Bezoars are deposits of undigestible material in the stomach or intestines. The word ‘bezoar’ comes from the Arabic ‘badzehr’ or the Persian ‘padzhar’ and means ‘antidote’ or ‘protection against poison’, and in traditional Chinese medicine the animal concretions are in fact used as antidotes in cases of poisoning. The first bezoar reported in a human is said to have been observed in 1779 during an autopsy, and was associated with gastric perforation and peritonitis.

A PubMed search showed that first case of gastric bezoars in the medical literature was reported in 1946, in a cat.[1] In humans, bezoars may be composed of vegetable fibres (phytobezoar), persimmon fibres (diospyrobezoar), human hair (trichobezoar), milk products (lactobezoar) or drugs (pharmacobezoar). The first case of trichobezoar was reported in 1948.[2]

Gastric trichobezoar with a long intestinal tail is known as Rapunzel syndrome, after the Grimm brothers’ story in which the prince climbed into Rapunzel’s tower using the tresses of her long hair.

Clinical diagnosis of bezoars may be difficult, especially when no psychiatric disorder associated with human hair ingestion (trichophagia, trichotillomania) is reported. (Trichotillomania is the desire to pull hair from the body, and trichophagia means eating of hair. They are usually associated with non-scarring alopecia.)

We present the case of a neglected young girl who presented with a hair and wool gastric bezoar.

Case report

An 18-year-old girl from a rural area, weighing 62 kg and with no history of mental disturbance, was admitted to the emergency department with nonspecific abdominal pain. Two years previously she had undergone surgical removal of a Meckel’s diverticulum, and 4 months previously she had given birth to a healthy baby. She gave no other past medical history.

Physical examination revealed her to be a healthy girl with long hair. No alopecia was noted. On palpation, a painful abdominal mass was identified in the epigastric region. An abdominal computed tomography (CT) scan revealed an obstructive non-homogeneous abdominal mass, and a gastric tumour was suspected.

Laboratory tests showed no abnormal results: specifically, a haemoglobin concentration of 12.4 g/dl and a haematocrit of 39.8% excluded anaemia, the while blood cell count was in the normal range, and liver and kidney function were normal.

A midline laparotomy followed by gastrotomy was performed. Because the stomach wall and mucosa were normal and the stomach was full with a soft, detachable mass, the obstructive foreign bodies were removed and the gastrotomy was closed. The bodies removed were a giant J-shaped (or stomach-shaped) hairball measuring 320×180×60 mm and two pieces of sheep’s wool measuring 150×45×60 mm and 160×30×100 mm (Fig. 1). The hairball was composed of human hair, mucus and sheep’s wool.

The patient recovered well from the operation, and psychiatric counselling was prescribed to prevent recurrence of the problem.

Discussion

The commonest human bezoars are composed of human hair and are called trichobezoars. To the best of our knowledge, only 8 previous cases of mixed bezoars have been reported in the literature, between 1969 and 2008; all of them have been hair-cotton bezoars.[3] This is the first case of a bezoar consisting of a mixture of human hair and sheep’s wool.

Bezoars are usually diagnosed in mentally retarded or psychiatric patients, but gastric dysmotility was a further problem...
in a depressed patient with a cotton bezoar.\textsuperscript{[3]} In 90\% of reported cases the patients were adolescent girls or young women between 13 and 30 years of age, with long hair and trichotillomania, tricophagia, picca, depression, obsessive-compulsive disorder or anorexia nervosa, a history of having been abused as children, or other emotional and psychiatric disorders.\textsuperscript{[4]} Psychological problems are not always evident, so a detailed history is indispensable.

Of patients with trichophagia, approximately 1\% develop a trichobezoar.\textsuperscript{[5]} In the early stages, when the bezoar is small, the patient may be asymptomatic. Larger bezoars may cause luminal stenosis and mucosal erosion. In these cases, the commonest symptoms are nonspecific abdominal pain, nausea, vomiting, and iron deficiency or megaloblastic anaemia.\textsuperscript{[6]} Sometimes these hairballs result in weight loss, intestinal malabsorption or gastro-intestinal obstruction, perforation, abscesses, pancreatitis, obstructive jaundice, gastric emphysema and even death, especially in children. Gastric fermentation of fats may give the patient's breath a putrid smell.

If any of the above symptoms give rise to suspicion that a trichobezoar may be present, endoscopy should be performed even if the patient appears mentally healthy. Our 18-year-old patient was neglected, but did not have a history of psychiatric disorders. An important element in her history was the birth of her baby 4 months previously. Pregnancy in a young girl can result in depression, and could have alerted the doctor to the possibility of a trichobezoar.

Small bezoars can be removed endoscopically. Gastrotomy or enterotomy is necessary to remove larger ones, followed by exploratory enteroscopy for any that might remain behind. Phytobezoars may be fragmented or completely dissolved with acetylcysteine, or with Coca-Cola or other carbonated beverages, in as little as 12 hours (periods of 7 days and as much as 12 months have also been reported). Trichobezoars are not as sensitive to these substances. Devices such as bezotomes or bezotriptors can be used to fragment huge, solid bezoars, but this may result in oesophageal perforation.\textsuperscript{[7]}

An accurate pre-operative diagnosis may avoid the necessity for gastroscopy. In our case, CT findings led to suspicion of a gastric tumour. Endoscopy was not performed because of the acute gastric obstruction, and gastroscopy was the best surgical option.

Bezoars are rare, and because the symptoms may be similar to those of gastric or intestinal tumours, the differential diagnosis may pose a real challenge. After removal of bezoars, patients need long-term psychiatric follow-up to prevent recurrence. Endoscopic follow-up is also advised.

What made our case unusual was the post-delivery diagnosis in a young girl without a psychiatric history, and the previously unreported mixture of human hair and sheep’s wool comprising the bezoar.

REFERENCES