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■ Case report ■

A case of gastric trichobezoar causing psychiatric problems

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= Abstract =

Trichobezoar is characterized by the accumulation of hair in the gastrointestinal tract and usually occurs in those who have trichotillomania, other psychiatric disorders, or neurologic problems. Trichobezoar typically presents as gastric obstruction, including abdominal pain, vomiting, anorexia, and weight loss. A 9-year-old girl visited our clinic with the complaint of abdominal discomfort and vomiting. A review of her medical history revealed that she had trichophagia since the age of 5, and she felt that her parents had been strict with her. She underwent gastrotomy, during which a large trichobezoar was removed. This case highlights the importance of psychiatric and comprehensive approaches in patients with trichobezoar. (Korean J Pediatr 2009;52:1167-1170)

Key Words : Trichobezoar, Trichophagia, Psychiatric problem

Introduction

Gastric trichobezoar is uncommon in childhood and generally results from emotional instability, neurologic or psychiatric problems particularly including trichotillomania, a compulsive behavior of eating hairs. And trichobezoar is associated with iron deficiency anemia, which often results in pica[1].

The most common presenting symptoms of trichobezoar include palpable abdominal mass, vomiting and noticeable hair loss. The methods in the history taking, which revealed a possibility of the trichotillomania and trichobezoar by looking into a sense of the uneasiness and the relationship between their parents, or brothers, and sisters, will be greatly required to the patients those who suffer abdominal pain and psychopathic disease. Furthermore, if a patient presents with gastointestinal symptoms, abdominal palpation must be performed during physical examination.

Five to 18% of the patients with trichotillomania ingest hairs. The incidence of trichobezoars in trichotillomania, however, is unclear[2]. Of those whom engage in trichophagy, approximately 37.5% are at risk of forming a trichobezoar[3].

The main current in the reports about bezoar is chiefly phytobezoar in contrast with the papers on trichobezoar. The fatality rate of trichobezor may be up to 30% of diagnosed patients primarily because of gastrointestinal bleeding, obstruction or perforation[4]. Even so, trichobezoars have failed to attract a scholastic interest in the field of pediatrics. There are only a few reports of trichobezoars.

We report a case of a giant trichobezoar in a 9-year-old girl who had a psychiatric disorder.

Case report

A 9-year-old girl visited our clinic because of abdominal discomfort and vomiting (4 times episodes a day), worsened after meals. The symptoms started 1 month before and became aggravated through 3 days before the visit. She had no history of previous surgery. Her medical history was notable for trichophagia since 5 years of age. She had a habit of pulling her brother's hair out. She had no other medical history and was not on any medication.

There was no family history of hair-pulling or of obsessive-compulsive disorder. However, the patient's mother suffered from depression in the past. And also, she felt that her parent was strict.

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Her social development was unremarkable and her school performance has been lagged behind. She was treated discriminatingly by her parents comparing to her elder brother.

On the admission, she was afebrile, with a stable vital sign, and there was no evidence of the hair loss from the scalp. Her height and weight were 131.0 cm (25–50 percentile), and 23 kg (10–25 percentile). On physical examination, she looked acutely ill and did not appear to be in toxicated. The abdomen was notable of two moderate, tender and firm masses detected in the mid-epigastrium (sized each 10×7 cm, 5×5 cm) without peritoneal sign. The routine laboratory findings, including a complete blood cell count, serum electrolyte, and liver and renal function test, were all within normal range and as were amylase and lipase.

Fig. 1. Unenhanced abdominal computed tomography (CT) image shows an ovoid intraluminal mass with mottled gas pattern within the stomach.

Fig. 2. A large trichobezoar weighing 105.5 g and measuring 14.7×8.7×4.2 cm.

Abdominal radiographs showed no free air, no air fluid level, but unusual gas pattern in the upper quadrant, with a rim of air inside was noticeable. Abdominal computed tomography (CT) showed a large and ovoid mass with air retained in the intestines within the stomach with a massive space-occupying lesion (Fig. 1). The diagnosis of trichobezoar was made and the patient was brought to the operating room where transverse gastrostomy was performed. A gastrostomy incision was made in the anterior wall of the stomach, and a large trichobezoar was removed. The mass weighed 105.5 gm and measured 14.7×8.7×4.2 cm (Fig. 2). There was no extension of the hair into the duodenum. We could not make endoscopic intervention for removal of the bezor (which has recently performed) due to lack of support of specialists and instruments.

After the operation, her abdominal symptoms were disappeared. She was discharged 8 days later and followed as an outpatient. She was seen in a consultation with a psychiatrist, but the parents refused psychiatric treatment repeatedly.

Discussion

Bezoars are persistent retained concretions of the undigested foreign bodies that accumulate in the gastrointestinal tract. Interestingly, the term "bezoar" is derived from Arabic *badzehr* or from Persian *panzehr*, both meaning counterpoison or antidote[4] respectively. Bezoar is divided into 4 types that are lactobezoars (milk or formula), phytobezoars (plant fiber), pharmacobezoars (pills or capsules), and trichobezoars (hair). Trichobezoar are formed by ingesting a many numbers of hairs, Debakey and Ochsner[5] reported that 80% of the patients were under 30 years of age, and 90% of the cases were female.

Tichobezoar usually result from trichotillomania (a behavioral disorder characterized by the compulsive urge to pull one's hair) combined with trichophagia (the ingestion of hair). Moreover, trichobezoar is associated with an iron deficiency anemia, which often results in pica (the compulsion to ingest non-nutritious substances). And pica may manifest, by itself, as trichotillomania and trichobezoar. It might be an evidence for the relationship between pica and trichobozoar that trichobezoar is found more in high risk factors for pica including female gender, childhood, mental retardation, and African-American and Aboriginal race[6].

Our patient suffered from trichotillomina (pulling out hairs of her older brother) and subsequent trichophagia (ingestion of hairs of her older brother, her own, and unidentified hairs in her house) - since 5 years of age.

Another important predisposition to bezoar formation is alteration of gastrointestinal tract anatomy or disordered gastrointestinal motility and physiology. Examples of the disease conditions, in which these can be found include, post-partial gastrectomy, post-vagotomy, diabetes mellitus with gastroparesis, Guillain-Barre's syndrome, myotonic dystrophy, hypothyroidism, cretinism, psychiatric illness and edentulous patient[7, 8].

The clinical feature depends on which part of the gastrointestinal tract it originates from. Esophageal involvement presents the clinical signs such as dysphagia, odynophagia, reflux, retrosternal pain and halitosis, while gastric involvement dose subtle abdominal pain, nausea, post-prandial vomiting, bloating, ulceration, gastrointestinal bleeding and secondary anemia, and easy fullness, and small bowel involvement dose partial or complete intestinal obstruction with or without perforation[8]. However, a left upper quadrant/epigastric mass is the most common finding.

The most frequently arising complication is intestinal obstruction which can occur in approximately 10% of cases, usually in the ileocecal area. In addition to intestinal obstruction, it may lead to other complications such as ulceration, perforation and peritonitis. The cause of such complications has not been identified. It is assumed that a persistent stimulation to intestinal mucosal walls might induce ulceration or protein-losing enteropathy. Additionally transient pancreatitis, intussusception, hypoproteinemia and megaloblastic anemia associated with vitamin B12 deficiency have been reported.

The diagnosis of bezoar may be presumptive through a plane abdominal radiograph, and confirmed with barium swallowing, ultrasonograph, endoscopy or abdominal CT (computed tomography). Main ultrasonic feature is a hyperechoic arch like surface with a marked acoustic shadowing. Endoscopy is the diagnostic technique of the choice for gastric and esophageal bezoars and has therapeutic potentails. In this case, the abdominal CT played the essential role in diagnosis. Sometimes, it would not be easy to perform barium swallowing or endoscopy to children. Such disadvantage of barium swallowing and ultrasonograpy may make it difficult to differentiate a bezoar from a intraluminal mass[9]. Accordingly, we recognize that abdominal CT can be used to diagnose bezoars.

The trichobezoar presents on CT as a large inhomogeneous intraluminal mass occupying almost the entire lumen. A characteristic finding on CT appears that various sizes of the minute air bubbles filled masses, which is a helpful sign for diagnosis. These masses are not attached to the gastric wall. Despite the presence of satellite distal mass, CT can also provide a valuable information about therapeutic determination - extension degree of trichobezoar into the gastrointestinal tract and size.

CT has a major role in the evaluation of gastrointestinal obstruction, and has some important advantages of distinguishing between a bezoar and other cause of the intraluminal mass such as lobulated tumor, gallstone ileus or feces[10]. Therefore diagnosis of the gastric trichobezoar can be made confidently with an abdominal CT.

Therapy for any bezoar necessitates removal and prevention of recurrence. Treatment for gastric bezoar is gastrotomy or endoscopic removal. During operative treatment, there must be a through exploration of the rest of the small intestine to check for retained bezoars. Despite the rarity of obstructing small intestinal trichobezoars, whole small intestine should be observed carefully. That is why such size of other bezoar increase that small intestine could be obstructed. Endoscopic retrieval and fragmentation are frequently used for proximal bezoars whose size and density are not prohibitive; however, the procedure can be technically challenging, and fragments may migrate distally and cause a small bowel obstruction[11]. And emergent surgical treatment may be required. In order to prevent the recurrence of the bezoars, we prihibit the ingestion of the foods rich in vegetable fiber.

This case magnifies the importance of psychiatric evaluation early in the course of trichobezoar management, particularly in the pediatric age group. Trichotillomania, as mentioned above, is a psychiatric disorder related with formation of trichobezoar. According to a study, trichobezoar is found in more than a third of trichotillomania[12]. Physicians should be aware that a trichobezoar may be present even in patients who show no signs of the hair loss. Furthermore, the family history must be attentively examined. A family history of wheather other family members have any disorders, alcohol dependence or attention deficit hyperactivity disorder (ADHD) should be taken. There was a study using genogram that trichotillomania could occur to members of the family with these psychiatric disorders[13].

Behavior therapy and habit-reversal therapy have been reported to be effective. Habit-reversal therapy includes the technique in which the patient monitors the undesirable behaviors and substitutes a desired behavior for the problem behavior. For children, supportive family counseling has also been reported to be effective[14, 15].

After trichobezoar removal, prognosis is good, if psychiatric therapy to control habitual trichophagia is successful. To prevent recurrences, psychiatric follow-up is needed for treatment of the underlying behavioral disorder presenting in up to 90% of patients[16].

Without a thorough attention to the complete history, the diagnosis of coeliac disease could be overlooked in the presence of another distracting diagnosis. Thus a bezoar should be considered in the differential diagnosis in a child with symptoms of the gastrointestinal obstruction and painless upper abdominal mass. Also, if children complaining of abdominal pain present psychiatric problems, hair loss, alopecia or pale appearance, an abdominal examination should be considered. The multimodal approaches using pediatricians, surgeons and psychiatrists are emphasized for an effective management of the pediatric patient with trichobezoar.

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정신적 문제로 인한 털위석 1례

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털위석은 위장관에 털이 축적되어 생기며, 발모광이나 다른 정신적인 문제나 신경학적인 문제가 있는 환자들에게서 주로 발생한다. 털위석은 복통, 식욕부진, 구토 같은 위장관 폐쇄의 증상들로 나타난다. 국내외에서 위석의 보고는 식물위석에 대한 것이 대부분이며, 털위석이 소아에게서 보고된 경우는 많지 않다. 9세 여아가 복부 불쾌감과 복부 종괴로 내원하였다. 환아는 5년 전부터 머리카락을 주어먹는 버릇이 있었고 부모로부터 오빠에 대한 차별적인 대우를 받는다고 생각하였으며 학교에서 성적은 뒤처지는 편이지만 그 외 특별한 병력은 없었다. 복부 컴퓨터단층촬영술을 시행하여 위 안에 거대한 털위석을 확인하였으며, 위절개술을 시행하여 털위석을 제거하였다. 그 후 정신과적인 상담과 치료를 권유하였으나 보호자의 거부로 중단되었다. 이에 저자들은 정신적인 문제가 수반된 환아가 복부 증상으로 내원시 털위석의 유무에 대한 접근과 진단의 중요성을 강조하는 바이다.

References

1) Osman YM, Wali YA, Osman OM. Craving for ice and iron-deficiency anemia: A case series from Oman. Pediatr Hematol Oncol 2005;22:127-31.
2) Bouwer C, Stein DJ. Trichobezoars in trichotillomania: case report and literature overview. Psychosom Med 1998;60:659-60.
3) Christenson GA, Crow SJ. The characterization and treatment of trichotillomania. Clin Psychiatry 1996;57:42-7.
4) Williams RS. The fascinating history of bezoars. Med J Aust 1986;145:613-4.
5) DeBakey M, Ochsner A. Bezoars and concretions: a compulsive review of the literature with analysis of 303 collected cases and a presentation of 8 additional cases. Surgery 1939;5:132-60.
6) Bhatia MS, Singhal PK, Rastogi V, Dhar NK, Nigam VR, Taneja SB. Clinical profile to trichotillomania. J Indian Med Assoc 1991;89:137-9.
7) Andrus CH, Ponsky JL. Bezoars: classification, pathophysiology, and treatment. Am J Gastroenterol 1988;83:476-8.
8) Ibuowo AA, Saad A, Okonkwo T. Giant gastric trichobezoar in a young female. Int J Surg 2008;6:e4-6.
9) Gayer G, Jonas T, Apter S, Zissin R, Katz M, Katz R, et al. Bezoars in the stomach and small bowel - CT appearance. Clin Radiol 1999;54:228-32.
10) Ko SF, Lee TY, Ng SH. Small bowel obstruction due to phytobezoar: CT diagnosis. Abdomen Imag 1997;22:471-3.
11) Diettrich NA, Gau FC. Postgastrectomy phytobezoar - endoscopic diagnosis and treatment. Arch Surg 1985;120:432-5.
12) Bouwer C, Stein DJ. Trichobezoars in trichotillomania: Case report and literature overview. Psychosom Med 1998;60:658-60.
13) Carr JR, Sholevar EH, Baron DA. Trichotillomania and trichobezoar: A clinical practice insight with report of illustrative case. J Am Osteopath Assoc 2006;11:647-52.
14) Tay Y, Levy M, Metry D. Trichotillomania in childhood: case series and review. Pediatrics 2004;113:e494-8.
15) Salaam K, Carr J, Grewal H, Sholevar E, Baron D. Untreated trichotillomania and trichophagia: Surgical emergency in a teenage girl. Psychosomatics 2005;46:362-6.
16) Assevero VL, Brooks DA, Cardozo WW, Gerson EF. Trichobezoar as an expression of emotional disturbance. AMA J Dis Child 1957;94:668-71.