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Trichobezoar with Rapunzel Syndrome Secondary to Trichophagia in Old Age a Case Report

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ABSTRACT

Bezoars are impacted concretions of ingested non-absorbable foreign material, formed most often in the stomach and sometimes in the intestine. When the bezoar is composed of hairs it is termed a trichobezoar and when its tail is extended into the small intestine and even up to the colon, it is called Rapunzel syndrome because of its long tail, like the tresses of the fictional character it is named after. It is usually associated with prior mental illnesses such as trichophagia and trichotillomania. Treatment includes surgical removal of the trichobezoar along with psychiatric management of the associated mental illness to prevent recurrence. It is common in young teenage females, but we present a rare case of a 71 years old adult female. She presented with chief complaints of epigastric pain, dysphagia and vomiting. All laboratory investigations turned out normal but physical examination revealed a hard-movable mass in the abdomen. The patient was reluctant to reveal her habit of eating hairs, which hindered the diagnosis. Endoscopy was done which confirmed presence of a trichobezoar, tethered to the mucosa of stomach, which made it difficult to remove the bezoar with forceps. She underwent open abdominal surgery and the trichobezoar with its tail was removed. The patient recovered and is undergoing psychiatric treatment for trichophagia.

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Introduction

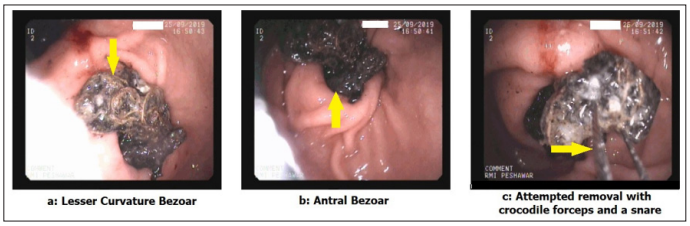
The name bezoar is said to be derived from the Persian word "Padjahar" meaning an antidote, as evident from ancient accounts of its occurrence in animals and men for centuries, they were collected and used as medicinal charms after presentation. Bezoars are impacted concretions of ingested non-absorbable foreign material, formed most often in the stomach and sometimes in the intestine [1]. They are mostly asymptomatic but with the passage of time they grow bigger in size and develop into a dense mass, because of their non-absorbable nature in gastrointestinal tract, causing a series of gastrointestinal symptoms [2].

The structural composition of bezoars determines the etiology and the type they are classified into [3]. There have been multiple case reports describing bezoars of various compositions which can be classified into phytobezoars (composed of vegetables or fruit fibers), trichobezoars (balls of hair or hair-like fibers, also known as Rapunzel syndrome), diospyrobezoars (of persimmon), pharmacobezoar (of pills), lactobezoars (of milk and curd), lithobezoars (fragments of stones) or plasticobezoars (plastic) [4]. Trichobezoars, which are usually associated with a psychiatric disorder viz trichophagia (habitual ingestion of hair) and trichotillomania (practice of pulling hair), are composed of balls of hair or hair like fibers when ingested they get trapped in the stomach mucosa and do not propagate with peristalsis. It was first described by Baudomant in 1779 [5]. When its tail is extended into the small intestine and even up to the colon, then it is termed Rapunzel syndrome [6]. Rapunzel, is the name of a maiden in a fairytale written by Grimm Brothers, had long hairs in tresses and was locked away in a tower by her witch step mother, the only way to climb that tower was with the help of her hairs [7]. This syndrome was first reported in 1968 by Vaughan et al [8]. Since then, less than 50 cases have been reported so far. In adults phytobezoar is the most commonly occurring type of bezoar, while trichobezoar usually occurs in children and young individuals aged 10 to 19, associated with a psychiatric illness like trichophagia or trichotillomania [9]. In some case reports, patients as young as three years have also been reported [10]. On the other hand a 27 year old female was found to have a trichobezoar with Rapunzel syndrome [4]. The case under discussion might be the first case ever reported in old age. Patients with trichobezoar usually present with symptoms of early satiety, nausea, vomiting,

loss of appetite, weight loss and abdominal pain. Some cases with features of intestinal obstruction, obstructive jaundice, perforation with peritonitis, pancreatitis and even death are also reported [11-15]. Diagnosis is generally based on ample history, clinical examination, endoscopic and radiological investigation. It should be noted that when taking the clinical history, the patient and the parents of the patient may hesitate to reveal the coexisting psychiatric illness, which leads to delayed diagnosis and other complications like obstruction, pancreatitis, perforation, and peritonitis. Treatment involves extraction of the bezoar either by upper gastrointestinal endoscopy, which is both diagnostic and therapeutic, laparoscopic or open surgery and if there is any coexisting psychiatric illness, this should also be treated with proper psychiatric consult [15,16].

Case Report

A 60-year-old female patient belonging to the indigenous population of northern Pakistan province of Gilgit Baltistan presented to us in outpatient department. She worked as a housewife with no other professional work history. There was no history of past interventions or surgeries, no significant co morbidities in her family or cases of psychiatric illnesses. Her chief complaints upon presentation were epigastric pain, dysphagia and vomiting for 10 days. laboratory investigations were all within normal limits viz; Hb 14 g/dl (12-16), WBC $5.6x10^3$/microl (4-11), Platelets $217x10^9$/L (150-450), Alk 114U/L (30-120), Alt 29U/l (5-35), Na 140mmol/l (135-148), K 4mmol/l (3.6-5.2). On physical examination a hard-movable mass was detected in the right upper quadrant of the abdomen. Along with this we found alarming features specific to intestinal obstruction which included her age i.e., 71 years, new onset dysphagia and post prandial vomiting which were suggestive of probable intestinal obstruction due to a bezoar. Keeping in mind her alarming features we suspected a case of bezoar and again asked her specifically about a habit of swallowing hairs or any other foreign body; she revealed that she'd had this habit of swallowing her hairs (trichophagia) for the last six months, which she had concealed until now, including her initial history. We opted for oesophago-gastro-duodenoscopy or upper gastrointestinal endoscopy as it is the gold standard for diagnosis of bezoars, which showed a normal esophagus and two large balls of hairs (Trichobezoars) at the lesser curvature and D1 regions, but further visualization of the stomach was hampered by the bezoars, (Figure 1). The removal of both the bezoars was attempted with crocodile forceps and a snare but the bezoars were tethered to the mucosa of the stomach (Figure1c). To investigate further we did computed tomography of the abdomen to confirm the extent and length of the trichobezoar. Computed tomography confirmed the trichobezoar with a tail extending beyond the ligament of Treitz; in other words, she was diagnosed with a trichobezoar with Rapunzel syndrome. On clinical assessment the trichobezoar was classified as grade 1, which requires gastrotomy or enterotomy. Surgical consult was obtained, and surgical removal of the bezoar was planned. The patient was transferred to the surgical department for surgical removal of the bezoar under general anesthesia by a surgical team led by consultant general surgeon with thirty-five years of experience in the field. Laparotomy was performed with a scalpel by giving upper midline incision, the trichobezoar location was identified with inflamed gut and gastrotomy with extraction of both the green colored trichobezoars with tail was done. The patient was closed with vicryl sutures, and the surgery finished uneventfully. The patient was transferred to the surgical intensive care unit in stable condition for 24 hours observation and was kept on intravenous antibiotics. She was then transferred to surgical ward and got discharged with home medications comprising of multivitamins, antibiotics, and home instructions for wound care. On follow up the patient recovered with no post-op complications. She is now under psychiatric treatment for Trichophagia.



a: Lesser Curvature Bezoar b: Antral Bezoar c: Attempted removal with crocodile forceps and a snare

Discussion

Phytobezoars are the most common type of occurring bezoar but in young individuals it is trichobezoar [1]. The disease is more prevalent in females in approximately 90% of cases with age group 13 to 19 years. Trichobezoars in old individuals are rare, there is no such case ever reported so far and we believe that our case might be the first case ever reported. Trichobezoars are mostly associated with prior psychiatric illness like trichotillomania and trichophagia in mentally disturbed individuals, that is why psychiatric treatment is important to avoid recurrence of the illness. As in our case the diagnosis was delayed due to two reasons; first being the communication gap with the patient as she was from the northern region of our country and the language there is less comprehensible in the rest of the country and secondly the patient was hesitant to reveal her habit of trichophagia which required extra counselling to obtain proper history. We believe history should be ample in such cases of trichobezoars. Trichobezoar is asymptomatic initially but in due course when a significant mass is formed occluding the lumen of the gastrointestinal tract causing symptoms depending on the location it is formed and the extent it has grown into. Symptoms usually include abdominal pain, nausea, vomiting, decreased appetite, early satiety, and that of intestinal obstruction. If left untreated it can lead to transient pancreatitis, ulceration, perforation, and peritonitis. Although our patient was diagnosed late, but we had enough time to initiate prompt treatment and prevented late complications. Other common complications include hematemesis, anemia, gastric mucosal erosion, gastric outlet obstruction, intussusception, obstructive jaundice, protein-losing enteropathy, acute pancreatitis, and death [11-15]. Endoscopy is the gold standard when it comes it diagnosing a bezoar, our endoscopy facility is the state of the art facility in the province but due the size of the bezoar we were unable to visualize full length of the trichobezoar or try to extract it with simple endoscopic snare. Trichobezoar's shiny glistening surface is because of the coating by the mucus secreted by the stomach, the acid secreted in the stomach denatures the protein making the trichobezoar appear black in color. Patients with trichobezoar has a rotten odor in breath, which is due to the fermentation and decomposition, of the entrapped food particles and fat, carried out in the gastrointestinal tract. Cases with bezoars found distally in the gastrointestinal tract have also been reported due to dislodgement and distal propulsion. Treatment is usually surgical removal of the bezoar through open abdominal surgery, other options including laparoscopic removal and endoscopic removal of the bezoar have also been used in some cases. Treatment of the associated psychiatric illness is important for the cure and to prevent recurrence of the disease [16].

Conclusion

Trichobezoar can lead to fatal complication if not diagnosed on time, it can also rarely occur in old individuals, treatment should include surgical removal of the trichobezoar with psychiatric treatment of the associated mental illness, like trichophagia, to prevent recurrence.

Data Availability
All data underlying the results are available as part of the article and no additional source data are required.

Consent
Written informed consent for publication of their clinical details and clinical images was obtained from the patient.

Grant Information
The authors declared that no grants were involved in supporting this work.

Competing Interests
Head of department is also the author of the manuscript and member of the ethics committee of the institute.

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