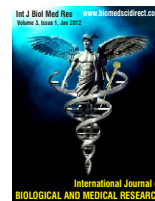


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Case report

Bezoars- A variety of presentations in tricobezoars

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ABSTRACT

Introduction: Human brucellosis is an important but ignored disease in India. Due to long standing fever and lack of typical signs and symptoms, patients with acute brucellosis are often tagged as the cases of Pyrexia of Unknown Origin (PUO). **Generally** these patients are investigated for diverse serological tests except for brucellosis. **Aims:** The present study was carried out to know the prevalence of brucellosis among the PUO cases and study the clinical and epidemiological aspects of brucellosis. **Materials and methods:** In this cross sectional study, 2379 PUO patients were investigated for the evidence of brucellosis. **Results:** Among 2379 cases, 114 patients were positive by RBPT. Significant titers could be demonstrated in 105 subjects by SAT and 2-ME tests. Brucellae could be isolated from 28 cases. Along with fever, joint pain and low backache were the commonest clinical symptoms. Stay in the rural area, animal exposure and raw milk ingestion were the major risk factors. **Conclusions:** Brucellosis accounted for 4.41 % of PUO cases. Serological tests are more sensitive when compared to blood culture. Efforts to create awareness regarding the existence of the disease among the physicians and preventive measures to be followed in rural population are needed.

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1. Introduction

A Bezoar is a tightly packed agglomeration of partially digested or undigested material in the intestinal tract. A typical patient is a teenage girl, but trichobezoar has been described in all age groups including the very young infants [1]. In children Bezoars are associated with pica, mental retardation and psychiatric disorders. A hair ball formed in stomach and intestine has been known as Trichobezoar. They present with varying symptoms and complications like anemia, malabsorption syndromes, haematemesis, intestinal obstruction, pancreatitis, jaundice etc.

2. Review

The first scientific study was done by "Imad ul oia' in early 16th century. It was known as badzehr in Arabic, padzhr in Persian and as panzehr in Turkish language, they all meaning it anti dot to poison or counter poison.

They are classified as Biological and Non-Biological Bezoars. Biological bezoars constitute trichobezoar, having hair as content; Phytobezoar again classified into opo bezoars, having fruit peelings and inio bezoars having plant fibers as content. Another term disopyrobezoar used for biological bezoars resulting due to

consumption of a fruit persimmon, commonly found in American continent, containing tannin and shibuol which polymerises in stomach promoting bezoars formation. Non biological bezoars are associated with pica and the contents are concretions, soap stones, clay, chalk, sand, slate, pencils, tooth paste, iron scrap, glass pieces, polythene (in animals). Similarly pharmacobezoars are masses of mostly tablets or semi solid drugs like sucralfate, aluminium hydroxide gel etc. Lacto bezoars are a specific type of food bezoar comprising of inspissated milk. It is most commonly seen in premature infants receiving formula milk food. Pseudo bezoar is an indigestible object introduced intentionally into digestive. Though common sites are stomach and intestine but a bezoar may be seen in the esophagus in young children. The bezoar in large intestine is known as choke.

3. Clinical presentation

Usual patients are adolescent, adult females having varying degree of psychiatric ailments like Trichotillomania with complaint of trichophagia, recurrent pulling out of own hair resulting into alopecia circumscripta. These hair get accumulated in stomach and may extend into intestine beyond ileocecal valve (Rapunjel syndrome).

Usually there are no symptoms until the bezoar reaches substantial size, a tricho bezoar of 2500 gm has been reported [2]. However, it will show itself earlier when there is a structural or functional narrowing of GI tract. (e.g. compression of duodenum by superior mesenteric artery) [3].

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The most common features are abdominal pain and intestinal obstruction, but the patient may also present with weight loss, anorexia or vomiting because the stomach is full, all the time.

Clinical features comprise of pain abdomen in epigastric region, poor appetite and early satiety (a pathognomonic feature) which ultimately leads to poor and inadequate nutrition leading to anemia, hypoproteinemia, cachexia, weight loss, develops multiple deficiency symptoms and his BMI gets reduced. Patient complains vomiting and vomits out food indigested days before. The psychological ailment either may develop or gets exaggerated. There will be tachycardia, low blood pressure and altered hemodynamic function. Mucosal erosions may lead to haematemesis.

Complications of bezoars include erosions, ulcer in gastrointestinal tract with or without bleeding, perforation, intussusceptions [4], intestinal obstruction, usually in the terminal ileum but sometimes of gastric outlet. A bezoar can also lodge in a duodenal diverticulum and cause cholestasis [5]. Malnutrition is a frequent accompaniment and a protein losing enteropathy has also been described in such patients [6]. Trichobezoars have been associated with Menetrier's disease [7] and transient pancreatitis [8]. Pressure necrosis of bowel wall may lead to perforation, peritonitis, a surgical emergency. Fatalities have been reported from obstruction[9].

Abdominal examination may reveal a mobile lump on epigastrium. There may be sign of obstruction usually in small intestine though colonic obstruction have been reported [10]. Sometimes a trichobezoar causes intermittent gastric outlet obstruction. The mass may be indentable on palpation – a physical sign introduced by Lamerton [11]. Some of these signs can lead to suspicion of malignant disease.

In Rapunzel syndrome, a long tail of hair extends from the main mass in the stomach along the small bowel to reach the ileum, caecum or beyond [12]. Only few cases have been published. The term Rapunzel syndrome was coined by Vaugan and associates in 1968 (Naik S, Gupta, Naik S et al)

4. Investigations

Haemogram and routine biochemistry will reveal features of anaemia, hypoproteinemia and various nutritional deficiencies.

5. Radiography

Plain films of abdomen are of limited use apart from confirming a diagnosis of obstruction. Sometimes plain film of left upper quadrant of abdomen may show Bezoar as a large mass with a convex upper border projecting into the gastric air bubble. An erect and a supine abdominal radiograph may show a prominent gastric outline with an intra gastric mottled mass, outlined by gas in the distended stomach, mimicking a food filled stomach.

Barium studies reveal an intra luminal filling defect without attachment to bowel wall. Also some times interstices of bezoar are filled with barium which remains in bezoar for hours after existing remainder of bowel, so it is expected to get delayed film if bezoar is suspected. Sometimes barium meal shows a near- perfect cast of stomach. Features of partial or complete bowel obstruction may be seen. Barium studies are of maximum benefit in the small bowel, in differentiating obstruction due to adhesions from obstruction caused by bezoars.

6. CT scan

The typical CT image of a gastric trichobezoar is a well defined ovoid intra luminal heterogeneous mass with interspersed gas [13,14] as in figure 01, if trichobezoar is distal to stomach and causing an obstruction, there will be dilated intestinal loops in addition to the intraluminal mass with gas retained in its interstices. Beyond the lesion, the bowel is collapsed. In the Rapunzel syndrome, CT scan have shown a hypo dense lesion in the stomach with mesh- like pattern. Oral contrast is sparse within the mesh, though prominent around margins (Figure 02). The presence of tail is reflected by small rounded areas of hypo density in other parts of small bowel [15].

Figure 01- CT scan of abdomen. Ovoid mass extending into stomach with inhomogeneous mottled appearance characteristic of bezoar (arrow) is readily identifiable.

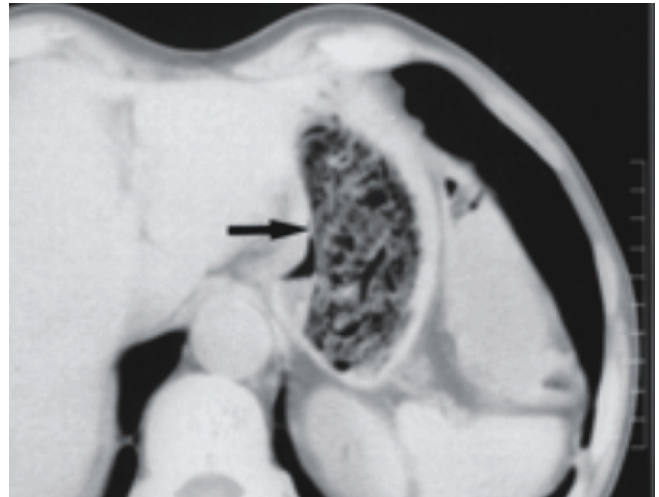
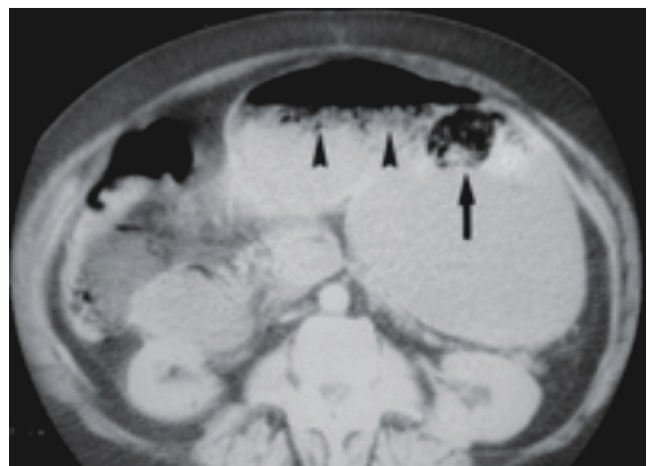


Figure 02- CT scan obtained without oral or IV contrast material shows inhomogeneous mass with mottled gas pattern (arrow) floating into distended stomach. This appearance differs from that of ingested food (arrowheads)



7. MRI

MRI seems less useful than CT scan for diagnosis of trichobezoars, because of low signal density and easily confused with air.

8. USG

Sonographic features are not pathognomonic, but Ko et al [16] noted that for trichobezoar in small intestine, an arc-like surface echo casting a clear posterior acoustic shadow within the dilated lumen can suggest diagnosis. Malpani et al reported in three patients that sonography revealed hyperechoic curvilinear dense strip at the anterior margin of the lesion associated with acoustic shadowing and no thorough transmission. The diagnosis was confirmed by barium meal and surgery. Similar findings were reported by McCracken colleague [17].

9. Endoscopy

Even if surgery is contemplated, a preliminary endoscopy is advisable. It will confirm the diagnosis, and occasionally the offending trichobezoar can be removed by this route. If a trichobezoar has been diagnosed elsewhere in the GI tract, endoscopy will also indicate whether the stomach contains any hair material. The typical color of a trichobezoar at endoscopy (and surgery) is black, irrespective of the original color of hair. This is due to denaturation of proteins in highly acidic gastric juice. There may be foetid odour due to entrapment of undigested fat in the hair mesh and bacterial colonization; this of course poses a threat of peritonitis after surgery [18].

Small bezoars can be removed endoscopically from the stomach like any foreign body, but small bezoars are not very common in clinical practice. Usually bezoars must be fragmented first or disrupted in some manner; by biopsy device or water jet under direct vision, the fragments then being removed through a large gastric lavage tube [19].

10. Minimally invasive surgery

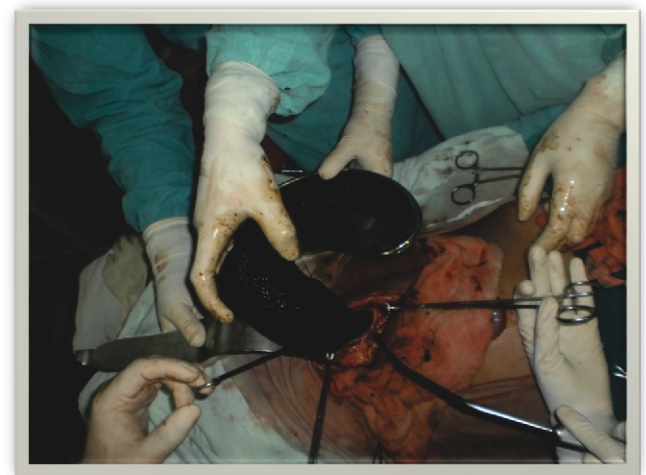
Many times, trichobezoar is an unexpected finding at laparotomy. An enterotomy is performed, the offending specimen is removed and the bowel defect is repaired. With a phytobezoar Robles et al [20] reported successful laparoscopic management by fragmentation with Babcock forceps and 'milking' into the caecum. Also laparoscopically, Nirsawa et al removed a large trichobezoar that extended from the stomach from the stomach into the duodenum [21]. In that case, endoscopic removal had been attempted twice under GA; trichobezoar was received through a gastroscopy and removed via a small suprapubic incision with excellent cosmetic result but operation time in 7 year-old girl was five hours [22].

Commonly, in diagnosed case of trichobezoar gastrostomy is done to remove it from stomach and enterostomy to remove bezoar from intestine.

Photograph 01- trichobezoar with long tail in a case presented with peritonitis.



Photograph 02- Removal of trichobezoar en-mass.



Photograph 03- Trichobezoar presented as Repunzal Syndrome.



Photograph 04- Trichobezoar presented as lump epigastium**11. Case reports****11.1. Case 1**

A 13 years girl was admitted in pediatrics ward having complaint of pain abdomen since last one week with distension abdomen and not passing flatus and motions since three days. She was also having vomiting since two days. Presentation was with vague symptoms, vomitus was mixed with blood. Patient was being treated for the symptoms of recurring episodes of anaemia and was transfused 5 units of blood during period of anemia. Her appetite was also gradually getting poor with symptoms of early satiety on increase since last two years. Patient was often febrile, feeling progressive weakness and losing weight significantly in last six months period, under nutrition and cachexia, reduced appetite. Although no perforation was noted in the patient, clinically the presentation created a diagnostic dilemma of intestinal perforation and peritonitis.

Patient was severely anaemic, dehydrated cachectic with fullness of abdomen, and swelling feet.

On examination she was conscious oriented but apathetic poorly built and malnourished with loss of buccal fat and sunken eyes sparse hair cachectic, with ugly look and long nails of all the fingers. She looked pale, had tachycardia with low volume pulse and hypotension. Gastric aspirate was hemorrhagic, dirty. Abdomen was distended. On palpation there was marked tenderness all over abdomen rigidity and guarding Bowel sounds absent, per rectal examination was found normal with no growth.

Investigations revealed anemia with Hb 6 gm%. there was significant leucocytosis with predominantly polymorphonuclear cells (78%). plain X-ray abdomen in standing position revealed few gas shadows and fluid levels with generalized haziness but there was no gas under diaphragm.

Ultrasonography showed moderate amount of fluid in peritoneal cavity and dilated loops of intestine. Other viscera were found normal. In diagnostic dilemma patient remained under conservative treatment for five days then was taken for surgery. On exploration under GA with right paramedian incision dirty fluid came out of peritoneal cavity. Small and large gut was found collapsed, few mesenteric lymph nodes were found enlarged, stomach was full, firm in consistency with normal mobility, surface of stomach was uniform and anterior wall of stomach freely mobile over luminal contents. Pylorus, duodenum and part of jejunum were also full and there was bluish uniform mass in continuity visible from serosal surface.

Considering mass in stomach of doubtful nature, a gastrostomy was done along and about 3 cm away and parallel to greater curvature of stomach. On opening dirty, brownish fluid with foul smell gushed out of stomach and the lumen showed a black bunch of hair, the diagnosis of trichobezoar was established, gastrostomy incision was extended. The mass of hair, filling whole stomach was retrieved out of incision wound gently, separating from mucosa of stomach and taken out en block (Photograph -01 & 02). This mass had continuity into pylorus, duodenum and part of jejunum where it was felt like a whip cord or tail and it could be squeezed out by external pressure and pulling out main mass gently. Stomach cavity was lavaged thoroughly with normal saline, Ryle's tube was repositioned and then stomach was closed in two layers as usual. On exploration there was no perforation seen in gut. A peritoneal drain was put into peritoneal cavity through a separate stab wound in right hypochondrial region and fixed to skin, postoperatively three units of blood transfused to patient. Patient had infection in stitch line of skin treated by removing two stitches on 5th day followed by dressing, remaining post operative period was smooth. Patient was discharged on 9th day after psychiatric counseling.

11.2. Case 2

She was a female of age 30 years having complaint of mild pain abdomen, anorexia, weight loss and fullness abdomen since last 8 months time, along with generalized weakness.

On examination there was elliptical palpable lump of 15 x 10 cm. size in epigastric region with mild tenderness. It was having smooth surface with clearly defined rounded margins. Patient look pale and mentally dull along-with features of cachexia with no symptoms of abdominal distension. Patient was passing flatus but had constipation since last 5 days.

Blood examination showed hemoglobin 7gm%, TLC 16,000/cmm, with predominantly polymorphonuclear cells 76% and lymphocytes 32%. USG abdomen showed big lump in epigastric region. There was no lymphadenopathy in supra clavicular region, both axillae, abdomen and pelvic region.

On exploration stomach found full of some mass. The stomach wall freely moving all over the underlying mass. The mass was extending into duodenum through pylorus. Gastrostomy done by an incision on the anterior stomach wall 3cm away from greater curvature in the line parallel to it. On opening there was a big ball of hairs occupying complete stomach and it was extending in to first part of duodenum through pylorus (Photograph -04). The ball of

hairs was nothing but trichobezoar and gradually it was removed through wound by separating easily it from mucosa of stomach with the help of fingers gently. The extension into duodenum could be pulled out easily as it was having glistening surface due to mucous covering it. After removal of trichobezoar the gastric cavity was washed thoroughly with normal saline solution and stomach was closed into layers. A peritoneal drain was put in the vicinity of stomach through separate stab incision in right hypochondriac region and fixed to skin. Abdomen closed in layers after proper homeostasis.

On third day the peritoneal drain was removed and on eight day patient stitches were removed. There was no infection in stitch line and patient was discharged next day after psychiatric consultation to her.

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11.3. Case -3

A girl mentally poor aged 15 year presented with 6-month history of pain and upper abdominal distension along with repeated episodes of vomiting. She had a habit of ingesting hair. On examination, a firm lump was noticed in upper abdomen easily palpable. Ultra sound and CT studies revealed a mottled filling defect in the stomach that extended into third part of duodenum.

Diagnosis-Rapunzel syndrome: A trichobezoar extending into ileum. (Photograph -03)

On gastrostomy a hair ball was seen in stomach. The mass was composed of long hair strands that formed a long tail extending into the ileum. The hair ball was pulled out of wound gradually, with gentle movement so that tail can be taken out en mass in continuity. Distal end of tail was also manipulated by squeezing pressure so that it helped tail of mass for easy retrieval from gastrostomy wound. Complete tail was taken out of wound at last successfully. Gastrostomy wound closed in layers. Abdomen closed in layers after proper hemostasis and putting a peritoneal drain into peritoneal cavity through a separate stab wound along with fixing drain with skin.

11.4. Case-4 :- A markedly distended stomach

A 25 year old female with autism had worsening pain abdomen and decreased appetite. Patient had history of intermittent, cramping pain in upper abdomen about two weeks back with no nausea and vomiting. She suffered weight loss of about 5 kg in last three weeks time. Her vitals were normal; lab investigation showed anemia with normal white cell counts. Chest X-Ray was normal; however a soft tissue shadow was seen in left upper quadrant.

Patient was kept NPO with support of IV fluid. Later a CT scan of her abdomen showed a markedly distended stomach suggestive of trichobezoar, confirmed by upper endoscopy, seeing to large size that filled whole abdomen gastrostomy was the only operative choice to remove the trichobezoar.

12. Discussion

Bezoars are rare masses formed of undigested materials found in the gastro intestinal tract. Bezoar formation is rare in healthy subjects. They are believed to be a physical manifestation of an underlying psychological disorder. The first case of human trichobezoar was described in 1779, by Baudamant [23], while the first surgical removal of trichobezoar was performed in 1883, by Schonborn. Incidence of bezoars peaks in the second decade. While 80% cases occur before the age of 30 years, more than 90% of bezoars occur in adolescent age girls. Bezoars though benign may cause serious complications. Early detection and surgical removal is associated with less than 4% mortality.

Trichobezoars arise from agglomeration of indigested hair with other indigestible organic fibers. Trichotillomania, the practice of picking hair out, and in association with habitual ingestion of hair (trichophagia) can predispose to the formation of trichobezoars.

Other concretions may arise from ingestion of various indigestible foreign materials. Delay in gastric emptying due to diabetes mellitus, mixed, connective tissue disease prior gastric acidity or hyperthyroidism, can predispose to the formation of bezoars [24]. Trichobezoars are formed over time by current ingestion of hair. The hairball enlarges over time as it enmeshes within itself all other indigestible fiber material. If left untreated, it may grow in size large enough to occupy the entire stomach and extend into the duodenum and small intestine. The most common presenting symptom is upper abdominal pain and weight loss. Patient also experiences stomach fullness, a 'dragging sensation' in the upper abdomen and periodic episodes of nausea and vomiting. If large in size, a uniformly firm, mobile, non tender abdominal mass can be palpated. A history of habitual ingestion of foreign substance like hair, or pica can usually be elicited and patients may be found to have focal alopecia [24]. An abdominal CT scan can delineate a well defined oval intraluminal mass with air bubbles retained within the interstices or a homogenous mottled appearance in the region of the stomach or intestine [25]. Upper GI contrast study would delineate a filling defect outlined by the barium, but is usually not necessary,

Presence of hair ball or a mobile mass within the stomach cavity directly visualized by upper GI endoscopy is confirmatory. A dark greenish brown or black mass with slimy surface and a fetid odor secondary to decomposition of various organic residue interspersed with hair is typically identified on upper GI endoscopy [26]. Upper endoscopy enables one not only to classify the type of bezoar depending upon its composition but also helps in deciding on the further mode of treatment.

Removal of bezoar is the main and primary treatment goal. Small bezoar may be fragmented into smaller pieces and aspirated endoscopically. Dissolution with papain, saline, acetyl cysteine, and cellulose are generally used for phytobezoars [27-28]. Various new techniques have been developed to tackle the issue of bezoars with variable success like water jet, bedside coca cola lavage [24] direct large channel endoscopic aspiration [29], dormia basket [30], forceps, polypectomy snare [31], ND YAG laser therapy, use of modified needle knife (bezotome) and modified lithotripter (bezotripter) [32].

However, endoscopic modalities are of limited use because the bezoar must be small and soft. Gastrostomy is the modality of choice for removal of large solid bezoars [27]. Enterotomy may be required when bezoars extend into small intestine. Recurrence cannot be avoided unless the underlying behavior is corrected. Behavioral training, in addition to serotonin re-uptake inhibitors or neuroleptic agents help to treat mood disorder and prevent recurrence. Prognosis is excellent after removal of mass, psychiatric counseling, and regular follow up.

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