CASE REPORT

Trichobezoar: a diagnosis which is hard to swallow and harder to digest

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SUMMARY

An 11-year-old girl with normal development and good academic progress at school presented with an abdominal mass. She lived with her aunt as her mother suffered from severe bipolar disorder and was unable to raise her. There were also multiple bereavements in the family. She had a history of alopecia and admitted to have eaten her hair to relieve stress. Examination was normal apart from a solid mass in the left hypogastrium. CT abdomen revealed a huge intra-abdominal mass covering most of the lumen. Open laparotomy was performed to remove the mass which was confirmed to be a trichobezoar. She made a good postoperative recovery and was discharged after 7 days. She has been referred to psychology services. Bereavement alone without psychiatric, physical disability or autism can be associated with trichobezoar. More studies are needed to study the association of trichotillomania, trichophagia and trichobezoar.

BACKGROUND

Foreign bodies can be lodged in any part of our body. Natural orifices are the most common sites. Inhalation and ingestion share almost equal incidences.¹ It can cause many complications ranging from simple discomfort, obstruction or extreme life-threatening scenarios. Foreign bodies in children are more serious, due to relatively small anatomical structures. Trichobezoar is an ingestional foreign body. Case reports of trichobezoar are still relatively uncommon. Only two cases of trichobezoar have been reported in Ireland. Very little is known about the association of trichotillomania (TTM), trichophagia and trichobezoar. Bereavement as a prodromal cause for trichobezoar is very uncommon.

CASE PRESENTATION

An 11-year-old girl presented to our accident and emergency department with abdominal pain and diarrhoea for 2 days. She had no history of fever, vomiting or urinary symptoms. She had a history, about 5 years prior to presentation, of alopecia in her frontal scalp which had self-resolved. The girl had normal development and good academic progress at school. She also did not have any allergies.

Vaccinations were up to date. She lived with her aunt since birth as her biological mother was suffering from severe bipolar disorder. Her aunt obtained legal custody after her biological mother was murdered 2 years ago. She had also lost her grandfather about 4 months ago to whom she was very close. She almost did all her daily routine in his

companionship. She reported that she felt lonelier after his death. Her aunt reported that she liked to stay indoors most of the time; however, she was lively and more mature for her age.

On examination of her abdomen, a 10×5 cm oblique mass extending from the left hypogastrium towards the epigastrium was observed, non-tender, not continuous with liver or spleen. Upper border was not felt as it was behind the left lower ribs. Other systemic examinations were within normal limits.

Discussion

Bereavement without psychiatric, physical disability or autism could be a cause for a trichobezoar. More studies are needed to study the association of TTM, trichophagia and trichobezoar.

INVESTIGATIONS

- ▶ Normal full blood count, urea, electrolytes, liver function test, erythrocyte sedimentation rate.
- ▶ Chest X-ray, plain film abdomen—no definite diagnostic finding.
- ▶ Abdominal ultrasound not able to penetrate the solid mass.
- ▶ CT abdomen. Intra-abdominal mass almost covering the whole lumen of the stomach (figures 1 and 2).

Figure 1 CT abdomen showing trichobezoar covering the whole lumen of the stomach.

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Figure 2 CT scan of the abdomen showing the cross section of the trichobezoar.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis includes splenic enlargement due to lymphoma, left liver lobe tumour, neuroblastoma and a rare carcinoma of the stomach.

TREATMENT

Open laparotomy, nil by mouth for first 48 h. Discharged home as she recovered well after 7 days of inpatient stay in the hospital.

OUTCOME AND FOLLOW-UP

Sixteen centimetre long trichobezoar was removed from her stomach (figure 3). The next surgical outpatient department appointment is scheduled in 6–8 weeks. She is awaiting psychology input at the earliest possible date.

DISCUSSION

A bezoar is a hard indigestible mass that is found in the stomach or intestine. There are three types of bezoars that occur in children namely, trichobezoars, lactobezoars and phytobezoars composed primarily of hair, milk and vegetable materials. Trichobezoars were described first by Baudamant in 1779. Trichobezoars are thought to form when hair, once swallowed, escapes peristalsis in the stomach due to its slippery surface, and becomes trapped in the gastric folds of mucosa. As more hair accumulates from repeated ingestion, peristalsis causes this ball to become enmeshed and harden which with time assumes the shape of the stomach. Ultimately, this may result in an obstructive foreign body or cessation of peristalsis.^{1–4} It is assumed that the pylorus is the hold up that allows the stomach to be the

Figure 3 Postoperative specimen of trichobezoar.

frequent location for bezoars.¹ The mucous that covers it gives a shiny surface, while the denatured protein secondary to gastric acid juices gives it its black colour.^{3, 4} A foul odour may be present in the bezoar and the patients' breath caused by decomposition and fermentation of fats.^{1, 3, 4}

TTM (hair pulling) and trichophagia (hair eating) may result in a trichobezoar. It is estimated that 1 in 2000 children worldwide suffer with TTM.³ Thirty per cent of those who suffer from TTM will also suffer from trichophagia, while only 1% of those who suffer from trichophagia will develop a trichobezoar.^{2, 3} Most cases of trichobezoar occur in women. Ninety per cent of trichobezoars occur in women, 80% of these occurring under the age of 30 years.¹ Most cases of TTM begin in childhood and predominantly in women.⁵ TTM has a bimodal age of onset in childhood. There is a positive association of focused hair pulling with age.⁵ These are very young children, less than 2 years old and onset in late childhood/early adolescence.⁵ A recent study has suggested onset within two further subsets between the ages of 7 and 18, one at age 7–8 and another at age 11–12.⁵ In children who begin pulling before 2 years of age, TTM is often time limited and has better prognosis.^{5, 6} There are two different types of pulling that occur; automatic pulling is habitual and often occurs without the individual's awareness whereas focused pulling, in contrast, is pulling with awareness. This is often carried out to decrease an unpleasant urge or as a reaction to negative emotional states (eg, stress, anxiety).^{5, 6}

The most common presenting features are abdominal pain and intestinal obstruction.⁴ Patients may be asymptomatic for years. Symptoms develop as the trichobezoar enlarges and begins to cause obstruction and may include abdominal pain, nausea, vomiting, obstruction and peritonitis.^{1, 4} There may also be weight loss, anorexia, halitosis and pain related to eating meals.^{1, 3} On examination, a mobile, well-defined mass may be palpable in the epigastrium. This may be indentable on examination, Lambertson's sign, as quoted by O'Sullivan *et al.*⁴ There may be evidence of alopecia on inspection while halitosis may also be present.¹ Differential diagnosis includes splenic enlargement due to lymphoma, a left liver lobe tumour, a neuroblastoma and carcinoma of the stomach. There are many complications associated with trichobezoars described, gastric ulceration, perforation, haemorrhage, malabsorption and obstruction among these.¹ Rapunzel syndrome refers to a rare form of trichobezoar that extends to the small intestine.

X-ray of the abdomen may show a radio-opaque mass in the stomach. Ultrasound reveals highly echogenic areas but is often not diagnostic. Barium studies show barium in the stomach interspersed within the gastric mass.³ CT abdomen is more readily available in many departments and is often the investigation of choice in diagnosis and well-defined heterogeneous masses, separate from the gastric lumen wall, are seen. If there is obstruction, dilated loops may also be present.^{3, 4}

There are two arms to the treatment of trichobezoar: treatment of the physical bezoar and treatment of any underlying psychiatric issues. Bezoars may be removed by endoscopy or surgery depending on its size and consistency. Small trichobezoars may be removed endoscopically. Bezotomes and bezotriptors have been used to fragment large trichobezoars.¹ Surgery may be performed by laparotomy or laparoscopy, the former necessary in larger solid trichobezoars, and achieved by gastrotomy or enterotomy.^{1, 3, 4} There is very little available information to guide treatment of young children.³ Behavioural therapy has shown some promise in treating TTM in adolescence and childhood with better outcomes than during adulthood.⁷ Trichophagia may occur due to emotional stress or personality disorder, not necessarily because of severe

neuropsychiatric disorders.³ However, case reports of trichobezoars and Rapunzel syndrome have linked trichophagia to anxiety, eight mental retardation, four neglect (fatal case p6) or bereavement (rap rec and redefined p7). There is a risk of recurrence in later life if not appropriately addressed.⁸

Learning points

- ▶ Do not forget trichobezoar as a differential for epigastric abdominal mass.
- ▶ Include trichobezoar in your differential for abdominal pain especially if patient has been under stress even if showing no outward psychological signs.
- ▶ Think of trichotillomania (TTM) when a paediatric patient has thinning hair/alopecia especially when this coincides with times of increased stress.
- ▶ Treat the trichobezoar and the psychological issues that have led to TTM and trichophagia.
- ▶ Recent study suggests that cognitive-behavioural therapy is the most appropriate treatment for TTM and this is important to prevent recurrence.
- ▶ Consider follow-up screening to ensure no recurrence of trichobezoar.

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