

Trichobezoar: A Case report.

Harpreet Singh¹, Malika Agrawal², Naveen Kumar Singh³, Roop kishan kaul⁴, Ikram Ilahi⁵

¹Associate Professor, Department of Surgery, Teerthankar Mahaveer Medical College, TMU, Moradabad.

^{2,4}Assistant Professor, Department of Surgery, Teerthankar Mahaveer Medical College, TMU, Moradabad.

³Professor, Department of Surgery, Teerthankar Mahaveer Medical College, TMU, Moradabad.

⁵Post Graduate Resident, Department of Surgery, Teerthankar Mahaveer Medical College, TMU, Moradabad.

Received: December 2016

Accepted: January 2017

Copyright: © the author(s), publisher. It is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Bezoars are persistent ingested concretions of human or vegetable fibers or concretions of foreign material that accumulate in the gastrointestinal tract mainly the stomach. Long term prognosis is good with proper psychological counselling and long term follow up. Fluoxetine or other SSRI'S have shown good results in treatment of trichotillomania. Parental counselling and education is important part of treatment.

Keywords: Parental counseling, Psychological counseling, Trichobezoar,

INTRODUCTION

Bezoars are persistent ingested concretions of human or vegetable fibers or concretions of foreign material that accumulate in the gastrointestinal tract mainly the stomach.^[1-4] The most common type of bezoar is the trichobezoar, in humans, made of mostly hair. Bezoars can also be made of fruit fiber (phyto-bezoars), vegetable (phyto-bezoars), milk curd (lacto-bezoars), or any indigestible material.^[5-7]

It is rare to see this condition. Approx 1% of patients suffering from trichophagia develop trichobezoar.^[6,3,4,7]

Name & Address of Corresponding Author

Dr. Malika Agrawal
Assistant Professor,
Department of Surgery, Teerthankar Mahaveer Medical
College,
TMU, Moradabad.

Large size bezoar can lead to decrease in blood flow to the mucosa of stomach/ small intestine leading to mucosal erosion, ulceration and sometimes perforation [Figure 1]. Obstruction at second part of duodenum where there is opening of papilla, may lead to obstructive jaundice and pancreatitis. Malabsorption leads to megaloblastic and iron deficiency anemia and protein losing enteropathy.^[8,6,9]

Hair when taken orally reaches stomach. These hair strands escapes peristaltic propulsion as they have slippery surface and are retained in gastric mucosal folds. Pylorus also creates resistance and this helps in holding up of hair in stomach. As more and more hair taken orally and reach stomach, peristalsis waves of stomach or intestine push them and hair

takes shape of ball. Slowly this ball increases in size and is unable to leave stomach because of its large size. Gradually more and more hair is entangled in this ball and it takes shape of stomach. Mucus secreted in stomach coats this trichobezoar and gives this a shiny glistening surface and acid secreted in stomach denature the hair protein which gives black colour to bezoar. Patient develops bad smell in her breath due to fermentation and decomposition of fats in bezoar.^[6, 7, 10-14]

Majority of patients having trichobezoars have psychiatric disorder like trichotillomania (pulling out of their own hair) and trichophagia (eating of hair). Rarely, some of these patients may eat hair from other sources. Some patient may remain asymptomatic for years. As the bezoar increases in size, it starts producing symptoms related to intestinal obstruction. Common clinical features are pain in abdomen, nausea, vomiting, features of intestinal obstruction and sometimes features of peritonitis. Other clinical features may be anorexia, weight loss, blood in vomitus, intussusceptions.^[6, 7, 15, 16]



Figure 1: Per-operative finding of Trichobezoar.

CASE REPORT

24 year old girl presented to the emergency department of Teerthankar Mahaveer medical college and research centre with diffuse abdominal pain, vomiting and abdominal distention from past 10 days duration, constipation from 15 days. No history of any previous illness, abdominal surgeries, abdominal injuries or allergies was found. Family history was not significant. On examination – Patient was conscious, oriented. Pulse rate – 115 beats per min, blood pressure – 90/70 mm of Hg, a febrile. On abdominal examination, distention, diffuse tenderness present, guarding, rigidity and rebound tenderness was found, bowel sounds were sluggish on auscultation. Per rectal examination was normal. Patient was resuscitated and relevant investigations sent. Hb – 11.9 gm/dl, TLC – 9,910 cells per mm³, neutrophils – 96%. X ray abdomen (AP view erect) showed features suggestive of small bowel obstruction. No free gas under left dome of diaphragm was present. Ultrasound scan of abdomen showed features suggestive of small bowel obstruction.

Patient was taken up for emergency exploratory laparotomy under general anesthesia on 16/12/2015.

Parts prepared and draped. Abdomen was opened in layers from midline with incision extending both above and below the umbilicus.

Small intestine was eviscerated through abdominal incision to enable examination of entire small bowel.

A black color lump obstructing the ileum (20 cms proximal to ileocaecal junction) causing pressure necrosis and perforation with multiple gangrenous patches in surrounding area were found [Figure 02]. Segment of ileum containing black color lump and perforated area of ileum was resected close to healthy margins both proximally and distally and the two ends of ileum delivered out as proximal ileostomy and distal mucus fistula in right iliac fossa.

Entire specimen was explored and black color lump was found to be having bunch of hairs Entire specimen was subjected to histopathological examination [Figure 03].



Figure 2: Per-operative finding of Trichobezoar.

No remaining portions of hair were found on careful palpation of rest of bowel. Operating surgeon requested the anesthetist to examine the patient's scalp for localized areas of missing or thinned hair, however none were found. Peritoneal cavity was irrigated using warm saline. Abdominal tube drain kept in pelvis and brought out through different incision from abdomen.



Figure 3: Black color lump was found to be having bunch of hairs.

Abdomen closed in layers. After the end of operation, the surgeon met with the patient's parents and husband and surgical findings were explained. In post operative period patient recovered well. On careful history asked in post operative period, patient gave history of eating hairs. Psychiatry consultation was taken for habit of eating hair and advice followed. The specimen sent for histopathology was reported as portion of ileum with necrosed portion containing bunch of human hair (trichobezor) [Figure 04].



Figure 4: Necrosed portion containing bunch of human hair.

DISCUSSION

Bezoars are persistent ingested concretions of human or vegetable fibers or concretions of foreign material that accumulate in the gastrointestinal tract mainly the stomach.^[1-4]

The most common type of bezoar is the trichobezoar, in humans, made of mostly hair.

Bezoars can also be made of fruit fiber (phyto-bezoars), vegetable (phyto-bezoars), milk curd (lacto-bezoars), or any indigestible material.^[5-7]

“Bezoar” word is derived from arabic word “bedzehr” which means “protecting against a poison”^[1]

In 1779, bezoar was found during autopsy of a patient died from gastric perforation causing peritonitis. This is the first reference of bezoar in human history.^[1, 17, 18]

It is rare to see this condition. Approx 1% of patients suffering from trichophagia develop trichobezoar.^[6,3,4,7]

Usually seen in adolescents and during the second decade of life.^[19,5]

Usually seen in females. This can be correlated with long hair in female. One reported case of male with trichobezoar ate hair of his sisters.^[20]

Majority of patients having trichobezoars have psychiatric disorder like trichotillomania (pulling out of their own hair) and trichophagia (eating of hair). Rarely, these patients may eat hair from other sources.^[3, 4]

Hair when taken orally reaches stomach. These hair strands escapes peristaltic propulsion as they have slippery surface and are retained in gastric mucosal folds. Pylorus also creates resistance and this helps in holding up of hair in stomach as more and more hair taken orally and reaches stomach, peristalsis waves of stomach or intestine push them and hair takes shape of ball. Slowly this ball increases in size and is unable to leave stomach because of it's large size. Gradually more and more hair is entangled in this ball and it takes shape of stomach. Mucus secreted in stomach coats this trichobezoar and gives this a shiny glistening surface and acid secreted in stomach denature the hair protein which gives black colour to bezoar. Patient develops bad smell in her breath due to fermentation and decomposition of fats in bezoar.^[6, 7, 10-14]

Entity named “rapunzel” syndrome is described in surgical text books .The name “rapunzel” syndrome comes from the grimm brothers’ fairy tale of a 12-year-old princess who was shut into a tower with neither stairs nor doors by an enchantress who climbed up the tower’s walls with the help of rapunzel’s long tresses.^[19,8]

Various criteria have been used in literature in defining rapunzel syndrome. Some consider it as any size bezoar which may cause intestinal obstruction. Some consider it as trichobezoar with a long tail which may extend up to jejunum or beyond jejunum. Some consider it as a gastric trichobezoar with tail extending till ileocaecal junction. Tail consists of distal end of fragments of hair attached to trichobezoar in stomach.^[7,17,21] It is a rare entity.^[7]

Usually patients are in 13 to 20 years of age group. Psychiatric disorder associated with rapunzel syndrome are trichotillomania (pulling out of their own hair) and trichophagia (eating of hair).^[7]

Majority of patients having trichobezoars have psychiatric disorder like trichotillomania (pulling out of their own hair) and trichophagia (eating of hair). Rarely, some of these patients may eat hair from other sources. Some patient may remain asymptomatic for years. As the bezoar increases in size, it starts producing symptoms related to intestinal obstruction. Common clinical features are pain in abdomen, nausea, vomiting, features of intestinal obstruction and sometimes features of peritonitis. Other clinical features may be anorexia, weight loss, blood in vomitus, intussusceptions.^[6,7,15,16]

Large size bezoar can lead to decrease in blood flow to the mucosa of stomach/ small intestine leading to mucosal erosion, ulceration and sometimes perforation. Obstruction at second part of duodenum where there is opening of papilla, may lead to obstructive jaundice and pancreatitis. Malabsorption leads to megaloblastic and iron deficiency anemia and protein losing enteropathy.^[6,8,9]

When in doubt, history of trichotillomania and trichophagia should be elicited. Physical examination may reveal pallor, features of malnourishment, halitosis, patchy alopecia.^[9]

This condition is easily identifiable on abdominal ultrasound and CECT abdomen. It appears as filling defect or as a mass.^[9,22] Gastroduodenoscopy is a gold standard in the diagnosis. Gives direct visualization, and aids in therapeutic intervention.^[9,8] Aims of management is bezoar removal and prevention of recurrence by psychiatric counselling.^[9]

It can be removed by using endoscopic techniques or surgery (laparoscopic approach or open removal. Choice of method depends on available resources, technical expertise, location, consistency and size of bezoar.^[9] Phytobezoars and lactobezoars are easy to remove using endoscopic techniques because of small size. It is less effective in trichobezoar removal as they are usually of large size. Bezoars can be broken into small pieces with the help of modified needle/knife (bezotome) or modified lithotripter (bezotripter) which uses acoustic waves.^[2,6,9]

Indications of surgical management includes large (particularly > 20 cms size), solid bezoar which cannot be removed endoscopically, Bezoar causing intestinal perforation, haemorrhage, Rapunzel syndrome (bezoar with significant extension)^[3]

Indications for Laparoscopic removal of bezors are small/ medium size bezoars. Advantages include reduced hospital stay, better cosmesis. Disadvantages includes peritoneal contamination is more as there are more chances of spillage of hair in peritoneal cavity. Consumes more time, difficult to examine rest of bowel for additional pieces which may lead to secondary obstruction.^[6, 8, 17, 23-25]

Open surgical procedures are Laparotomy with enterotomy or gastrotomy for removal of bezoar/ trichobezoar is most common approach.^[8, 17, 23]

Other methods for trichobezoars removal tried are introducing chemical substances in stomach for example—coca soft drinks, enzymes (pancreatic lipase, cellulose) to dissolve the bezoar. Limited success with this approach was found. [23, 26]

CONCLUSION

Long term prognosis is good with proper psychological counselling and long term follow up^[5] Fluoxetine or other SSRI'S have shown good results in treatment of trichotillomania. Parental counselling and education is important part of treatment. [5, 18, 24, 27, 28]

REFERENCES

1. Wang C, Zhao X, Mao S, Wang Y, Cui X. Management of SAH with traditional Chinese medicine in China. *Neurol Res.* 2006; 28:436-444.
2. Macksey L. Aspirated bezoar in a pediatric patient: a case study. *AANA J.* 2006;74(4):295-298.
3. Phillips MR, Zaheer S, Drugas GT. Gastric trichobezoar: case report and literature review. *Mayo Clin Proc.* 1998; 73:653-656.
4. Irving PM, Kadirkamanathan SS, Priston AV, Blanshard C. Education and imaging. *Gastrointestinal: Rapunzel syndrome. J Gastroenterol Hepatol.* 2007;22:2361.
5. Gonuguntla V, Joshi DD. Rapunzel Syndrome: A comprehensive Review of an Unusual Case of Trichobezoar. *Clinical Medicine & Research.* 2009;7(3): 99-102
6. Gorter RR, Kneepkens CM, Mattens EC, Aronson DC, Heij, HA. Management of trichobezoar: case report and literature review. *Pediatr Surg Int.* 2010;26(5):457-463.
7. Gonuguntla V, Joshi D. Rapunzel syndrome: a comprehensive review of an unusual case of a trichobezoar. *Clin Med Res.* 2009; 7(3):99-102.
8. Middleton E, Macksey LF, Phillips JD. Rapunzel Syndrome in a Pediatric Patient: A Case Report. *AANA Journal.* 2012; 80(2):115-119.
9. Wang YG, Seitz U, Li ZL, Soehendra N, Qiao XA. Endoscopic management of huge bezoars. *Endoscopy* 1998; 30:371–374.
10. Pace AP, Fearn C. Trichobezoar in a 13 year old male: a case report and review of literature, *Malta Med J.* 2003; 15:39-40.
11. Deslypere JP, Praet M, Verdonk G: An unusual case of the trichobezoar: the Rapunzel syndrome. *Am J Gastroenterol.* 1982; 77:467-470.
12. Sidhu BS, Singh G, Khanna S: Trichobezoar. *J Indian Med Assoc.* 1993;91:100-101.
13. Sharma V, Sahi RP, Misra NC. Gastro-intestinal bezoars. *J Indian Med Assoc.* 1991;89:338-339.
14. O'Sullivan MJ, McGreal G, Walsh JG, Redmond HP. Trichobezoar. *J R Soc Med.* 2001;94:68-70.
15. Jiledar, Singh G, Mitra SK. Gastric perforation secondary to recurrent trichobezoar. *Indian J Pediatr.* 1996;63:689-691.
16. Klipfel AA, Kessler E, Schein M. Rapunzel syndrome causing gastric emphysema and small bowel obstruction. *Surgery.* 2003; 133:120-121.
17. Naik S, Gupta V, Naik S, Rangole A, Chaudhary AK, Jain P, Sharma AK. Rapunzel syndrome reviewed and redefined. *Dig Surg.* 2007;24:157-161.
18. Vaughan ED Jr, Sawyers JL, Scott HW Jr. The Rapunzel syndrome. An unusual complication of intestinal bezoar. *Surgery.* 1968; 63:339-343.
19. Grimm Brothers: Rapunzel. Translated by Godwin-Jones R. Richmond, Virginia Commonwealth University Department of Foreign Languages, 1994-1999.
20. Hirugade ST, Talpallikar MC, Deshpande AV, Gavali JS, Borwankar SS: Rapunzel syndrome with a long tail. *Indian J Pediatr.* 2001;68:895-896.
21. Kaspar A, Deeg KH, Schmidt K, Meister R. [Rapunzel syndrome, an rare form of intestinal trichobezoars] *Klin Padiatr.* 1999;211:420-422.
22. Taori K, Deshmukh A, Rathod J, Sheorain V, Sanyal R. Rapunzel syndrome: a trichobezoar extending into the ileum. *Appl Radiol.* 2008 March; 34-35.
23. Groenewald CB, Smoot RL, Farley DR. A football-sized gastric mass in a healthy teen. *Contemp Surg.* 2006; 62:531-534.
24. Eryilmaz R, Sahin M, Alimoçglu O, Yildiz MK.[A case of Rapunzel syndrome. *Ulus Travma Acil Cerrahi Derg.* 2004; 10:260-263.
25. Fraser JD, Leys CM, St Peter SD. Laparoscopic removal of a gastric trichobezoar in a pediatric patient. *J Laparoendosc Adv Surg Techn A.* 2009;19(6):835-837.
26. Lopes LR, Oliveira PSS, Pracucho EM, Camargo MA, de Souza Coelho Neto J, Andreollo NA. The Rapunzel syndrome: an unusual trichobezoar presentation. *Case Rep Med.* 2010;2010:841028 (doi:10.1155/2010/841028.)
27. Memon SA, Mandhan P, Qureshi JN, Shairani AJ. Recurrent Rapunzel syndrome – a case report. *Med Sci Monit.* 2003; 9:CS92-CS94.
28. Swedo SE, Leonard HL, Rapoport JL, Lenane MC, Goldberger EL, Cheslow DL. A double-blind comparison of clomipramine and desipramine in the treatment of trichotillomania (hair pulling). *N Engl J Med.* 1989;321:497-501.

How to cite this article: Singh H, Agrawal M, Singh NK, Kaul RK, Ilahi I. Trichobezoar: A Case report. *Ann. Int. Med. Den. Res.* 2017; 3(2):SG01-SG04.

Source of Support: Nil, **Conflict of Interest:** None declared