Case Reports in Gastroenterology

Case Rep Gastroenterol 2024;18:28–38
DOI: 10.1159/000534548
Received: January 18, 2023
Accepted: October 6, 2023
Published online: January 19, 2024

Single Case

Trichobezoar Found Accidently while Diagnosing Resistance to Thyroid Hormone

Kinga Szopińska[a] Karolina Tracz[a] Żaneta Malczyk[b] Anna Jarzumbek[b]
Andrzej Grabowski[c] Katarzyna Bąk-Drabik[b]

[a]Faculty of Medical Sciences in Zabrze, Students Association, Medical University of Silesia, Katowice, Poland; [b]Department of Paediatrics, Faculty of Medical Sciences in Zabrze, Medical University of Silesia, Katowice, Poland; [c]Department of Children's Developmental Defects Surgery and Traumatology, Medical University of Silesia, Katowice, Poland

Keywords

Abdominal pain · Bezoar · Trichobezoar · Trichotillomania · Abdominal mass · Resistance to thyroid hormone · Complications · Foreign body

Abstract

Introduction: Bezoars are masses of indigestible foreign material in the gastrointestinal tract, usually in the stomach. These materials could be indigestible fruits, hair, milk products, or tablets. In children, the most common type of bezoar is trichobezoar (formed from hair). ***Case Presentation:*** We describe a female patient who has been complaining about deterioration of mood, collapse without losing consciousness, scotomas, and cardiac arrhythmia for 2 years. Based on the results of thyroid hormone, resistance to thyroid hormone (RTH) was suspected. Physical examination during hospitalization revealed a palpable upper abdominal mass. Several diagnostic examinations were performed. The abdominal ultrasound showed acoustic shadowing caused by a pathological structure in the upper abdomen. Therefore, the contrast X-ray of the digestive tract revealed a deficit of contrast with an irregular shape in the stomach body and the pylorus region. Due to these results, a gastroscopy was performed, which revealed a large trichobezoar of the stomach. The trichobezoar was surgically removed without complications. ***Conclusion:*** The case presented shows that these nonspecific symptoms and laboratory test suggesting RTH require multi-path diagnostics and the cooperation of many specialists, ultimately giving a surprising diagnosis. It is crucial to interpret diagnostic examinations with regard to the patient's physical condition. Diagnosis of trichobezoar requires a detailed search of causes to avoid another incident.

Correspondence to:
Katarzyna Bąk-Drabik, katarzyna.drabik@sum.edu.pl

Case Rep Gastroenterol 2024;18:28–38
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Introduction

Bezoars are masses of indigestible foreign material in the gastrointestinal tract, usually in the stomach. They can be formed from different indigestible materials swallowed intentionally or accidentally. There are various types of bezoars, including phytobezoars (formed from plant material such as skins, seeds, or fibers), pharmacobezoars (formed from medications), lactobezoars (formed from milk protein), and trichobezoars (formed from hair) [1]. The most common type of bezoar in children is trichobezoar [2, 3], found mainly among girls [4]. It is usually located in the stomach. If it extends to the small intestine and colon, it is known as Rapunzel syndrome [1, 3, 5–7]. There is a strong connection with psychiatric comorbidities such as trichophagia, trichotillomania, obsessive-compulsive disorder, or depression [8, 9]. Trichobezoars may not be recognized in their early stages due to their nonspecific presentation or even lack of symptoms [2, 3, 9, 10].

Treatment of trichobezoar is the surgical removal of the mass preferable by a minimally invasive approach. However, in large masses, open surgery is inevitable [1, 8, 9]. If the bezoar is small, dissolution using enzyme therapy is also an option [9, 11].

We describe the case of a 12-year-old girl diagnosed due to suspicion of resistance to thyroid hormone (RTH), based on the entire clinical picture and the results of laboratory tests. Trichobezoar was diagnosed by accident, due to a detailed physical examination and an in-depth interview.

Case Description

An adolescent female patient, aged 12 years, with a suspicion of RTH came to the endocrinology ward for a further diagnostic evaluation. For 2 years, she has suffered from mood deterioration, collapsing without losing consciousness, scotomas, and cardiac arrhythmia. The patient underwent a cardiology consultation, and no abnormalities were found. A routine blood test was performed twice in outpatient care. Thyroid function test showed a non-suppressed serum thyroid-stimulating hormone (TSH) despite a normal or elevated free triiodothyronine (FT3) and free thyroxine (FT4 level) (Table 1).

Due to upper abdominal pain that persisted for approximately 2 months, she had a gastroenterology consultation. On the basis of the *Helicobacter pylori* antigen in the stool, eradication treatment, including amoxicillin, metronidazole, and proton pump inhibitors, was applied. Abdominal pain subsided after this treatment.

On admission, the girl's general condition was good. She appeared alert, oriented, and cooperative. No abnormalities were found in the respiratory and cardiovascular systems. The only abnormality in the physical examination was a poorly palpable mass of the upper abdomen approximately 5 cm in diameter, without palpation abdominal tenderness, with normal peristaltic sound. During examination, no signs of hyperthyroidism were revealed, including tachycardia and thyroid enlargement.

The patient's weight was 45.3 kg (50th to 75th percentile) and height was 155.6 cm (25th to 50th percentile). The development of secondary sex characteristics was assessed according to the Tanner scale M4 P4 Ax3.

The results of routine laboratory tests and ECG were normal. Thyroid function tests were repeated on the ward and showed elevated levels of FT3 and FT4 and normal TSH (Table 2). She had low antibody titers of anti-thyroid peroxide and thyrotropin receptor antibody. On ultrasound examination, the thyroid gland was in a typical location and had a homogeneous appearance and normal size. Based on the whole clinical picture and the results of laboratory tests, thyroid hormone resistance was suspected. A more in-depth medical history of the

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Table 1. Thyroid function tests before hospitalization

Date	TSH, μIU/mL (N: 0.51–4.3)	FT3, pmol/L (N: 3.84–6.06)	FT4, ng/dL (N: 0.93–1.7)
Nov 2019	0.84	4.61	2.52
Feb 2020	1.23	4.66	2.48

TSH, thyroid-stimulating hormone; FT3, free tri-iodothyronine; FT4, free thyroxine.

patient revealed that her mother had intermittent tachycardia, and her laboratory tests showed elevated FT3 and FT4 and normal TSH. The patient underwent molecular genetic tests, but pathogenic variants were not found in the analyzed regions of the thyroid hormone receptor beta (THRB) gene.

Due to the poorly palpable mass in the upper abdomen, the diagnostic had to be expanded. Abdominal ultrasound showed acoustic shadowing caused by a pathological structure in the upper abdomen approximately 5 cm in diameter (Fig. 1). Next, a contrast X-ray of the digestive tract showed smoothing of the gastric mucosa and a contrast deficit with an irregular shape and blurred lines in the stomach body and the pylorus region (Fig. 2–4). Trichobezoar or lymphoma was suspected. Thus, a gastroscopy was performed that revealed a large trichobezoar occupying the whole stomach (Fig. 5).

Psychological consultation was suggested. The mother of the patient told the psychologist that she had been playing with her hair for 2 years, including putting it in her mouth while watching TV and reading books. At this time, she also had a nail-biting habit. Surprisingly, she was unaware that she had eaten the hair. Since the transition to online classes (during the COVID-19 pandemic), she has stopped this activity, probably due to less stressful situations. On account of these symptoms, the psychologist suggested mood disorders and further observation. After discharge from the hospital, the patient was prescribed cognitive-behavioral therapy (BT).

After a surgical consultation, scheduled admission to the department of surgery was recommended as there were no indications for urgent intervention. A month later, a minilaparotomy was performed with laparoscopy under general anesthesia. The operation started with laparoscopy during which the gastric wall was prepared and then retrieved through a widened abdominal incision. Then, the wall was cut and the hair was removed consecutively. After complete removal of the trichobezoar, gastric wall was closed in double layers and the ventricle was put back into the peritoneal cavity. Trichobezoar was removed without complications (Fig. 6).

All patient's symptoms resolved after trichobezoar removal except for periodic nausea. After discharge from the hospital, genetic diagnostics extended to next-generation sequencing. No pathogenic or potentially pathogenic variants were found in the study. Further genetic diagnosis depended on the results of thyroid control hormones. Because thyroid hormone level after discharge was normal, no further genetic diagnostics was needed.

Discussion

RTH is a rare inherited condition associated with reduced responsiveness of target tissues to thyroid hormones [12, 13]. It is usually caused by mutations in the β form of the thyroid hormone receptor (TRβ), particularly receptor TRβ2; mutations of TRα1 and TRβ1 receptors are less common [13].

The incidence of the syndrome is estimated to be 1:40,000–1:50,000 live births. In patients with RTH, 80% of cases are inherited in an autosomal dominant manner. The de novo

Case Rep Gastroenterol 2024;18:28–38
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Table 2. Thyroid function tests during hospitalization

Date	TSH, μIU/mL (N: 0.51–4.3)	FT3, pmol/L (N: 3.84–6.06)	FT4, ng/dL (N: 0.93–1.7)
17 Jul 2020	1.17	–	2.1
21 Jul 2020	0.994	6.56	1.76

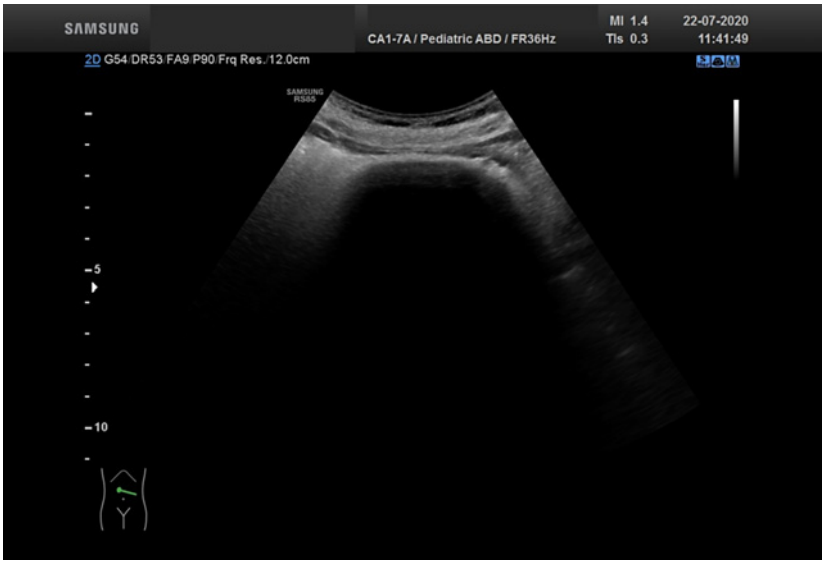

Fig. 1. Abdominal ultrasound with acoustic shadowing.

mutation rate is estimated at 20% [12]. The clinical presentation of this condition is non-specific, characterized by variable symptoms resulting from differential expression of individual receptor isoforms in the body. TRβ1 is the predominant isoform in the liver and kidneys, while TRβ2 expression is restricted principally to the pituitary gland. TRα1 is especially abundant in the myocardium, central nervous system, bones, and skeletal muscles. Thus, mutations in isoform β may result in tachycardia [12, 13].

Based on the elevated levels of FT3 and FT4 and normal TSH in our patient, the RTH was suspected. But the molecular test of our patient performed during the hospitalization did not confirm the autosomal dominant RTH. After discharge from the hospital, diagnostic extended to next-generation sequencing without showing pathogenic or potentially pathogenic variants. Surprisingly, multiple thyroid hormone measurements in the girl after discharge from the hospital were correct. The symptoms reported by the child have also subsided. Of the symptoms remained only periodic nausea.

Trichobezoars rarely occur in everyday clinical work. They were discovered in 1779 during an autopsy of a patient who died of gastric perforation [10]. Nowadays, they are described in the surgical literature, and fewer cases are found in psychiatric sources [14]. Trichobezoars can appear asymptomatic or cause a variety of symptoms and signs from the gastrointestinal tract, such as nausea, vomiting, flatulence, loss of appetite, abdominal pain, halitosis, or ulceration that leads to bleeding and/or perforation [2, 10]. They are reported to cause subacute or acute gastric obstruction, ulceration, or even perforation [2, 7, 10]. Bezoars can also cause anemia, bloody or tarry stool, hemoemesis, and fainting [2].

Case Rep Gastroenterol 2024;18:28–38
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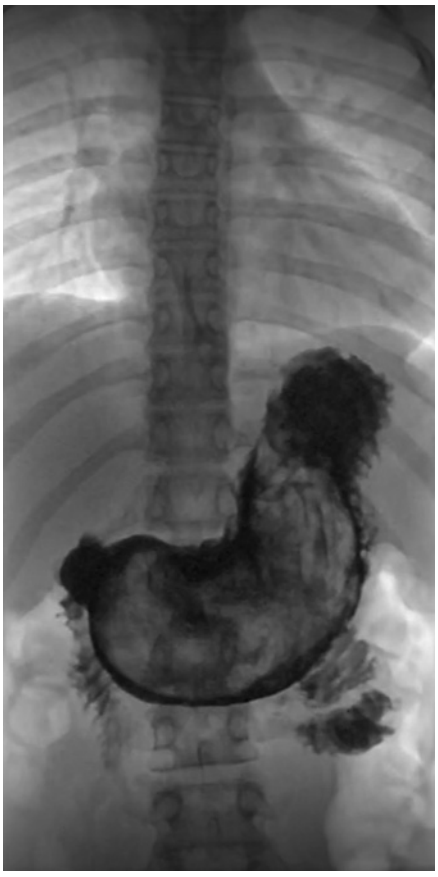

Fig. 2. Contrast X-ray. AP projection. It shows a mass that causes a deficit of contrast in the stomach.

In our patient, none of the symptoms described above was observed. There was only abdominal pain in the history, but it subsided after eradication of *H. pylori*. It may have been caused by proton pump inhibitor, which had healed a possible inflammation or ulceration of the stomach by inhibiting hydrochloric acid.

Other patient's symptoms such as mood deterioration, collapsing without losing consciousness, scotomas, and cardiac arrhythmia have resolved. They could be caused by trichobezoar physical presence but also they could be the manifestation of stress and psychological disorders.

Trichobezoars are formed because hair is a smooth and indigestible material. It makes them resistant to peristaltic movements, so they are affected in gastric folds. They are mixed in the stomach with mucous and food elements and are usually compact as trichobezoar, which adapts to the shape of the stomach [3]. Sometimes, they can extend to the duodenum and intestine, called Rapunzel syndrome [10]. The tail in the intestine can trigger peristaltic movements that result in colicky abdominal pain or obstruction [3, 7]. In our case, we did not observe that condition.

During physical examination, a palpable mass can be found in the epigastric region, but only when a large amount of hair creates a solid mass [7]. The findings of the clinical examination with trichobezoar are diverse and depend on the stage at which the bezoar is formed [3]. Most trichobezoars remain asymptomatic during the initial stages [14]. It may lead to delay in diagnosis or even misdiagnosis. In the case we report, a poorly palpable mass in the upper abdomen was found.

Generally, bezoars are palpable as well-delimited and movable tumors within the left epigastrium. For this reason, the differential diagnosis should cover liver lobe

Case Rep Gastroenterol 2024;18:28–38
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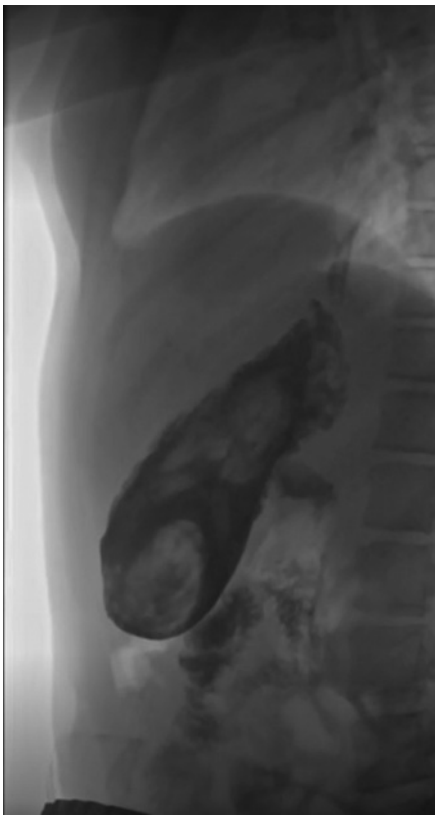

Fig. 3. Contrast X-ray. Lateral projection.

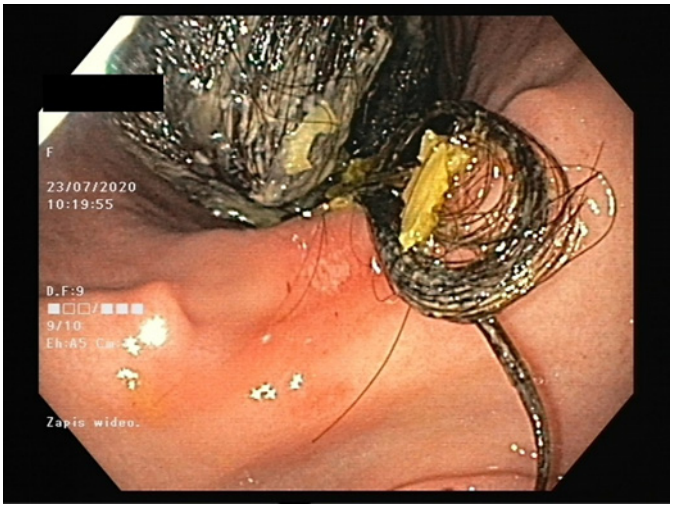

Fig. 4. Gastroscopy image. Large trichobezoar in the stomach.

tumors, splenomegaly, neuroblastoma, and gastric tumors [15]. Additionally, firm or hard masses in the abdomen during physical examination are usually suspected of being a malignant process, especially in adults or the elderly. But in young women and children, trichobezoar should always be considered in differentiation, especially when they have a history of psychiatric disorders and complain of indigestion or abdominal pain [10].

Case Rep Gastroenterol 2024;18:28–38
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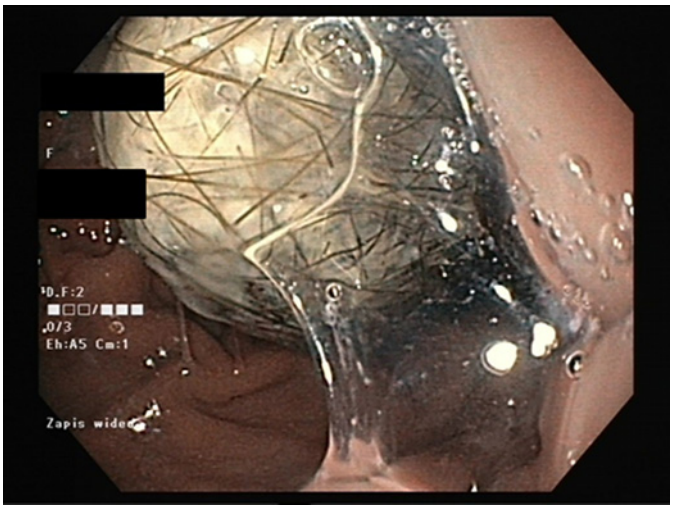

Fig. 5. Gastroscopy image.

In our case, there was no clear evidence of the problem in the gastrointestinal tract; the patient complained of collapse, mood deterioration, and scotomas. Only abdominal pain in the medical history and proper physical examination were signs that could guide to a successful diagnosis. That is why precise medical interviews and questions are crucial in such cases.

The etiology of the formation of trichobezoar is multicausal. Most cases of trichobezoars occur in women under 30 years of age [3]. The most common causes of trichobezoar described in the literature are trichophagia and trichotillomania. They occur in 1–2% of the population [3]. Three types of trichotillomania have been identified: early or infancy-onset, automatic, or focused. The first typically affects children up to 8 years of age and resolves without treatment or after psychotherapy. In automatic trichotillomania, the patient unknowingly pulls out the hair without paying attention. "Focused" trichotillomania is different; it is associated with powerful emotions and typically manifests in menopausal women. A trichobezoar may also form due to habitual, not psychologically conditioned, chewing of hair ends. Furthermore, patients with trichobezoar may be affected by psychological disorders and the associated trichotillomania at the time of symptoms, act intermittently, or have only a history of events [2, 3]. Several factors may predispose to the formation of trichobezoar: gender, mental retardation, excessive ingestion of food or non-food substances, gastric dysmotility, and an underlying behavioral disorder leading to pica [7, 10].

The case we are referring to here showed a few characteristic symptoms. Information that the patient was playing with her hair while watching TV or reading helped us establish that she was suffering from automatic trichotillomania. She was unconscious of eating hair. This behavior could be explained as a result of ongoing stress and poor coping skills, because it was lessened when the patient started online lessons.

As described above, clinical symptoms related to trichobezoars are characterized by late onset and lack of specificity, leading to late diagnosis. As a result, diagnostic procedures are commonly performed due to acute abdominal symptoms or abnormalities in abdominal palpation [14].

A conclusive diagnosis is made based on medical imaging, including radiography, abdominal ultrasound, computed tomography (CT), endoscopy, or even magnetic resonance imaging. The first option, usually the cheapest and most accessible in every hospital, is ultrasound. But it is of limited value in the diagnosis of trichobezoar [15]. Typically, examination reveals a lesion with hyperechoic reflections. However, the presence of numerous

Case Rep Gastroenterol 2024;18:28–38
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Fig. 6. Postoperative image of the trichobezoar.

acoustic shadows because of air bubbles trapped between the hair and the high echogenicity of the hair limits its assessment and description capabilities. There is a strong possibility that an ultrasound scan would not recognize a trichobezoar. In the reported case, an ultrasound showed acoustic shadowing, which, in correlation with a palpable mass in the upper abdomen, indicated the need to extend the diagnostic evaluation.

Radiological imaging also plays a crucial role in the diagnostic approach. Although plain radiograms of the abdominal cavity usually reveal a soft tissue mass in an enlarged stomach, they can also show levels of airflow that could suggest intestinal obstruction [2, 15]. A preferable choice is the contrast-enhanced scan of the upper gastrointestinal tract, using barium drank by the patient. The image may show a mass within a stomach, separated from the wall of the stomach. That examination might help differentiate the disorder or make the decision about the next steps. In the case of our patient, only a contrast radiograph led to suspicion of trichobezoar.

CT scanning is undoubtedly the best tool in the diagnostics of trichobezoars. It is the radiological method of choice, characterized by high sensitivity (up to 90%) and specificity (up to 60%) [2]. The image is quite specific, consisting of a well-delimited, nonenhanced mass filling the stomach, with a spotted pattern due to air being trapped between hairs. CT scans allow for the estimation of tumor size, location, and potential complications [15].

Endoscopy usually makes an unequivocal diagnosis by visualizing the hair mass, which appears black due to the denaturation of the hair proteins by stomach acid. In addition, it facilitates the evaluation of the type of bezoar and the existing complications [3, 10]. In the case of our patient, several imaging studies were performed, starting with an ultrasound and finishing with an endoscopy, which confirmed the diagnosis, showing a large hair mass in the stomach.

Treatment options include endoscopic, laparoscopic, or laparotomy removal [10]. Paschos, K.A. and Chatzigeorgiadis, A. proposed a treatment scheme based on the volume of trichobezoar, chemical synthesis, location, and associated pathology. For example, for bezoars located in the stomach, the first choice treatment should be endoscopic removal; however, trichobezoars usually have very hard consistency and require surgical procedures [2, 3].

Bezoar removal by open surgery has been well established for decades [2]. It has been described as a method with a 100% success rate and a low risk of comorbidities. In addition, a

complex and careful examination of both the intestine and the stomach prevents secondary intestinal obstruction due to satellites in a shorter time [8].

The advent of laparoscopic surgery, due to excellent cosmetic results and perfect visualization, has become a promising alternative [16]. Despite smaller incisions, fewer complications, and shorter admission times, it is still limited in large bezoars [2, 3, 17]. In addition, successful laparoscopic removal is difficult to achieve due to the complexity of the operation and its rarity [2, 8]. Thus, laparotomy remains the standard surgical approach for most trichobezoars [17].

In our case, the patient underwent a laparoscopy with a mini-laparotomy. All patient's symptoms resolved after trichobezoar removal except for periodic nausea.

For prevention of recurrence, trichotillomania should be treated if present [3]. Wolski et al. [11] proposed a scheme of endoscopic control of patients with trichotillomania. They recommended performing an endoscopic examination of the upper gastrointestinal tract in all patients diagnosed with trichotillomania. In patients after surgical removal of trichobezoar, a follow-up endoscopic examination should be scheduled after 6 months and another after 12 and 24 months since the last examination. If residual hair is found in the gastrointestinal tract in any endoscopic examination, the next follow-up should be scheduled in 6 months.

The follow-up gastroscopy was not performed in our patient due to lack of symptoms and the COVID-19 pandemic. It was an extremely difficult time for a non-COVID-19 hospitalization. But the control gastroscopy is scheduled in coming months.

Psychiatric treatment is an inherent part of follow-up after surgical removal of trichobezoar to avoid another accident. It includes both medications and psychotherapy [3, 14]. Drugs such as selective serotonin reuptake inhibitors, olanzapine, clomipramine, Nacetylcysteine, naltrexone, topiramate, dronabinol, and atypical neuroleptics are effective [14, 18]. Psychotherapy adds efficacy to pharmacotherapy. BT and cognitive therapy have been reported to be beneficial for patients. Currently, cognitive-BT is indicated to be the most empirically validated treatment for trichotillomania, appearing to be more effective than pharmacological treatment [18]. Cognitive-BT and psychological care were recommended to the patient in question. Pharmacological treatment was not essential at that stage. However, the patient did not undergo treatment. Nevertheless, her mother claimed she is watching her to avoid playing with her hair and eating it unconsciously.

Conclusions

The presented case shows that an interview with the patient and a physical examination are key to effective diagnosis and treatment. Trichobezoars require multi-way diagnostics and the cooperation of many specialists. It is crucial to remember that bezoars are complex problems, usually associated with other disorders. Therefore, the patient needs extended treatment and observation to avoid another incident. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000534548).

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the parent of the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

This research was funded by the Medical University of Silesia, Grant No. PCN-1-171/K/0/K.

Author Contributions

Kinga Szopińska: an author, substantial contributions to the conception or design of the work, final approval of the version to be published, and wrote the largest share of the report. Karolina Tracz: co-author, substantial contributions to the conception or design of the work. Żaneta Malczyk, Anna Jarzumbek, and Andrzej Grabowski: co-author, attending physicians overseeing the care of the patient described in the case and substantial contributions to the conception or design of the work. Katarzyna Bąk-Drabik: senior author, an attending physician overseeing the care of the patient described in the case.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author. Contact: katarzyna.drabik@sum.edu.pl.

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