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Systematic review of Rapunzel Syndrome

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RESUMEN

Revisión sistemática del Síndrome de Rapunzel

Introducción. El síndrome de Rapunzel involucra una forma inusual de tricobezoar gástrico que se localiza desde el estómago llegando a traspasar el píloro y extendiéndose hasta el intestino delgado y/o colon derecho. El tricobezoar, uno de los bezoares que más se presentan en la práctica clínica, consiste en una masa formada por cabello debido a la ingestión de este, ya sea de manera deliberada o no intencionada.

Objetivo. Describir la frecuencia de signos y síntomas del síndrome de Rapunzel en los reportes de caso y presentar una comparación entre las edades pediátrica y adulta.

Método. Revisión sistemática en las bases de datos PubMed, Google académico, EBSCO y Scopus, empleando los términos "Rapunzel syndrome" y "case report" o "clinical case".

Resultados. Un total de 110 casos de 98 artículos de reporte de caso fueron analizados, encontrando diferencias por sexo y edad. La tricotilomanía se distribuyó sin predominancia por grupo de edad. Se identificaron diferencias en el reporte de algunas manifestaciones clínicas al analizarlas por grupo de edad, incluyendo diarrea, constipación, vómitos y dolor abdominal. Los casos recidivantes representaron un 9 %.

Conclusiones. El síndrome de Rapunzel es una entidad que se encuentra en pacientes con patologías psiquiátricas como la tricotilomanía y la tricofagia. Es más frecuente en mujeres que en hombres. La técnica diagnóstica por elección fue la endoscopía y el tratamiento elegido incluyó la remoción quirúrgica y la psicoterapia.

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ABSTRACT

Introduction. Rapunzel syndrome involves an unusual form of gastric trichobezoar located from the stomach into the pylorus and extending to the small intestine, and or right colon. The trichobezoar, one of the bezoars that most occur in clinical practice, consists of a mass formed by hair due to ingestion of it either deliberately or unintentionally.
Objective. Describe the frequency of signs and symptoms of Rapunzel syndrome in published case reports and present a comparison between the condition at pediatric and adult ages.
Method. A systematic review in the PubMed database, EBSCO, and Scopus, using the terms "Rapunzel syndrome" AND "case report" OR "clinical case."
Results. A total of 110 cases of 98 case report articles were analyzed. Differences between sex and age were found. Trichotillomania was distributed without predominance by age group. Also, differences in the reporting of some clinical manifestations analyzed by age group, including diarrhea, constipation, vomiting, and abdominal pain, were found. Recurrent cases represented 9%.
Conclusions. Rapunzel syndrome is an entity found in patients with psychiatric disorders such as trichotillomania and trichophagia. It is more common in women than in men. Endoscopy was the chosen diagnostic technique, and the treatment selected included surgical removal and psychotherapy.

Keywords. Rapunzel syndrome, bezoar, diagnosis, trichotillomania.

INTRODUCTION

Rapunzel syndrome was first described in 1968 by Dr. Vaughan; the syndrome name refers to the fairy tale written by the Grimm brothers about Princess Rapunzel, who let her hair fall through the window of the tower so that his prince could climb to rescue her. (1) Rapunzel syndrome is an unusual form of gastric trichobezoar, which is formed by fractions of hair. The resultant bezoar can extend throughout the intestine. In this syndrome, trichophagia and trichotillomania can be observed with depressive, anxiety and particularly, obsessive-compulsive disorders (2).

Rapunzel syndrome occurs mainly in young women in 90% of cases, of which 80% are under 30 years of age. (3) Trichobezoar can be asymptomatic until it has grown enough to generate symptoms of gastrointestinal obstruction, such as recurrent abdominal pain, nausea, vomiting, anorexia, weight loss, diarrhea, constipation, jaundice and malabsorption of trace elements. When this condition is not diagnosed, gastric bezoars may cause anemia, either due to malabsorption or gastrointestinal bleeding. (1, 4) Additionally, gastrointestinal obstruction, peritonitis, perforation, gastrointestinal bleeding, intussusception, pancreaticobiliary obstruction, pancreatitis, and complications due to malabsorption are complications reported of this syndrome (1).

The diagnosis and prognosis of Rapunzel syndrome can be modified in the transition from pediatric age to adulthood (5), which is consistent with the ontogenetic period of establishment of other psychiatric disorders (6, 7), particularly in women. Based on this, the aim of this study was described the frequency of signs and symptoms in case reports and establishing a comparison between the clinical characteristics in the pediatric population and adults, using as a cut-off point the age of 15 years.

MATERIALS AND METHODS

A systematic review of the medical literature on Rapunzel syndrome was conducted; signs, symptoms, and complications were included. PubMed, EBSCO, and Scopus databases were systematically searched with terms: "Rapunzel syndrome" AND "case report" OR "clinical case" that included a description of the patient. All case report studies of Rapunzel syndrome, available in English or Spanish (with summary in English), were selected. Exclusion criteria were the lack of clinical data, the absence of a digital version, and articles published in languages other than English, or Spanish.

The data entry of signs, symptoms, complications, age (as a binary variable with a cut-off point

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at 15 years), and sex were captured in Excel. Subsequently, columns were generated to identify the presence or absence of signs, symptoms, and complications. Descriptive statistics were applied to obtain the measures of central tendency to present the frequencies by age group, the means and standard deviation of the numerical variables, as well as the percentages and proportions of the dichotomous variables. The statistical significance of the difference between groups was evaluated through hypothesis testing, using the chi-square (c^2) test to compare proportions and Student's t-test for the comparison of means. The statistical significance was established for a threshold of P-value < 0.05, and all statistical analyses were carried out with Stata 14 software.

RESULTS

A total of 133 articles were initially selected, which 98 met the inclusion criteria. We excluded 12 studies because they were clinical or radiographic images without a description of the patient, four studies because they were not online and were not obtained by correspondence with the author, and nine because they were written in another language. Ten articles were duplicated in different versions; when duplicates were removed, 110 cases of Rapunzel syndrome were collected from the 98 studies. The most outstanding data of the publications are shown in **Table 1**.

Table 1. Literature review of signs, symptoms, and complications in the cases reviewed with Rapunzel syndrome diagnosis (n=110).

Reference No.	Reference	Sex	Age	Signs-Symptoms	Complications	Result
1	Nettikadan *et al*., 2018 *	Female	19	Trichobezoar, trichophagia, abdominal pain, nausea, vomiting, anorexia, palpable mass, paleness of teguments, anemia.	Gastrointestinal obstruction.	Alive
2	Appak *et al*., 2018	Female	12	Trichobezoar, tricophagia, trichotillomania, palpable mass.	Gastrointestinal obstruction.	Alive
3	Bargas *et al*., 2018	Female	14	Trichobezoar, tricophagia, trichotillomania, abdominal pain, gastric satiety, nausea, palpable mass, alopecia, onychophagia, diarrhea, constipation, weight loss.	Gastrointestinal obstruction.	Alive
4	Cannalire *et al*., 2018	Female	14	Trichobezoar, tricophagia, abdominal pain, gastric satiety nausea, vomiting, anorexia, palpable mass, alopecia, paleness of teguments, anemia, weight loss, splenomegaly.	Gastrointestinal obstruction, Upper Gastrointestinal Bleeding, pleural effusion.	Alive
5	Frey *et al*., 2005	Female	7	Trichobezoar, tricophagia, trichotillomania, palpable mass, paleness of teguments.	Gastrointestinal obstruction.	Alive
9	Obinwa *et al*., 2017*	Female	25	Trichobezoar, tricophagia, abdominal pain, gastric satiety, palpable mass, anemia.	Gastrointestinal obstruction, gastric ulcer, pleural effusion, lung infection.	Alive
15	Naik *et al*., 2007	Female	21	Trichobezoar, tricophagia, abdominal pain.	Gastrointestinal obstruction, peritonitis, perforation, intussusception.	Alive
15	Naik *et al*., 2007	Female	18	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting.	Gastrointestinal obstruction, perforation.	Alive

15	Naik *et al*., 2007	Female	16	Trichobezoar, trichotillomania, vomiting, anorexia, palpable mass, anemia.	Gastrointestinal obstruction.	Alive
16	Nour *et al.*, 2017	Female	4	Trichobezoar, tricophagia, abdominal pain, palpable mass, paleness.	Gastrointestinal obstruction.	Alive
17	Kohler *et al*., 2012	Male	16	Trichophytobezoar, tricophagia, abdominal pain, Trichophytobezoar.	Pancreatitis.	Alive
18	Dindyal *et al*., 2008	Male	55	Trichobezoar, abdominal pain, nausea, palpable mass, alopecia, fever, constipation.	Gastrointestinal obstruction, perforation.	Alive
19	Jain *et al*., 2011	Male	3	Trichobezoar, palpable mass, Trichobezoar	Gastrointestinal obstruction.	Alive
20	Emre *et al*., 2008	Male	18	Cotton bezoar, Trichobezoar, abdominal pain, nausea, vomiting, gastrointestinal bleeding, melena, anemia, fever, weight loss.	Splenomegaly, Gastrointestinal obstruction, ulcers, lung infection.	Dead
21	Hirugade *et al*., 2001	Male	6	Trichobezoar, tricophagia, abdominal pain, vomiting, anorexia, palpable abdominal mass, weight loss.	Gastrointestinal obstruction.	Alive
22	Sarin, 1998	Female	1	Phytobezoar, vomiting, palpable abdominal mass, anemia, dehydration, diarrhea.	Gastrointestinal obstruction.	Dead
29	Gonuguntla and Joshi, 2009	Female	5	Trichobezoar, tricophagia, abdominal pain, gastric satiety, vomiting, anorexia, palpable mass.	Gastrointestinal obstruction.	Alive
30	Jones *et al*., 2010*	Female	37	Trichobezoar, tricophagia, trichotillomania, abdominal pain, nausea, vomiting, weight loss.	Intestinal obstruction.	Alive
31	Matějů *et al*., 2009	Female	3	Trichobezoar, trichotillomania, vomiting, alopecia, paleness.	Gastrointestinal obstruction, lung infection.	Dead
34	Hassan and Panesar, 1989 *	Female	8	Wool bezoar, trichotillomania, abdominal pain, nausea, vomiting, diarrhea.	Gastrointestinal obstruction, intussusception intestinal.	Alive
35	Balik, Ulman, Taneli, and Demircan, 1993	Female	15	Trichobezoar, tricophagia, abdominal pain, vomiting, anorexia, palpable abdominal mass.	None reported.	Alive
36	Şeker, 1996	Female	6	Trichobezoar, trichotillomania, nausea, vomiting, palpable abdominal mass, weight loss.	Gastrointestinal obstruction.	Alive
37	Pul and Pul, 1996	Female	12	Trichobezoar, vomiting, anorexia, palpable abdominal mass, constipation, weight loss.	Gastrointestinal obstruction, perforation.	Alive
38	Bonnet, El Arbi, Chelly, and Girodet, 1996	Female	11	Trichobezoar, tricophagia, vomiting, anorexia, palpable abdominal mass, weight loss.	Gastrointestinal obstruction.	Alive
39	Senapati and Subramanian, 1997	Female	8	Trichobezoar, abdominal pain, vomiting, Hematemesis, anorexia, palpable abdominal mass, paleness of teguments, anemia, fever, dehydration, weight loss.	Gastrointestinal obstruction.	Alive

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40	West and Duncan, 1998	Female	5	Trichobezoar, tricophagia, trichotillomania, abdominal pain, anorexia, palpable mass, alopecia, weight loss.	Gastrointestinal obstruction.	Alive
41	Singla, Rattan, Kaushik, and Pandit, 1999	Female	9	Trichobezoar, tricophagia, abdominal pain, vomiting, anorexia, palpable abdominal mass.	Gastrointestinal obstruction.	Alive
42	Dalshaug, Wainer, and Hollaar, 1999	Female	7	Trichobezoar, trichotillomania, abdominal pain, nausea, vomiting, diarrhea.	Gastrointestinal obstruction.	Alive
43	Faria, Silva, Santos, Avilla, and Silveira, 2000	Female	7	Trichobezoar, tricophagia, abdominal pain, vomiting, alopecia, paleness of teguments, anemia.	Gastrointestinal obstruction, peritonitis, perforation.	Alive
44	Uçkun, Sipahi, Igde, Üner, and Çakmak, 2001	Female	13	Trichobezoar, trichotillomania, abdominal pain, vomiting, palpable abdominal mass.	Gastrointestinal obstruction.	Alive
45	Curioso Vilchez, Rivera Vega, Abriojo, and Walter, 2002	Female	22	Trichobezoar, abdominal pain, nausea, anorexia, palpable abdominal mass, paleness of teguments, asthenia, weight loss.	Gastrointestinal obstruction,	Alive
46	Memon, Mandhan, Qureshi, and Shairani, 2003*	Female	12	Trichobezoar, trichotillomania, abdominal pain, nausea, palpable abdominal mass, paleness of teguments, anemia.	None reported.	Alive
47	Klipfel, Kessler, and Schein, 2003	Female	14	Trichobezoar, abdominal pain, vomiting.	Gastrointestinal obstruction.	Alive
48	Durkhure, Singh, and Singhal, 2003	Female	35	Cotton bezoar, abdominal pain, vomiting, anorexia, palpable mass, paleness, anemia, constipation, weight loss.	Gastrointestinal obstruction.	Alive
51	Ventura *et al*., 2005	Female	5	Trichobezoar	Peritonitis, perforation, death.	Dead
49	Pérez *et al*., 2005	Female	16	Trichobezoar, trichotillomania, abdominal pain, nausea, anorexia, asthenia, anemia, fever, weight loss.	Gastrointestinal obstruction, perforation.	Alive
50	Maldonado, 2005	Female	23	Trichobezoar, tricophagia, abdominal pain, gastric satiety, nausea, vomiting, anorexia, palpable mass, halitosis, paleness of teguments, anemia, weight loss.	Gastrointestinal obstruction, perforation.	Alive
51	Durán Ferreras, López Bernal, Martínez Vieira, Álamo Martínez, and Docobo Durántez, 2005	Female	12	Trichobezoar, abdominal pain, vomiting, anorexia, palpable mass, asthenia, anemia, weight loss.	Gastrointestinal obstruction.	Alive
52	Mathai *et al*., 2007	Female	5	Trichobezoar, tricophagia, abdominal pain, anorexia, palpable mass, alopecia, paleness of teguments, anemia, weight loss.	Gastrointestinal obstruction.	Alive
52	Mathai *et al*., 2007	Female	10	Trichobezoar, anorexia, palpable mass, weight loss.	Gastrointestinal obstruction.	Alive

53	Henry, Nair, Jemila, and Krishna, 2007	Female	10	Jute fiber bezoar, abdominal pain, vomiting, palpable mass, anemia, dehydration, constipation, weight loss.	Gastrointestinal obstruction,	
54	Rabie *et al*., 2008	Female	11	Trichobezoar, abdominal pain, gastric satiety, vomiting, palpable mass, dehydration.	Gastrointestinal obstruction, gastrointestinal ulcer.	Alive
4	Rabie *et al*., 2008	Female	19	Trichobezoar, abdominal pain, vomiting, anorexia, palpable mass, anemia, dehydration, constipation, weight loss.	Gastrointestinal obstruction, perforation, intussusception.	Alive
55	Western, Bokhari, and Gould, 2008	Female	14	Trichobezoar, tricophagia, abdominal pain, nausea, vomiting, anorexia, palpable mass, asthenia, anemia.	None reported	Alive
56	Mohite, Gohil, Wala, and Vaza, 2008	Female	28	Trichobezoar, abdominal pain, palpable mass, edema.	Gastrointestinal obstruction, perforation.	Alive
57	Anzieta *et al*., 2008	Female	16	Trichobezoar, trichotillomania, abdominal pain, vomiting, dehydration, weight loss.	Gastrointestinal obstruction.	Alive
58	Al Wadan, Al Kaff, Al Senabani, and Al Saadi, 2008	Female	7	Trichobezoar, abdominal pain, vomiting, palpable mass, weight loss.	Gastrointestinal obstruction.	Alive
59	Mehta and Bhutiani, 2009	Female	14	Trichobezoar, abdominal pain, vomiting, anorexia, palpable mass, alopecia, anemia, dehydration.	Gastrointestinal obstruction.	Alive
60	Agrawal, Joshi, Jain, and Gupta, 2009	Female	16	Plastic bezoar, abdominal pain, vomiting, palpable mass, paleness of teguments, constipation.	Gastrointestinal obstruction, perforation, volvulus.	Alive
60	Agrawal *et al*., 2009	Female	13	Trichobezoar, tricophagia, abdominal pain, palpable mass.	None reported.	Alive
61	Vila *et al*., 2009	Female	16	Trichobezoar, abdominal pain, nausea, vomiting, palpable mass, paleness of teguments, anemia, weight loss.	Gastrointestinal obstruction.	Alive
62	Koç, Yıldız, Narcı, and Şen, 2009	Female	14	Trichobezoar, abdominal pain, anorexia, palpable abdominal mass, anemia, weight loss.	Gastrointestinal obstruction, perforation.	Alive
63	Hernández-Peredo-Rezk *et al*., 2009	Female	12	Trichobezoar, abdominal pain, vomiting.	Gastrointestinal obstruction.	Alive
63	Hernández-Peredo-Rezk *et al*., 2009	Female	7	Trichobezoar, tricophagia, abdominal pain, palpable abdominal mass, alopecia.	None reported	Alive
64	Lopes *et al*., 2010	Female	22	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, alopecia, paleness of teguments, weight loss.	Gastrointestinal obstruction, intussusception, perforation.	Alive
65	Rajakannu, 2010	Female	15	Trichobezoar, tricophagia, trichotillomania, palpable abdominal mass, alopecia.	Gastrointestinal obstruction.	Alive

66	Raikar, Wali, and Khan, 2010 *	Female	12	Trichobezoar, tricophagia, abdominal pain, vomiting.	Gastrointestinal obstruction.	Alive
67	Chogle *et al*., 2010	Female	3	Trichobezoar, tricophagia, abdominal pain, vomiting, asthenia, fever, jaundice, weight loss.	Intussusception.	Alive
68	Tayyem, Ilyas, Smith, and Pickford, 2010	Female	23	Trichobezoar, abdominal pain, vomiting, hematemesis, paleness of teguments, dehydration, constipation, weight loss.	Gastrointestinal obstruction, peritonitis, perforation, gastric ulcer.	Alive
69	Patel, Abubacker, and Nour, 2010	Female	13	Trichobezoar, tricophagia, palpable abdominal mass, alopecia, anemia.	Gastrointestinal obstruction.	Alive
70	Gorter, Kneepkens, Mattens, Aronson, and Heij, 2010*	Female	9	Trichobezoar, tricophagia, palpable abdominal mass, weight loss.	Intussusception.	Alive
70	Gorter *et al*., 2010 *	Female	14	Trichobezoar, tricophagia, abdominal pain, vomiting.	Peritonitis, pancreatitis	Alive
70	Gorter *et al*., 2010 *	Female	15	Trichobezoar, tricophagia, abdominal pain, palpable abdominal mass, alopecia, weight loss.	Gastric ulcer.	Alive
70	Gorter *et al*., 2010 *	Female	7	Trichobezoar, tricophagia, vomiting, abdominal pain, palpable abdominal mass, anemia.	Intestinal ulcer.	Alive
71	Dorn, Gillick, and Stringel, 2010	Female	17	Trichobezoar, trichotillomania, abdominal pain, anorexia, onychophagia, weight loss.	Intestinal obstruction.	Alive
72	Dhinakar and Balkhair, 2010	Female	Non reported	Trichobezoar, abdominal pain, palpable abdominal mass.	Intestinal obstruction.	Alive
73	Bashir, Samiullah, Sadiq, Yusuf, and Karim, 2010	Female	8	Trichobezoar, abdominal pain, gastric satiety, palpable abdominal mass, alopecia, paleness, diarrhea.	Intestinal obstruction.	Alive
74	Tiwary, Kumar, Khanna, and Khanna, 2011 *	Female	10	Trichobezoar, abdominal pain, vomiting, anorexia, palpable abdominal mass, paleness, anemia.	Abdominal obstruction.	Alive
75	Singh *et al*., 2011	Female	5	Trichobezoar, abdominal pain, vomiting, palpable abdominal mass, paleness, anemia, dehydration, constipation, weight loss, edema.	Abdominal obstruction, perforation.	Alive
76	Cook, Beaver, Brislin, and Elitsur, 2011	Female	12	Trichobezoar, trichotillomania, abdominal pain, gastric satiety, vomiting, anorexia, palpable abdominal mass, alopecia, weight loss.	Abdominal obstruction, intussusception,	Alive
77	Kawoosa and Zargar, 2011	Female	12	Trichobezoar, abdominal pain, vomiting, palpable abdominal mass, weight loss.	Abdominal obstruction.	Alive

78	Mansour-Ghanaei, Herfatkar, Sedigh-Rahimabadi, Lebani-Motlagh, and Joukar, 2011	Female	19	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, palpable abdominal mass, constipation.	Abdominal obstruction.	Alive
79	Dogra, Kulkarni, and Rao, 2012	Female.	24	Trichobezoar, abdominal pain, vomiting, paleness of teguments, anemia, dehydration, constipation.	Gastrointestinal obstruction.	Alive
80	Elizabeth Middleton and Lynn Fitzgerald, 2012	Female.	2.5	Trichobezoar, abdominal pain, vomiting.	Intestinal obstruction, intussusception.	Alive
81	Ozdemir, Ozdemir, Sahiner, and Senol, 2012	Female.	13	Trichobezoar, tricophagia, trichotillomania, abdominal pain, nausea, vomiting, palpable mass, anemia.	None reported.	Alive
82	Petrović *et al*., 2012	Female.	19	Trichobezoar, trichotillomania, abdominal pain, nausea, vomiting, burning retrosternal, palpable mass, alopecia, anemia, weight loss.	Gastrointestinal obstruction.	Alive
83	Phavichitr and Vathanasanti, 2012	Female.	10	Trichobezoar, tricophagia, trichotillomania, palpable mass.	Gastrointestinal obstruction.	Alive
84	Tiago *et al*., 2012*	Female.	27	Trichobezoar, abdominal pain, nausea, vomiting, palpable mass, anemia, weight loss.	Gastrointestinal obstruction, gastric ulcer.	Alive
85	Neychev, Famiglietti, and Saldinger, 2013*	Female	26	Trichobezoar, abdominal pain, nausea, palpable mass, constipation.	Gastrointestinal obstruction.	Alive
86	Veloso *et al*., 2013	Female	7	Trichobezoar, abdominal pain, vomiting, palpable mass.	Intestinal obstruction.	Alive
87	Belsky, Whitcomb, Zimmerman, and Stankovic, 2014	Female	6	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, constipation.	Gastrointestinal obstruction.	Alive
88	Germani, Beltrá Picó, and Hernández Castelló, 2014	Female	8	Trichobezoar, abdominal pain, nausea, anorexia.	Gastrointestinal obstruction.	Alive
89	İslek, Sayar, Yilmaz, Boneval, and Artan, 2014	Female	5	Trichobezoar, vomiting, palpable mass, anemia, weight loss.	Gastrointestinal obstruction.	Alive
90	Marwah, Pandey, Raj, Jangra, and Sharma, 2015	Female	15	Trichobezoar, tricophagia, abdominal pain, vomiting, palpable mass.	Gastrointestinal obstruction, intussusception.	Alive
91	Adhikari, Vankipuram, Tiwari, Chaphekar, and Satardey, 2015	Female	29	Trichobezoar, abdominal pain, vomiting, fever.	Gastrointestinal obstruction.	Alive
92	Zeraatian, Ameri, Tabesh, and Kamalzadeh, 2015	Female	13	Trichobezoar, abdominal pain, gastric satiety, vomiting, anorexia, palpable mass, paleness of teguments, anemia, dehydration, weight loss.	Gastrointestinal obstruction.	Alive
93	Czerwińska, Bekiesińska-Figatowska, Brzewski, Gogolewski, and Wolski, 2015	Female	16	Trichobezoar, tricophagia, trichotillomania, palpable mass.	Gastrointestinal obstruction.	Alive

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93	Czerwińska *et al*., 2015	Female	13	Trichobezoar, tricophagia, palpable mass, alopecia, anemia.	None reported.	Alive
93	Czerwińska *et al*., 2015	Female	15	Hair and plaster, abdominal pain, palpable mass, alopecia, anemia, weight loss.	None reported.	Alive
94	Wolski *et al*., 2016	Female	13	Trichobezoar, abdominal pain, alopecia, anemia, weight loss.	Gastric ulcer.	Alive
94	Wolski *et al*., 2016	Female	16	Trichobezoar, trichotillomania, abdominal pain, palpable mass.	None reported.	Alive
94	Wolski *et al*., 2016	Female	15	Trichobezoar, abdominal pain, alopecia, anemia, weight loss.	None reported.	Alive
95	Kim, Kim, and Kim, 2016	Female	8	Trichobezoar, tricophagia, trichotillomania, abdominal pain, palpable mass, alopecia, anemia, weight loss.	Abdominal obstruction.	Alive
96	Guzmán Rojas, Bravo Paredes, and Pichilingue Reto, 2016	Female	16	Trichobezoar, tricophagia, abdominal pain, gastric satiety, nausea, vomiting.	Gastrointestinal obstruction.	Alive
97	Parakh, McAvoy, and Corless, 2016	Female	18	Trichobezoar, tricophagia, abdominal pain, nausea, vomiting, weight loss.	Gastrointestinal obstruction, perforation.	Alive
98	Ahmed, Baloch, Baber, and Ahmed, 2016	Female	13	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, palpable mass, paleness of teguments, anemia, constipation.	Gastrointestinal obstruction, perforation, intussusception.	Alive
99	Kumar, Thippeswamy, and Rangaswamaiah, 2016	Female	15	Trichobezoar, tricophagia, trichotillomania, abdominal pain, nausea, vomiting, palpable mass, alopecia.	Gastrointestinal obstruction.	Alive
100	Dixit, Raza, and Tiwari, 2016	Female	20	Trichobezoar, tricophagia, abdominal pain, vomiting, palpable mass.	Gastrointestinal obstruction.	Alive
101	Chauhan, Kumar, and Bhoil, 2016	Female	19	Trichobezoar, tricophagia, abdominal pain, vomiting, anorexia, palpable mass, constipation, weight loss.	Gastrointestinal obstruction.	Alive
102	Caiazzo, Di Lascio, Crocoli, and Del Prete, 2016	Female	9	Trichobezoar, tricophagia, abdominal pain, gastric satiety, nausea, vomiting, anorexia, palpable mass.	Gastrointestinal obstruction.	Alive
103	Beristain-Silva, Cordero-Barberena, and Beristain-Hernández, 2016	Female	10	Trichobezoar, tricophagia, abdominal pain, nausea, vomiting, palpable mass, paleness of teguments, anemia, dehydration, weight loss.	Gastrointestinal obstruction.	Alive
104	Yik and How, 2016	Female	13	Trichobezoar, trichotillomania, abdominal pain, vomiting, anorexia, palpable mass, alopecia, paleness, anemia.	Gastrointestinal obstruction, intussusception.	
105	Nwankwo *et al*., 2017	Female	7	Trichobezoar, tricophagia, trichotillomania, abdominal pain, nausea, vomiting, palpable mass.	Gastrointestinal obstruction.	Alive
106	Lalith, Gopalakrishnan, Ilangovan, and Jayajothi, 2017	Female	12	Trichobezoar, abdominal pain, vomiting, anorexia, palpable mass, paleness of teguments, asthenia, anemia, dehydration, constipation.	Gastrointestinal obstruction.	Alive

107	Iwama, Nambu, and Hara, 2018	Female	13	Trichobezoar, tricophagia, trichotillomania, abdominal pain.	None reported.	Alive
108	Hamid *et al*., 2017	Female	16	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, palpable mass, onychophagia, paleness of teguments, anemia, weight loss.	Gastrointestinal obstruction.	Alive
110	Nour *et al*, 2019	Female	4	Trichobezoar, tricophagia, abdominal pain, palpable mass, paleness.	None reported.	Alive
111	Kumar *et al*, 2019	Female	19	Trichobezoar, tricophagia, trichotillomania, abdominal pain, vomiting, hematemesis, palpable mass, paleness, anemia.	Gastrointestinal obstruction, perforation, gastric ulcer, intussusception.	Alive
112	Soria *et al*, 2019	Female	16	Trichobezoar, tricophagia, trichotillomania, abdominal pain, gastric satiety, nausea, palpable mass, paleness, weight loss.	Perforation	Alive

*Recurrent cases

Of the 110 cases included for appraisal and data extraction, 95% (105) occurred in female and 5% in male ($p < 0.001$). The age range was between one and 55 years; 72% were <15-years-old (n = 65), and the remaining cases were ≥15-years-old (n = 45).

The selection criteria bezoar was reported in all cases. Bezoars exclusively composed of hair, known as trichobezoar were found in 95% of the (n = 104); while in the remaining 5% (n = 6) bezoars were also formed by plaster, plastic, and wool or jute fibers. Bezoars of materials other than hair fibers were more frequent in the group of patients <15 years. The trichophagia was reported in 47% (n = 51), while trichotillomania in 33% (n = 37) of the cases reported.

Gastrointestinal signs and symptoms were the most frequent. Abdominal pain, palpable mass, and vomiting were the three most reported in descending order. The frequency of abdominal pain was 83% (n = 91) in the general population, and significantly more frequent in the group <15 years ($p = 0.003$). Additionally, a mass could be palpated in 72% (n = 79) of the cases, which 77% (n = 50) was found in the <15 years group, and there was a significant difference between the two groups studied $p = 0.048$. **Table 2** shows the signs and symptoms found in the sample studied.

Table 2. Signs and symptoms by age group reported in the cases included for the systematic review (n=110)

Variable	Total	<15 years (n=65)		≥ 15 years (n=45)		***P***
		Percentage	n	Percentage	n	
Trichobezoar	110	100	65	100	45	1.000
Tricophagia	54	51	33	46	21	0.405
Trichotillomania	37	31	20	38	17	0.258
Abdominal pain	91	75	49	93	42	0.003*
Gastric satiety	13	14	9	9	4	0.188
Nausea*	28	19	12*	36	16*	0.015*
Vomiting	73	68	44	64	29	0.379
Burning retrosternal	2	0	0	4	2	0.107

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Hematemesis	4	3	2	4	2	0.381
Melena	2	0	0	4	2	0.107
Anorexia	34	34	22	27	12	0.234
Palpable mass*	79	77	50*	64	29*	0.048*
Alopecia	26	26	17	20	9	0.237
Onychophagia	5	3	2	7	3	0.166
Halitosis	1	0	0	2	1	0.107
Paleness	30	29	19	24	11	0.414
Asthenia	7	8	5	4	2	0.373
Anemia	42	40	26	36	16	0.46
Fever	7	5	3	9	4	0.082
Dehydration	15	15	10	11	5	0.374
Jaundice	2	3	2	0	0	0.208
Diarrhea*	5	8	5	0	0	0.049*
Constipation*	18	11	7	24	11	0.039*
Weight loss	47	39	25	49	22	0.133
Splenomegaly	2	2	1	2	1	0.381
Edema	3	2	1	4	2	0.166

* significant values

Constipation was found in 16% (n = 17) of the patients in the sample, but when data were analyzed by age, constipation was more frequent in >15 years group (p = 0.039). The feeling of early satiety was present in 11% (n = 13) of the sample. Hematemesis was reported in 2 patients <15 years and 2 cases in patients ≥15 years. Alopecia was the most frequent sign (23.6%, n = 26).

The complications most frequent in descending order were: gastrointestinal obstruction (80.9%, n = 89), gastrointestinal perforation (19%, n = 21), intussusception (10%, n = 13), gastrointestinal ulcers (10%, n = 11) and peritonitis (5%, n = 6.3) (**Table 3**).

Table 3. Complications of Rapunzel syndrome by age group in the cases included for the systematic review (n = 110)

Variable	Total	<15 years (n=65)		≥ 15 years (n=45)		*p*
		Percentage	n	Percentage	n	
Obstruction GI	89	79	51	84	38	0.164
Peritonitis	7	6	4	7	3	0.493
Perforation	21	15	10	24	11	0.100
UGB	4	3	2	4	2	0.381
Ulcer GI	11	8	5	13	6	0.307
Intussusception	13	12	8	11	5	0.408
Volvulus	1	0	0	2	1	0.107
Pancreatitis	3	3	2	2	1	0.381
Pleural effusion	3	3	2	2	1	0.381
Lung infection	4	3	2	4	2	0.166

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Death	4	5	3	2	1	0.271
Recurrence	10	8	5	11	5	0.154

GI: gastrointestinal, UGB: Upper Gastrointestinal Bleeding

Four cases out of 110 resulted in the death of the patient (4%). Most cases were in the group <15 years (5%, n = 3) and all were female. A single case was reported in the group >15 years and was male. 8% of the cases were recurrent (n = 10), and all of them occurred in female.

DISCUSSION

Rapunzel syndrome is a trichobezoar that extends from the stomach to the small intestine. This syndrome is frequently in young women with mental disorders such as trichotillomania, which could be accompanied by trichophagia (8). Trichotillomania is defined as the irresistible desire of pull out the hair and is included in the manual of statistical diagnosis (DSM-5) of the American Psychiatric Association since 2013, as an obsessive-compulsive disorder (9). Trichotillomania is often associated with trichophagia, which is defined as the act of eating hair (10). According to Bryant-Waugh, in patients in whom this type of disorder has been reported, the presence of dissatisfaction with self-perceived appearance, feelings of shame, as well as low self-esteem, anxiety disorders, and the need for self-rewards have been identified. (11) In some patients, it has been reported limitation to establish social relationships, feelings of isolation, loneliness, and alienation. (12) In 82% of cases, patients suffer from other psychiatric morbidities and problems associated with the abuse of alcohol and other substances (13).

Trichobezoars are rare and usually are present in patients that more likely to have underlying psychiatric conditions (14). In a review conducted in 2007 by Naik et al., 28 cases of Rapunzel syndrome (27 women and one man) were evaluated, with an age range of four and 22 years (15). Nour et al. (2017) reported four cases of Rapunzel syndrome in male patients (16), although the presence of trichobezoar was greater in females, with only five cases reported in males (17-21). The age ranged between one to 55 years. According to Dindyal et al., the male population is located in older ages, while the female population in younger ages (22).

The presence of the trichobezoar in the female sex has been attributed to the fact that women and girls have long hair, which tends to get tangled up, fall off and get swallowed, forming the trichobezoars. Similarly, this could be since trichotillomania is much more common in women than in men and could be the reason for the difference between sexes (23, 24).

Trichotillomania is rare as an isolated disorder; occasionally it is part of a broader spectrum of repetitive behaviors focused on the body, involving repetitive self-stimulatory activities such as: pulling, tearing, biting or scratching hair, skin or nails, resulting in injuries corporal (25). It is estimated that the frequency of this phenomenon is 1.5% for men and 3.4% for women (10). In the present review, trichotillomania was found in 33% of the cases, of them, 100% was female. This disproportion between sexes increases with age, and it is estimated that in adults the female: male ratio is higher than 10:1 (26). Trichotillomania presents a peak incidence between 4 and 17 years, and the average age of the revelation of symptoms is between 11 and 13 years (13, 27). In this review, patients diagnosed with Rapunzel syndrome range from one to 55 years. The number of reported cases that presented trichotillomania was higher in the group <15 years (31%, n = 20), which is consistent with the incidence peak mentioned above. It is believed that the early onset of symptoms is usually associated with a mild course and with a better prognosis, while a later start is associated with more severe symptoms, resistance to treatment and higher morbidity (28).

It is estimated that 5 to 20% of cases of trichotillomania are accompanied by trichophagia. Case reports of children with trichobezoars or

Rapunzel syndrome are rare, and it has been associated trichophagia to neglect or abuse during childhood, psychiatric conditions, mental retardation, or grief (29). Of the 110 cases reviewed, only 50% mentioned the presence of trichophagia. The difference between the number of cases between trichotillomania and trichophagia could be associated to the fact that trichotillomania is an entity of difficult diagnosis since patients usually do not come for help and deny having any problem when they come to the consultation. Similarly, difficulties and inaccurate diagnostic criteria complicate the correct identification (24, 25). Therefore, the fact that trichotillomania is not reported in a case of Rapunzel syndrome does not exclude its presence.

Failure to treat trichotillomania leads to social, occupational, and other deterioration of the patient's functioning. Long-term complications can result in general deterioration of health and, occasionally, the need for surgical intervention (26). Signs, symptoms, and complications emerge when the trichobezoar begins to form. This occurs when the consumed hair is retained in the folds of the gastric mucosa. This hair is retained in the stomach because these are indigestible and the stomach mucosa is slippery, which prevents the propulsion of this material by peristalsis. The more hairs are added, the peristalsis causes the formation of a too large mass to leave the stomach and eventually forming a gastric mold (30).

In the present review, gastrointestinal obstruction was the most frequent complication. Symptoms include recurrent abdominal pain, nausea, vomiting, anorexia, weight loss, and malabsorption of trace elements. When not correctly diagnosed, gastric bezoars can cause severe anemia, either due to malabsorption, or gastrointestinal bleeding (4). In a retrospective study conducted by Abou-Shady (2016), the symptoms present in gastric bezoars, included malabsorption, were found in eight patients in Egypt (14).

Trichotillomania can often generate alopecic areas on the scalp; typically, patients pluck hairs from more than one area of the body. Alopecia caused by trichotillomania has different severities, from discrete to very significant (10). Out of a total of 110 cases, only 23.6% (n = 26) presented alopecia; the higher proportion was in the group <15 years. This symptom can be a diagnostic key to suspect Rapunzel syndrome (29); however, sometimes, it can present discreetly, especially when the patient does not report the symptoms of trichotillomania, which masks the condition.

In addition to gastrointestinal obstruction, there are other complications reported: peritonitis and perforation (18.3%), gastrointestinal bleeding (10%), intussusception (7.4%), pancreatobiliary obstruction, pancreatitis, and malabsorption (1).

Trichotillomania is a rare and poorly diagnosed entity, which if not treated in time, the complications generated can lead to death. Out of the 110 cases reported as Rapunzel syndrome, only four had a fatal outcome (19, 22, 31, 32). Of these, 75% (n = 3) was in women, with an age range between one and five years. The only male case was 18-years-old (20).

Twenty percent of cases of Rapunzel syndrome is recurrent in the absence of psychotherapy. Therefore, as mentioned by Bargas et al. (2018), beyond the surgical resolution, the trichobezoar should be approached integrally with psychological therapy and psychiatric counseling. If trichotillomania-trichophagia is not resolved, could result in the recurrence and formation of new trichobezoars (3). Of the cases of recurrence, the highest frequency was in the group of >15 years; out of total cases, only in one, the presence of trichotillomania was reported. Since trichotillomania is an entity challenging to diagnose, the absence of evidence of this behavior does not exclude his presence. In any case of trichotillomania, psychotherapy is the standard treatment, and the first step should always be psychoeducation (31). The method of choice is cognitive-behavioral therapy (10), and a complementary one is the habit-reversal training, which includes self-observation, mindfulness, impulse control, relaxation techniques, and social support (33). Likewise, part of the treatment must involve the patient or a member of their family in support groups.

Some limitations arose in this study, the studies included were explicitly obtained from the central PubMed, Scholar Google, Ebsco and Scopus database, and the data only included the clinical manifestations reported in the manuscript; therefore, if there were others not reported, the clinical manifestations would be underrepresented. However, it is pertinent to mention that the selection criteria to be systematized did not imply a bias in the selection of authors, as indicated by the definition of systematic reviews.

In conclusion, Rapunzel syndrome is caused by psychiatric pathologies such as trichotillomania and trichophagia, if these are not diagnosed and treated promptly, lead to the development of trichobezoars, or in case of recurrence and complications, could cause death. Since these entities are underdiagnosed, it is essential to inquire directly to women <15 years of age who are clinically compatible with symptoms to offer a diagnosis and timely treatment.

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