WORLD JOURNAL OF PHARMACEUTICAL RESEARCH
SJIF Impact Factor 7.523
Volume 6, Issue 3, 1346-1353. Review Article ISSN 2277– 7105

A RARE AND UNUSUAL PRESENTATION OF ABDOMINAL PAIN IN YOUNG ADOLESCENTS. CASE SERIES AND REVIEW

***Waseem Raja, Rohey Jan, Rifat Khan, Sabiya Gul, Hilal Wani, Salma Maryam**

[1]Department of Medicine (Gastroenterology Division), Government Medical College Srinagar.

[2]Department of Critical Care & Anesthesiology, SKIMS Soura, Jammu & Kashmir, India.

[3]Department of Surgery, Government Medical College Srinagar. Jammu & Kashmir, India.

Article Received on
17 Jan. 2017,

Revised on 07 Feb. 2017,
Accepted on 27 Feb. 2017

DOI: 10.20959/wjpr20173-7959

***Corresponding Author**
Dr. Waseem Raja
Department of Medicine (Gastroenterology Division), Government Medical College Srinagar.

ABSTRACT

Trichobezoars are an uncommon cause of acute abdominal pain. A bezoar refers to a mass of undigestible foreign material found in the gastrointestinal tract, mainly stomach. The second most common bezoar is the trichobezoar, which usually occurs in the young psychiatric female patients with history of trichotillomania and trichophagia. We reported a case of a 16 year-old young psychiatric female patient with frontal patchy alopecia admitted with history of recurrent abdominal pain, vomiting abdominal distension and weight loss. Endoscopy confirmed a large hair ball occupying whole of the stomach and CT abdomen was also suggestive of trichobezoar. Laproscopic surgery was done and long hair ball with tail was removed in piecemeals. Another case we reported was a 18 year old male known case of Mental retardation and seizure disorder on antiepleptics, who presented with recurrent abdominal pain and hemetemesis - endoscopy revealed multiple metallic Nails inside stomach with erosions and was successfully removed by raw tooth forcep without any complications. Thus, trichobezoar should be considered as differential diagnosis of abdominal pain in a young psychiatric female patients. **Conclusions:** Early diagnosis and intervention are crucial, but not always sufficient to avoid serious complications. Further research is needed to increase knowledge regarding the etiology and treatment of this psychiatric condition.

KEYWORDS: Bezoar, trichobezoar, trichophagia, hair ball, hemetemesis, metallic nails.

INTRODUCTION

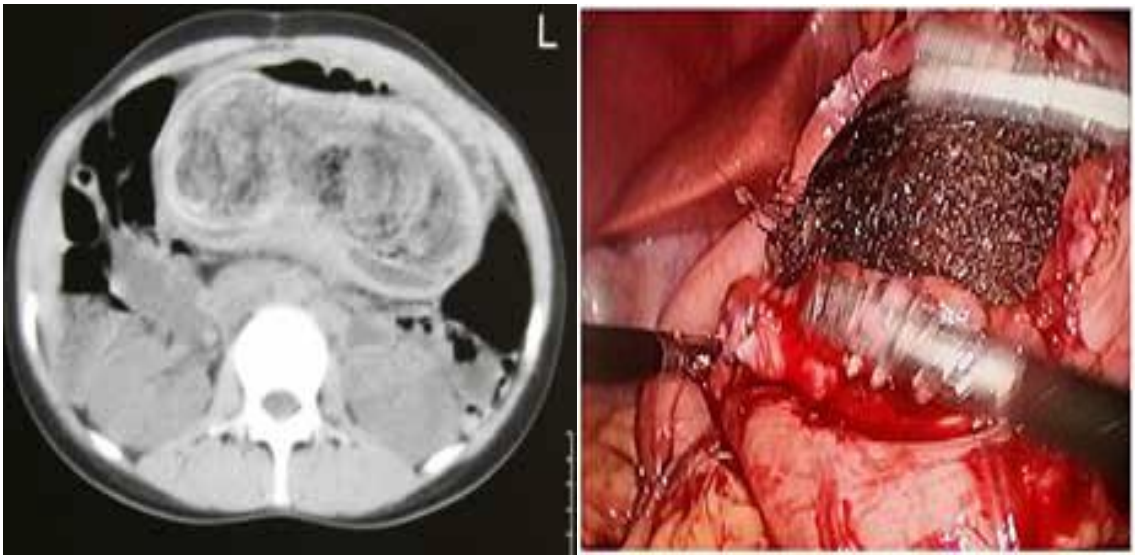
Bezoars are accumulations of human or plant fiber located in the gastrointestinal tract of both humans and animals. Trichobezoar is a rare condition presenting in just 0.06–4% of the population[1] defined as a mass of swallowed hair in the stomach.[1,2] Due to the smooth and indigestible nature of hair, when swallowed it accumulates in the gastric mucosa and over time it mixes with food and mucus, becoming compacted and forming a solid mass known as a trichobezoar.[1,3] When the gastric bezoar extends into the duodenum and the jejunum it is called Rapunzel syndrome and it is associated with higher risk of complications.[4] Presence of trichobezoar is associated with the conditions trichotillomania and trichophagia. Trichotillomania is a psychosomatic condition, involving the pulling out of one's hair.

Trichophagia is often associated with mental retardation and psychiatric conditions such as personality and eating disorders[5] and these patients often have some common pathophysiological and neurobiological risk factors for another pediatric eating disorder, known as pica.[5,6,7] Although rare and usually asymptomatic[5], trichobezoar should be suspected in young females presenting with nonspecific symptoms such as epigastric pain or mass, weight loss and fatigue. Other common signs or symptoms include vomiting, constipation, alopecia, iron deficiency, hypochromic anemia and vitamin B12 deficiency[3,8,5,9] Most commonly, the bezoars produce obstructive symptoms but rarely may cause ulceration and even bleeding. Diagnosis is suggested by upper GI series and confirmed by endoscopy. Treatment options, as described in the literature, include enzyme therapy (papain, cellulase and acetyl-cysteine), endoscopic fragmentation, or surgical excision.

CASE NO 1

A 16 year-old young girl presented to the Emergency Department with recurrent epigastric pain, of increasing frequency and severity, associated with nausea, vomiting, abdominal distension and weight loss of two months duration. There was no associated history of fever, night sweats, diarrhea, joint swellings and bleeding P/R. General physical examination revealed pallor and patchy alopecia in frontal region of her head. Per abdominal examination revealed the presence of a hard, solid mass from epigastric to periumbilical region, mobile and painless with no other remarkable features. Blood routine investigations –microcytic hypochromic anemia, Serum amylase, lipase, absolute eosinophillic count were within normal limits. Abdominal CT reported a distended stomach, filled with heterogeneous density material, probably bezoar **(Fig. 1**). An upper GI endoscopy was performed, confirmed

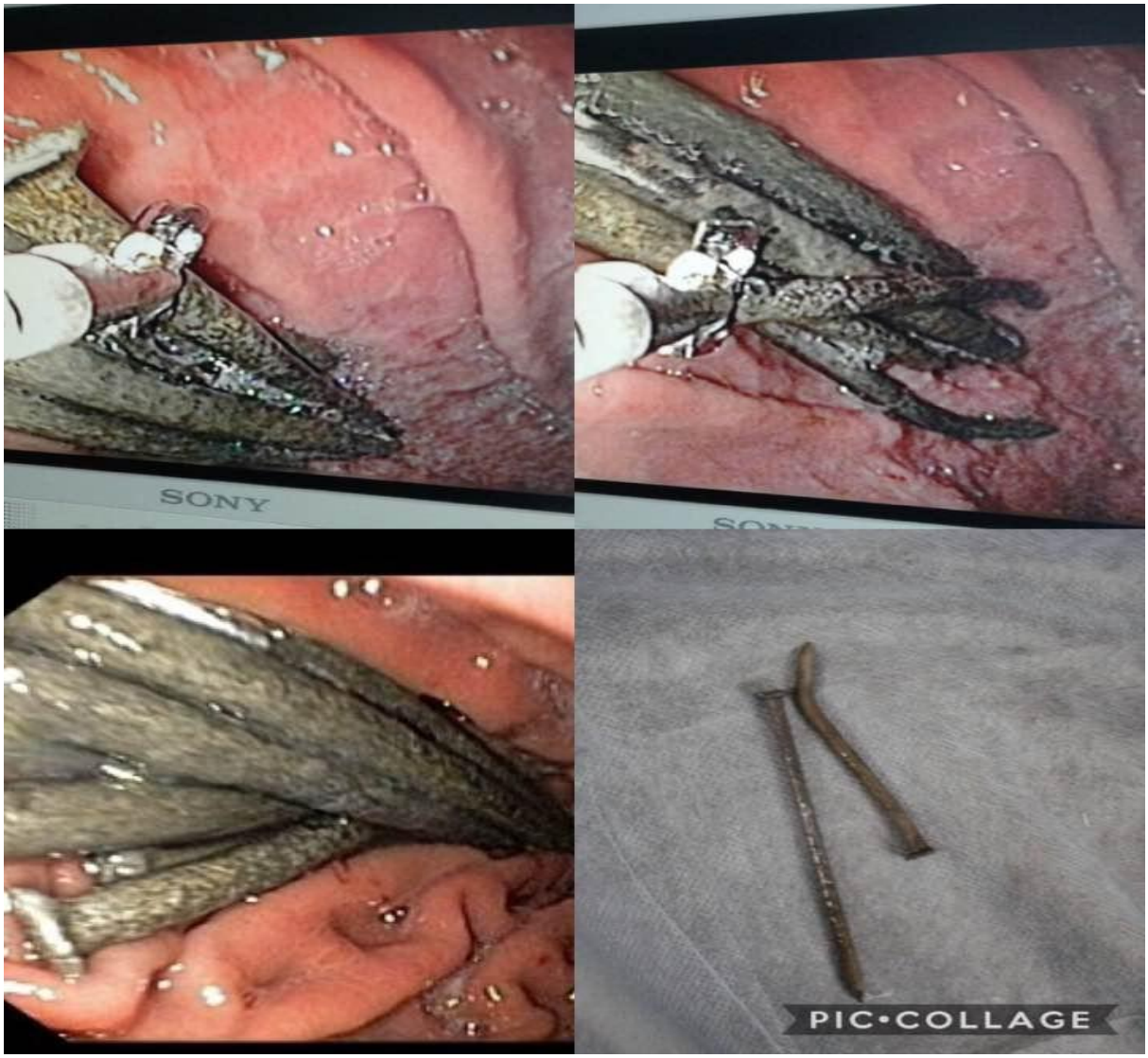
the presence of a Hair ball (trichobezor) occupying all gastric cavity. On revealing her past history, She has been addicted to hair eating for the past six years (Trichophagia). The laproscopic surgery was done – hair ball was cut by endo-scissors into piecemeals and was removed. **(Fig. 2**). Psychiatric consultation was also sort and proper counseling and anti psychiatric treatment was started. After six months of follow up she has no abdominal symptoms and is feeling better with antipsychotic medications.



(A). CT abdomen - distended stomach, filled with heterogeneous density material, probably bezoar. (B). Intraoperative Laproscopic removal of gastric hair ball.

CASE NO 2

An 18 year old young male who presented with recurrent abdominal pain and multiple episodes of coffee ground vomitus (hemetemesis). There was no history suggestive of EHPO, NSAIDs intake and no significant family history of Peptic ulcer disease or Malignancy and was managed conservatively with PPI infusion, Upper GI endoscopy revealed multiple metallic iron nails inside stomach of size 3-4 inches with superficial erosions and were successfully removed by raw tooth forcep **(Figures 1-4),** without any complication. On revealing his history, the patient was a known case of mental retardation with seizure disorder on drugs. Historically whenever patient would get a seizure, the parents would keep an iron nail between the teeth to avoid tongue bite as advised by some local quack. Most of the times patient would ingest the nail without any recognition. After 3 months of follow up, he has no abdominal symptoms and is on anti psychiatric treatment.



Figures 1- 4: Endoscopic removal of Iron Nails from stomach, in a mentally retarded, Seizure disorder patient who presented with UGI Bleeding.

DISCUSSION

Bezoar is an uncommon but potentially serious cause of abdominal pain in children. Several types of bezoars exist, including phytobezoars (plant material), lactobezoars (formula or milk, seen exclusively in infants)[10], pharmacobezoars (medications) and trichobezoars (hair). Bezoars are most commonly seen in the stomach, although may also involve the small bowel, particularly in the case of trichobezoars. "Rapunzel syndrome" is the name given to trichobezoars that extend into the small bowel, potentially traversing the ileocecal valve, with associated with obstructive symptoms.[11] While some phytobezoars and lactobezoars may be managed conservatively with enzyme-based therapy or observation, respectively, trichobezoars typically require surgical intervention in the form of laparotomy.[12]

Trichobezoars overwhelmingly tend to occur in females over males, with predisposing factors of trichotillomania and trichophagia. A recent case series of 7 patients with trichobezoars, all of whom were girls, found a mean age at presentation of 11.5 years.[13] Common presenting signs and symptoms of trichobezoars include abdominal pain, chronic gastrointestinal complaints, a palpable abdominal mass and small bowel obstruction.[13] Imaging plays an important role in diagnosis. On abdominal radiography, the diagnosis may be suspected when there is abundant mottled material admixed with gas distending the stomach. On CT, bezoars present as a heterogeneous, intraluminal mass typically interspersed with gas, distending but not adherent to the stomach or small bowel.[14] CT is helpful in delineating small bowel extension including more distal satellite lesions.[14]

Small gastric bezoars can be removed endoscopically however; there is limited evidence of success with only a 5% success rate in a recent study.[3] Endoscopic fragmentation and enzymatic dissolution prove to be problematic due to the difficulty of fragmenting large, dense, indigestible and solid nature of trichobezoars.[1] There is a significant risk of migration once fragmented which could result in residual satellite bezoars which can cause later obstructive phenomena. Endoscopy also poses the potential risk of iatrogenic esophageal perforation[3,6] and is therefore used more for diagnostic reasons rather than treatment.[6] Thus the role of endoscopy has advanced more towards diagnostic management than treatment. It allows us to differentiate, in the context of a gastric mass of an unknown nature, between a trichobezoar and foreign bodies that can be extracted or fragmented *via* endoscopy.[15]

In terms of surgical approach, this is where most of the debate lies with respect to management. Laparotomy, laparoscopy, endoscopic removal and even chemical dissolution in the case of phytobezoars have all been proposed. The choice is made on the basis of the size and composition of the bezoar.[16,17] As far as surgical removal is concerned, it is important to differentiate between the classic and the laparoscopic approaches. The first report of a successful result of laparoscopy for a trichobezoar was published in 1998 regarding a 7-year-old girl.[18] For the removal of small sized masses, laparoscopic surgery could be considered and an overall success rate of just 70% in experienced laparoscopic centers.[1]

To date the case reports endorsing this technique have been few, mainly in the pediatric population, limited mainly by the size and the presence of satellites in the intestine.[19,20,21,22-25] The combination of techniques has been used, implementing fragmentation

laparoscopically and removal endoscopically.[26] The advantages of laparoscopy as it relates to a trichobezoar are a lower rate of postoperative complications, a reduced hospital stay and a better cosmetic result. The disadvantages are a longer operating time, greater complexity in the review of the intestine in search of satellites, and the risk of contaminating the abdominal cavity with hair fragments.[27]

The laparotomy has been successful in most cases: over 100 cases of successful results with this technique have been described.[28] Complications with this technique are described in 12% of the cases[29] and these include intestinal perforation during removal of the trichobezoar[30,31], infection of the surgical wound[32], pneumonia and paralytic ileum.

In summary, our case highlights trichobezoar as an uncommon, yet important potential cause of abdominal pain in girls presenting with acute abdominal pain. The diagnosis of trichobezoar may be suspected in female patients with a history of trichotillomania, trichophagia, gastrointestinal symptoms and a palpable abdominal mass. However, imaging is a mainstay for accurate diagnosis, particularly of potential complications.

Disclosure: The authors report no conflicts of interest in this work.

REFERENCES

1. I. S. Al-Janabi, M. A. Al-Sharbaty, M. M. Al-Sharbati, L. A. Al- Sharifi and A. Ouhtit, "Unusual trichobezoar of the stomach and the intestine: a case report," *Journal of Medical Case Reports*, vol. 8, article 79, 2014.
2. *Concise Medical Dictionary*, Oxford University Press, Oxford, UK, 8th edition, 2014.
3. R. R. Gorter, C. M. F. Kneepkens, E. C. J. L. Mattens, D. C. Aronson and H. A. Heij, "Management of trichobezoar: case report and literature review," Pediatric Surgery International, 2010; 26(5): 457–463.
4. J. O. Nieto, J. V. Su´arez, J. J. V. Vergara and J. M. T. Molina, Rapunzel syndrome. A case report and literature review," *Revista Colombiana de Gastroenterologia*, 2011; 26(1): 70– 73. 4 Case Reports in Surgery.
5. M. J. O'Sullivan, G. McGreal, J. G. Walsh and H. P. Redmond, "Trichobezoar," *Journal of the Royal Society of Medicine*, 2001; 94(2): 68–70.
6. G. Chatzimavroudis, P. Christopoulos, S. Atmatzidis et al., "Pica: an uncommon cause of acute abdominal pain in children," *Indian Journal of Pediatrics*, 2011; 78(7): 886–887.

7. J. E. Grant and B. L. Odlaug, "Clinical characteristics of trichotillomania with trichophagia," *Comprehensive Psychiatry*, 2008; 49(6): 579–584.
8. J. R. Carr, E. H. Sholevar and D. A. Baron, "Trichotillomania and trichobezoar: a clinical practice insight with report of illustrative case," *The Journal of the American Osteopathic Association*, 2006; 106(11): 647–652.
9. A. Islek, E. Sayar, A. Yilmaz, C. Boneval, and R. Artan, "A rare outcome of iron deficiency and pica: rapunzel syndrome in a 5- year-old child," *Turkish Journal of Gastroenterology*, 2014; 25(1): 100–102.
10. Otjen JP, Iyer RS, Phillips GS, Parisi MT. Usual and unusual causes of pediatric gastric outlet obstruction. Pediatr Radiol, 2012; 42(6): 728e37.
11. Naik S, Gupta V, Naik S, Rangole A, Chaudhary AK, Jain P, et al. Rapunzel syndrome reviewed and redefined. Dig Surg, 2007; 24(3): 157e61.
12. Gorter RR, Kneepkens CM, Mattens EC, Aronson DC, Heij HA. Management of trichobezoar: case report and literature review. Pediatr Surg Int, 2010; 26(5): 457e63.
13. Fallon SC, Slater BJ, Larimer EL, Brandt ML, Lopez ME. The surgical management of Rapunzel syndrome: a case series and literature review. J Pediatr Surg, 2013; 48(4): 830e4.
14. Gayer G, Jonas T, Apter S, Zissin R, Katz M, Katz R, et al. Bezoars in the stomach and small boweleCT appearance. Clin Radiol, 1999; 54.
15. Gaia E, Gallo M, Caronna S, Angeli A. Endoscopic diagnosis and treatment of gastric bezoars. *Gastrointest Endosc*, 1998; 48: 113-114.
16. Andrus CH, Ponsky JL. Bezoars: classification, pathophysiology and treatment. *Am J Gastroenterol*, 1988; 83: 476-478.
17. Lee J. Bezoars and foreign bodies of the stomach. *Gastrointest Endosc Clin N Am*, 1996; 6: 605-619.
18. Nirasawa Y, Mori T, Ito Y, Tanaka H, Seki N, Atomi Y. Laparoscopic removal of a large gastric trichobezoar. *J Pediatr Surg*, 1998; 33: 663-665.
19. Pogorelić Z, Jurić I, Zitko V, Britvić-Pavlov S, Biocić M. Unusual cause of palpable mass in upper abdomen--giant gastric trichobezoar: report of a case. *Acta Chir Belg*, 2012; 112: 160-163.
20. Tudor EC, Clark MC. Laparoscopic-assisted removal of gastric trichobezoar; a novel technique to reduce operative complications and time. *J Pediatr Surg*, 2013; 48: e13.

21. Kanetaka K, Azuma T, Ito S, Matsuo S, Yamaguchi S, Shirono K, Kanematsu T. Two-channel method for retrieval of gastric trichobezoar: report of a case. *J Pediatr Surg* 2003; 38.
22. Cintolo J, Telem DA, Divino CM, Chin EH, Midulla P. Laparoscopic removal of a large gastric trichobezoar in a 4-year-old girl. *JSLS*, 2009; 13: 608-611.
23. Meyer-Rochow GY, Grunewald B. Laparoscopic removal of a gastric trichobezoar in a pregnant woman. *Surg Laparosc Endosc Percutan Tech*, 2007; 17: 129-132.
24. Hernández-Peredo-Rezk G, Escárcega-Fujigaki P, Campillo- Ojeda ZV, Sánchez-Martínez ME, Rodríguez-Santibáñez MA, Angel-Aguilar AD, Rodríguez-Gutiérrez C. Trichobezoar can be treated laparoscopically. *J Laparoendosc Adv Surg Tech A*, 2009; 19: 111-113.
25. Levy RM, Komanduri S. Images in clinical medicine. Trichobezoar. *N Engl J Med*, 2007; 357.
26. Yau KK, Siu WT, Law BK, Cheung HY, Ha JP, Li MK. Laparoscopic approach compared with conventional open approach for bezoar-induced small-bowel obstruction. *Arch Surg*, 2005; 140: 972.
27. Memon SA, Mandhan P, Qureshi JN, Shairani AJ. Recur-rent Rapunzel syndrome - a case report. *Med Sci Monit* 2003.
28. Al-Janabi IS, Al-Sharbaty MA, Al-Sharbati MM, Al-Sharifi LA, Ouhtit A. Unusual trichobezoar of the stomach and the intestine: a case report. *J Med Case Rep*, 2014; 8: 79.
29. Perera BJ, Romanie Rodrigo BK, de Silva TU, Ragunathan IR. A case of trichobezoar. *Ceylon Med J*, 2005; 50: 168-169.
30. Larsson LT, Nivenius K, Wettrell G. Trichobezoar in a child with concomitant coeliac disease: a case report. *Acta Paediatr*, 2004; 93: 278-280.
31. Zent RM, Cothren CC, Moore EE. Gastric trichobezoar and Rapunzel syndrome. *J Am Coll Surg*, 2004; 199: 990.
32. Varma A, Sudhindra BK. Trichobezoar with small bowel obstruction. *Indian J Pediatr*, 1998; 65: 761-763.