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Trichotillomania, trichophagia, trichobezoar – summary of three cases. Endoscopic follow up scheme in trichotillomania

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Summary

Aim. Trichotillomania is a lack of control of one's hair pulling. It is estimated that about 1% of population develops trichotillomania. In up to 20% of patients with trichotillomania hair pulling is followed by swallowing – trichophagia. Trichobezoar forms in about 30% of patients with trichophagia.

Material and methods. In 2008–2014 three patients patient underwent surgery due to trichobezoar. One patient has had a history of trichotillomania. On admission abdominal Xray and ultrasonography revealed abdominal mass. Diagnosis was confirmed in abdominal computed tomography.

Results. All trichobezoars were removed from the intestinal tract during laparotomy with gastric wall opening. In one case – Rapunzel syndrome – hair mass was also removed from the small bowel. After the surgical treatment all patients were referred to psychiatrist.

Conclusions. In patients after a surgery due to trichobezoar as well as other patients with trichotillomania, control of hair accumulation in the gastrointestinal tract remains a problem. The authors propose endoscopic follow up scheme in 6, 12, and 24 months after the surgery as well as for other patients with trichotillomania.

Key words: trichotillomania, trichobezoar, endoscopic follow up

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Introduction

Trichotillomania is defined as lack of control of one's hair pulling. Hair loss can be present in all body regions, although they are most common in head and face [1]. Other behaviours like hair swallowing, chewing etc. can also be present [1].

Trichotillomania often presents as part of a wider clinical picture. Comorbidity with depression, anxiety disorders, addictions or eating disorders can be present [1]. Detailed clinical description of trichotillomania is presented in "Trichotillomania and trichophagia – diagnosis, treatment, prevention. The attempt to establish guidelines of treatment in Poland" [2].

Aetiology of trichotillomania remains unknown. Genetic as well as environmental factors can lead to disease development [3]. In DSM-5 and ICD-10 trichotillomania is classified in the impulse-control disorders category.

Incidence is hard to measure, mainly due to the characteristics of the disorder and the fact that patients are ashamed of revealing the symptoms. It is estimated that about 1% or paediatric population develops trichotillomania. Among children and adolescents up to 18 years the disorder is much more common than in adult population [4]. Females are also at higher risk than men [5, 6, 7].

In up to 20% of patients with trichotillomania swallowing follows hair pulling – trichophagia [1, 8, 9]. In extreme cases patients swallow also human and animal hairs found in surrounding environment.

Trichobezoar forms in about 30% of patients with trichophagia [9]. Little compliance to peristaltic movements leads to hair accumulation in the gastrointestinal tract and mass formation. This can lead to passage, bile and pancreatic juice obstruction. Pressure on the bowel wall can cause mucosa erosion and perforation. Among other complications intussusception, mechanical jaundice, malabsorption and pancreatitis are noted [10, 11].

Trichobezoar has to be taken into consideration in differential diagnosis in patients presenting malabsorption, anaemia, weight loss, persistent abdominal pain – especially in the population of adolescent girls.

Abdominal ultrasonography, and radiography confirm presence of the abdominal mass with possible signs of ileus. MRI and CT of the abdomen show higher sensitivity. Diagnosis can be confirmed during endoscopy of the gastrointestinal tract or intraoperatively [12].

In most cases the method of choice in treatment of trichobezoar in laparotomy with wide gastric and – in specific cases when bezoar mass extends beyond the gaster (Rapunzel syndrome) – also small bowel opening. Among other methods endoscopic and laparoscopic removal and enzymatic fragmentation are mentioned in the literature [13–16].

Aim

Aim of this paper is to present three trichobezoar cases in paediatric patients.

Material and methods

Between 2008 and 2014 three female patients at the age between 13 and 16 years underwent surgery due to trichobezoar. Patients' documentation was analysed retrospectively.

Table 1. **Patients who underwent surgery due to trichobezoar**

Patient	1	2	3
Sex	W	W	W
Age	15	16	13
Medical history of psychiatric illness		Trichotillomania	
Symptoms			
hair loss	yes	no	yes
stomach pain	yes, since 2 weeks	yes	yes, since 6 months
abdominal mass	no	yes	no
weight loss	4 kg	no	6 kg in 6 months time
Laboratory tests			
Hb g%	8.9	12.1	10.6
imagining (diagnosis)			
USG	gastric mass	gastric mass	gastric mass
X-ray	abdominal mass with calcifications	abdominal mass	abdominal mass with calcifications
CT		bezoar, Rapunzel Syndrome	bezoar with calcification
Endoscopy	huge trichobezoar filling the entire stomach		
Duration of hospitalisation (days)	16	23	15

Material analysis

Among patients who underwent surgery due to trichotillomania only one patient had been previously treated for trichotillomania, other two had not been previously diagnosed with psychiatric disorders. The patient with a 10 years history of trichotillo-

mania was admitted to the hospital with abdominal pain and a palpable abdominal mass. The second patient was treated with anaemia and in the third patient trichobezoar was found in endoscopy. During the procedure mucosal erosion was also noted. Because of the risk of perforation and ileus as effect of trichobezoar fragmentation during endoscopy the child was referred to the surgical ward.

In two cases there was a palpable mass in the abdomen. Hair loss was also observed in two patients. One patient presented with pale skin, skin rash and tachycardia. Anaemia was present in two cases – haemoglobin levels 8.9 and 10.6 g/dl.

Abdominal X-ray and ultrasonography revealed abdominal mass. Diagnosis was confirmed in abdominal computed tomography – CT (Figure 1). All three trichobezoars were removed from the intestinal tract during laparotomy with wide gastric wall opening (Figure 2). In one case – Rapunzel syndrome – hair mass was removed also from the duodenum and small bowel (Figure 3). There were no complications in the postoperative period. Hospitalisation time was approximately two weeks. Oral feeding was administered in 10th postoperative day. After the removal of trichobezoar all patients were referred to a psychiatric treatment.

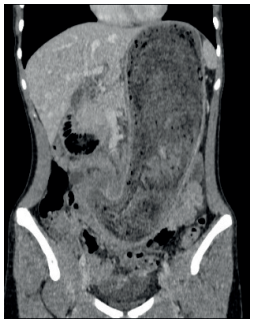

Figure 1. **CT image, stomach filled with trichobezoar**

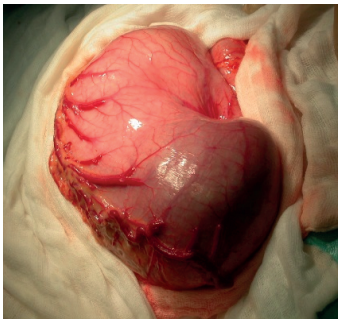

Figure 2. **Intraoperative image**

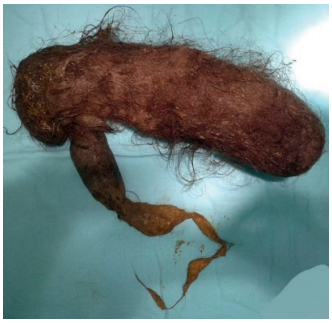

Figure 3. **Rapunzel Syndrome – trichobezoar mass reaching the small intense**

Discussion

Rarity of the trichobezoar prolongs time to diagnosis. Two patients were previously treated with anaemia, in one abdominal tumour was suspected. Only in the patient with psychiatric treatment history trichobezoar was considered in differential diagnosis.

Sensitivity of abdominal ultrasonography and X-ray in diagnosing trichobezoar is 25% and 18% respectively, whereas for abdominal CT it reaches 97% [12]. In the presented material diagnosis was confirmed in abdominal CT.

Duration of the hair accumulation process leads to malabsorbtion, passage obstruction and gastrointestinal wall pressure. Gastrointestinal perforation and pancreatitis

are the "acute" complications of trichobezoar [10, 11]. Among the three patients one underwent surgery due to trichobezoar as urgency procedure due to partial intestinal obstruction.

Surgical treatment in trichotillomania is reserved for complicated cases of trichophagia. Among available treatment methods laparotomy remains a treatment of choice, especially in case of large trichobezoars. Endoscopy, laparoscopy and enzymatic treatment are also noted, as well as experimental methods i.e. laser dissolvement of gastric mass [13–16]. Endoscopic removal of trichobezoar is mostly limited to smaller trichobezoars which are possible to remove without fragmentation. Fragmentation during endoscopy carries a risk of complications such as perforation of the gastric wall and intestinal obstruction [14]. Trichobezoars can be removed during laparoscopy. Gaster is visualised with the laparoscope, the walls are open and fragmented bezoar can be removed. Skin trauma restriction possibility is the advantage of this method. Disadvantages are longer operation time and possible abdominal cavity contamination with bezoar fragments. In case of large trichobezoar removal enlargement of the skin incision is sometimes required [14–16].

In most cases treatment of choice is laparotomy with gastric and in some cases – Rapunzel syndrome – small bowel wall opening and hair removal. This method allows for removal of gastric mass of any size and lower intestines inspection as well as minimises the risk of abdominal cavity contamination with hair. Enzymatic fragmentation of gastric bezoars with digestive enzymes (chymopapain, cellulase, acetylcysteine) have shown low efficacy [13]. In this material laparotomy was a method of choice, in one patient after failed endoscopic trial.

Main treatment of trichotillomania is psychotherapy with pharmacotherapy [17]. All patients who underwent surgery in our centre were transferred to psychiatric treatment.

Accurate data regarding recurrence rate and efficacy of treatment are lacking. Efficacy of specific methods in acquiring response to treatment and long lasting effectiveness is estimated to be 50–80% [1, 5, 8, 18].

Recapitulation

1. Due to rarity of the disease diagnosis of trichophagia is often concluded after formation of trichobezoar.
2. Surgical removal or trichobezoar is considered to be complication treatment in trichophagia.
3. Until now control scheme of patients with trichotillomania is lacking.

Three cases of trichobezoar treated in our centre in a relatively short time were inspiration to create a follow up scheme of patients with trichotillomania.

Endoscopy of the upper gastrointestinal tract should be performed in all patients diagnosed with trichotillomania. If hair is not visualised, next endoscopy should take place in 12 months. If there is no hair in second endoscopy another one is not necessary.

If hair is visualised in the gastrointestinal tract during endoscopy, it should be removed and another endoscopy should be performed in 6 months. If in the second endoscopy no hair is found in the stomach or intestines, next endoscopies should be scheduled 12 and 24 months from previous screening. In case of finding hair in any of the screening endoscopies next one should be performed after 6 months and the scheme should be repeated (12 and 24 months from previous endoscopies in case that no hair is found).

In patients who underwent surgery due to trichobezoar control endoscopy should be scheduled after 6 months and next after 12 and 24 month from previous ones. If hair is visualised in any of the screening endoscopies next one should be scheduled after 6 months.

In any moment of the follow up and treatment of patients with trichotillomania, symptoms like anaemia, weight loss, malabsorption, abdominal mass should alarm the physician, as they suggest hair accumulation in the gastrointestinal tract. In such a case additional endoscopic evaluation of the upper gastrointestinal tract is necessary.

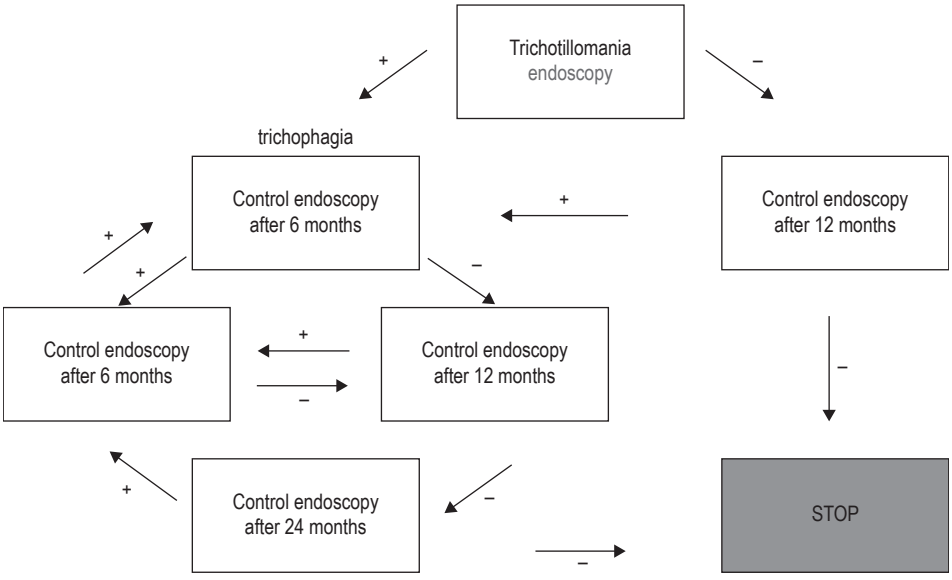

Figure 4. **Endoscopic follow-up scheme in patients with trichotillomania**

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