females below 20 years of age forming 90% of the cases with trichobezoars [9], the occurrence of bezoars in males was around 27% in our institute. But all of them were intestinal bezoars. The psychiatric abnormality was present in only two out of the 11 cases. Trichobezoars are commonly associated with history of trichotillomania and trichophagia, but few cases have been described without the association with trichotillomania [10]. Fatality has been reported in 3 out of 49 cases of Rapunzel in a review by Kim SC et al., [11]. The diagnosis can be made with the help of ultrasonography or CT scan of abdomen. Findings on ultrasound include hyperechoic band-like lesions and

Patient	Age (years)	Gender	Presenting complaints	Type of bezoar	Extent	Surgical procedure	Postoperative complications	Psychiatric manifestations
1	4	M	Intestinal perforation, peritonitis, severe wasting	Trichobezoar	Intestinal	Enterotomy, gut resection, end to end anastomosis	Expired	--
2	6	M	Lower GI obstruction, mass, weight loss	Trichobezoar	Intestinal	Enterotomy, resection and end to end anastomosis	none	Absent
3	7	M	Lower GI obstruction, mass, weight loss	Phytobezoar	Intestinal	Enterotomy, resection and end to end anastomosis	none	Absent
4	10	F	Abdominal lump, weight loss, trichotillomania, trichophagia	Trichobezoar	gastric	Gastrotomy	none	Absent
5	14	F	Abdominal lump, Upper GI obstruction, weight loss, trichotillomania, trichophagia	Trichobezoar	Gastric	Gastrotomy	None	Absent
6	13	F	Abdominal lump, Upper GI obstruction, weight loss, trichophagia	Trichobezoar	gastric	gastrotomy	none	Anxiety, depression
7	14	F	Abdominal lump, weight loss, loss of scalp hair	Trichobezoar	Gastric	Gastrotomy	none	Absent
8	16	F	Abdominal lump, vomiting, upper GI obstruction, loss of scalp hair	Trichobezoar	Gastric	gastrotomy	none	Anxiety, depression
9	15	F	Abdominal lump, weight loss, trichotillomania, trichophagia	Trichobezoar	rapunzel	Enterotomy	None	Absent
10	14	F	Abdominal lump, weight loss, Upper GI obstruction	Trichobezoar	rapunzel	Enterotomy	None	Absent
11	15	F	Pain abdomen, abdominal lump weight loss, upper GI obstruction	Trichobezoar	rapunzel	Enterotomy	None	Absent

[Table/Fig-2]: Summary of 11 cases of bezoars.



[Table/Fig-3]: CECT abdomen: non-adherent spindle shaped mass in the stomach, with the contrast surrounding, coating and infiltrating trichobezoar with extension into duodenum.

[Table/Fig-4]: Pre-operative photograph of large gastric bezoar.

CT scan shows intraluminal mass with bubbly gaseous lucencies. Abdominal radiographs after barium contrast may be required in some cases. They demonstrate a filling defect outlining the bezoar with air entrapped in it. The diagnosis can also be confirmed by endoscopy which can also be used for removal of the mass if it is small.

The management of bezoars depends on the site and the size of the bezoar. For large gastric bezoars and Rapunzel syndrome, treatment of choice is surgical removal through gastrotomy. In Rapunzel syndrome, if the tail is free, only gastrotomy is indicated. But, if the tail is adherent to jejunum at solitary or multiple sites, then solitary or multiple jejunotomies are required. The tail should be pulled gently; otherwise forceful extraction may result in jejunal perforation [7]. For small bezoars, endoscopic or laparoscopic removal may be tried. Coca cola has also been used to fragment the phytobezoars, which can then be removed endoscopically [12]. The mechanisms proposed are mucolytic effects of sodium bicarbonate, and the acidifying effects of carbonic and phosphoric acid present in diet coke. Dissolution of trichobezoars with papain, cellulose or acetylcysteine has largely been unsuccessful [13]. A literature review by Al-Janabi IS et al., found laparotomy as the best and most effective methods for management of the bezoars [14]. The laparoscopy and endoscopy being reported as unsuccessful in most of the case reports, while laparotomy followed by gastrotomy was associated with a success rate of around 99% [8,10,15]. But, the advantage of endoscopy and laparoscopic removal is that it is non-invasive, while laparotomy with surgical removal is associated with various complications, like wound infection.

The further management is necessary in order to avoid further recurrences. Upto 20% of the patients can have recurrent bezoars [16]. This can be minimized by prompt psychiatric referral and follow-up. We referred all our patients for psychiatry follow-up.

CONCLUSION

A high index of suspicion is necessary for diagnosis. Trichobezoars should be suspected in the cases presenting with weight loss, abdominal pain or abdominal lump. The best method of management is laparotomy or gastrotomy. Recurrences can be minimized by appropriate psychiatry follow-up.

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