



Case Report

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Gastric Trichobezoar without trichotillomania: A diagnostic dilemma

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Abstract

Trichobezoar (hairs ball in stomach) is an uncommon clinical entity. Plucking of hairs Trichotillomania and swallowing (trichophagia) of hairs are two essential conditions needed in development of trichobezoar. It is very rare to suffer with trichobezoar in the absence of trichotillomania. In this report we present two cases of trichobezoar associated with trichophagia in the absence of trichotillomania. A 32-years-old female mother of three children suffered with trichobezoar without trichotillomania as she developed taste and urge for hairs and stated eating hairs thrown by other ladies in the family. An 11-year-old girl presented with trichobezoar. Examination of scalp and other body parts did not show any evidence of pulling of hair/short hair as she was eating hairs thrown by others as she developed a taste and strong desire to eat hairs. To conclude, these two cases highlight that patients can have trichobezoar and trichophagia even in the absence of trichotillomania and can pose diagnostic dilemma for physician.

Keywords: Trichobezoar, Trichophagia, Trichotillomania.

INTRODUCTION

Bezoars can be formed from undigested materials. Trichobezoar is formed as a result of hair ingestion either self-hair or from others and is an uncommon disorder in humans. It is mostly seen in young females and was first reported in 1779 by Baudamant^[1]. The word is a combination of "trich" and "bezoar", with the former meaning hair in Greek and the latter meaning poison antidote in Arabic or Persian^[2]. Plucking of hairs (trichotillomania) and swallowing of hairs (trichophagia) are essential conditions in formation of trichobezoar. Trichobezoars are associated with trichophagia (compulsive eating of hair) which may be due to trichotillomania or due to pica –an eating disorder manifested by an appetite for nonnutritive substances like soil, dirt, hairballs, ice, paint and sand, etc. However, trichophagia and trichobezoars are rarely described in the absence of trichotillomania^[3,4]. Here we report two cases of trichobezoar associated with trichophagia but without trichotillomania.

CASES REPORT

1st Case

A 32-years-old female mother of three children presented with pain in abdomen with vomiting for the last 6 months. On clinical examination, there was a big 14x12 cm lump in epigastric area which was moving with respiration. Blood investigations like hemoglobin level, complete blood counts, serum electrolytes, liver function test, renal function test, pancreatic enzymes and routine stool examination were within normal limits. Ultrasound was done for initial diagnosis and it revealed trichobezoar. Barium study and upper GI endoscopy [Fig-1, Fig-2] were done to confirm the diagnosis which revealed gastric trichobezoar. As examination of scalp and other body parts did not show any evidence of pulling of hair/short hair, so, psychiatry analysis was done. Detailed history revealed that the patient used to do abnormal actions and would behave as some evil soul has entered in her body. Tantric used to feed her hairs mixed with ash for curative purpose to remove evil soul from her body. Subsequently, she developed uncontrollable taste and urge for hairs and started eating hairs thrown by other ladies in the family. However, this patient denied pulling of her own hairs.

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Fig 1: Barium study of 1st showing space occupying lesion ? trichobezoar



Fig 3: Ultrasound of 2nd case showing ? trichobezoar



Fig 2: Endoscopy picture of 1st case showing trichobezoar

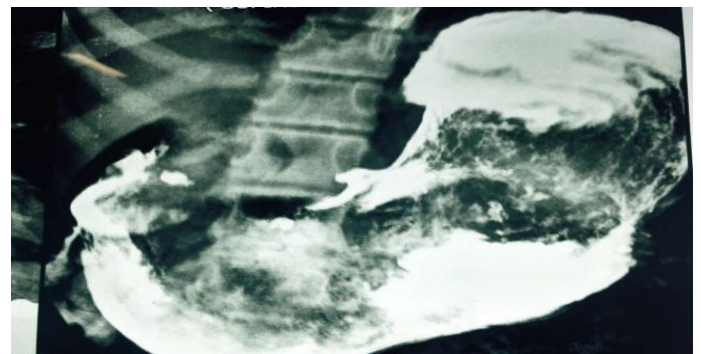


Fig 4: Barium study of 2nd case showing space occupying lesion ? trichobezoar

2nd Case

An 11-year-old girl presented with complaints of pain in abdomen and vomiting off and on for the last 9 months. Physical examination revealed a 10 × 8 cm lump in epigastrium firm which was mobile with respiration. Blood investigations like hemoglobin level, complete blood counts, serum electrolytes, liver function test, renal function test, pancreatic enzymes and routine stool examination were within normal limits. Ultrasound abdomen revealed a bizarre lesion in the right upper abdomen suggestive of gastric bezoars [Fig-3]. In view of trichobezoar as clinical diagnosis, barium study and upper gastrointestinal endoscopic were carried out to confirm the diagnosis [Fig-4, Fig-5]. As examination of scalp and other body parts did not show any evidence of pulling of hair/short hair, so, psychiatry consultation was sought. Detailed history revealed that the patient was eating hairs and clay since early childhood. Due to taste and strong urge to eat hairs, she would go out in search of thrown hairs. There was family history of mental illness in her mother. The patient was illiterate and was from a poor background and was suffering with signs of anxiety neurosis. She was emotionally labile. Under stressful condition she would have urge to eat hair and clay. She was considered to be a case of "Pica of infancy and childhood".



Fig 5: Endoscopy picture 2nd case showing trichobezoar

Laparotomy was carried out to treat both these patients [Fig-6] and trichobezoars were removed [Fig-7a, 7b]. Counseling was done in postoperative period and both the patients were told about the harmful consequences of eating hairs and clays and were encouraged to leave this bad habit. Patients were taught to tackle stressful conditions and to manage urge and impulse. Family members were also educated about the illness and were asked to be vigilant to note

the patient behavior of eating hairs and clay. The patients were improved with the surgery and by counseling. Follow-up at 3 and 6 months did not reveal any behavior suggestive of trichophagia.

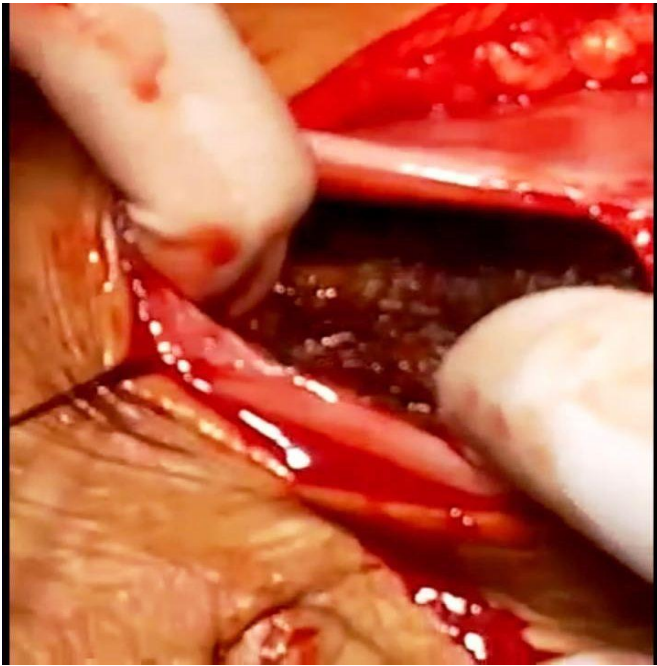


Fig 6: Operative Procedure

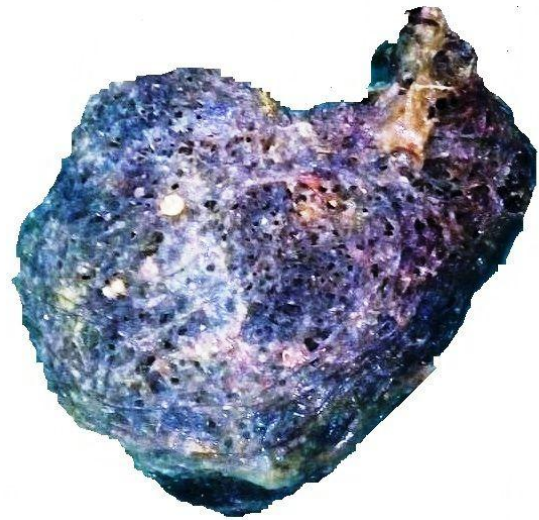


Fig 7b

Fig 7: a, b Showing removed trichobezoars



Fig 7a

DISCUSSION

Trichobezoar are formed due to hair ingestion whether self-hair or from others. It is an uncommon disorder in males but common in young females suffering with psychological problems such as trichotillomania or subtypes of pica disorder^[5].

Different types of bezoars are described in humans and according to the composition are named as phytobezoar (containing vegetable fibres), lactobezoar (milk products), pharmacobezoar (medications) and trichobezoar (hairs). Trichobezoars occur mostly in the second decade of life. They account for 12% of bezoars. Up to 90% of the all trichobezoars occur in girls younger than 20 years old probably due to their long hairs^[6]. Rarely, bezoars are can extend into the duodenum, jejunum, ileum, colon, appendix and Meckel's diverticulum. This condition, called Rapunzel syndrome and was first described by Vaughan et al. in 1968^[5].

The pathogenesis of bezoars is not clear. However, healthy individual stomach is able to clear even large foreign bodies in up to 80 to 90% of the cases, but hairs get trapped in mucosa of stomach as these are not pushed ahead by peristalsis due to the smooth surface of hairs. This results in bezoar formation. It means that bezoar formation occurs in the presence of both altered gastric anatomy or physiology and due to continuous eating and ingestion of the offending substance^[3].

Trichobezoar can be asymptomatic in the beginning and becomes symptomatic as it enlarges in size. A bezoar may also lead to mechanical obstruction, gastric perforation, gastrointestinal bleeding, anaemia and ulcer formation. Less common manifestations and complications include acute appendicitis, obstructive jaundice, nutritional deficiencies, and intussusceptions^[7].

Clinical suspicion should be high for trichobezoars in women with psychiatric problems presenting with abdominal pain, while diagnosis in a healthy patient requires a high index of suspicion, as it can present with nonspecific symptomatology. If suspected, trichobezoars can be diagnosed with radiological (ultrasound, barium study, Computerized Tomography) and by endoscopic techniques^[7].

Trichophagia may be more often present than the majority of physicians would expect. Many series of trichobezoar cases have been reported in literature but a few cases without trichotillomania have been reported so far^[3,4]. Trichotillomania results in highly variable patterns of hair loss, ranging from small undetectable patches of hair loss to total baldness and because of ingested hair accumulation, it

may present with trichobezoar. It can occur at any age, but starts most often in adolescence^[9]. However occurrence and definite association of trichobezoar and trichotillomania is not clear. Depending on the case series, 5 to 30% of the patients with trichotillomania engage in trichophagia, while 1 to 37,5% of these will develop a trichobezoar^[9].

Not all cases of trichotillomania have trichophagia, also all of patients with trichophagia will not have trichobezoars^[10]. Other psychiatric disorders, however, such as mental disorders, abuse, pica, obsessive compulsive disorder, depression and anorexia nervosa may also be associated with trichobezoar^[8].

An explanation for such discrepancy may be the fact that most of symptomatic trichobezoars are referred directly to surgeon due to possible complications whereas trichotillomania patients are mostly taken to psychiatry units by their relatives. Most of the time, due to urgency of surgery, treating surgeons may not notice psychiatric problem. Similarly, patients in acute surgical agony also avoids giving history of trichophagia due to shame and guilt.

A bezoar also means “protection against poison” or Ghosts and even for superstitions associated with good luck (as in our 1st case, tantric used mixture of hairs and ash for curative purpose against bad soul)^[9].

Trichobezoar have also reported in “Pica of infancy and childhood” condition without trichotillomania (as in our 2nd case). Pica is understood as a clinical condition in which the subjects often eats non-nutritive materials like soil, dirt, hairballs, ice, paint and sand, etc., It is usually seen in the setting of low intelligence(as in our second case); however, it can also occur in children with normal intelligence^[4,11].

Treatment of trichobezoars can be either endoscopic or surgical removal. Surgery can be open or laparoscopic approach. The endoscopic approach can be used for small size trichobezoar. Other minimally invasive modalities like extracorporeal lithotripter, endoscopic lithotripter and laser fragmentation are up-and-coming modalities. Endoscopic techniques have two advantages: first, we can see an extension of the trichobezoar in the intestine, and second, small trichobezoars can be removed.

Laparoscopic and minilaparotomy approaches are widely used as minimally invasive procedures. Laparoscopic treatment of bezoar was first described by Nirasawa and co-workers in 1998^[12]. A giant trichobezoar cannot be removed easily with laparoscopic technique as there are chances of spillage of hairs in peritoneum. Thus minilaparotomy could also be a substitute to laparoscopy in cases of large trichobezoars. We used open approach as it is a safe procedure and there was nil chances of contamination of the peritoneal cavity and wound with undigested hair. In our both patient’s six months follow up and long-term prognosis is excellent. Behavioral therapy was used to control their trichophagia.

CONCLUSION

Most of the cases of trichophagia and trichobezoar are described in patients with trichotillomania, which is considered to be an impulsive control disorder or other psychiatric disorders. The present cases exemplifies that patients of trichobezoar can have isolated trichophagia and can exist even in the absence of trichotillomania.

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