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Potential Prognosis and Side-Effects of Trichophagia Associated with Psychiatric, as well as Immunological Parameters

[1]Alexandros Vlachos, MD, [2*]Simon Vassiliadis, PhD

[1]Department of Pediatric Surgery, Penteli General Children's Hospital, 8 Hippocrates Street, Palaia Penteli, Athens, 15236, Greece
[2]Association of Greek Immunology Graduates, 33 Voriou Ipirou Street, Maroussi, 15125 Athens, Greece

***Corresponding Author:** Vassiliadis, S., Ph.D., Association of Greek Immunology Graduates, 33 Voriou Ipirou Street, Maroussi, 15125 Athens, Greece.*

ABSTRACT

Rapunzel syndrome (RS) is an extremely rare intestinal condition in humans resulting from ingesting hair (trichophagia) usually of psychogenic origin. It is named after the long-haired girl Rapunzel in the fairy tale by the Grimm Brothers. Since published reports point to possible relationships between trichotillomania and celiac disease -as associated condition- or trichotillomania and ileo-ileal intussusception -as side-effect-, further investigation and an in-depth patients' history has to urgently be compiled. Even though their possible connection is under intense scrutiny, it becomes obvious that elucidation of any potential association appears of pivotal importance in terms of prognosis and/or undesired side-effects.

Keywords: *Trichophagia, Bezoar, Rapunzel Syndrome, Trichotillomania, Laparotomy, Gastrotomy, Celiac Disease, Intussusception*

INTRODUCTION

Rapunzel, the eponymous heroine of a German fairy tale written by the Grimm Brothers in 1812, was a 12-year-old princess imprisoned for many years by a witch in a tall tower with neither stairs nor doors. To be rescued, she lowered her long hair from her window to the ground allowing a young prince to climb up and save her.[1]

As a metaphor, RS refers to trichotillomania and ingesting hair (trichophagia). The outcome of hair ingestion is a mass of hair accumulated in the stomach that usually extends into the small intestine termed bezoar from the Persian word "Padzahr", meaning antidote.[2] This particular feature, *i.e.* the extension (tail) that enters the small intestine, establishes the diagnosis of RS.[3,4] Trichobezoars are rarely described in the absence of trichotillomania. However, a case of trichobezoar associated with trichophagia in the absence of trichotillomania has been reported in an18-year-old thin built girl.[5]

Trichobezoars are often encountered in young women and are frequently associated with an underlying psychiatric/psychogenic disorder.[6]

Celiac disease (CD), a serious autoimmune disorder of the upper small intestines affecting 1 in 100 people worldwide, is characterized by intestinal villous atrophy and crypt hyperplasia and caused by an abnormal immune reaction to wheat gliadin.[7] As, however, the literature reports cases associating CD and trichobezoars,[8,9,10,11] an in-depth history must be taken and further lab tests should be prescribed.

By the same token, intussusception, a serious condition in which part of the intestine slides into an adjacent part of the intestine,[4] should also be suspected to occur secondary to unsuccessful surgical bezoar removal since, according to Yik and How, the small intestinal "baby" bezoar may well be the pathological lead point for intussusception.[12,4]

There is only one reported case of trichotillomania in Greece[13] and another one in our Hospital (data not published due to the incidence-free outcome) both successfully resolved surgically with an uneventful postoperative hospital stay.

GENERAL AND SURGICAL BACKGROUND

As previously reviewed,[13] bezoars can be classified into six types: (i) phytobezoars (composed of indigestible plant material), (ii) trichobezoars (hairball or hair-like fibers); more often found in children and teenage girls, (iii)

lithobezoars (fragments of small stones, pebbles or gravel stones), (iv) pharmacobezoar (mostly tablets or semiliquid masses of drugs), (v) plasticobezoars (plastic) and (vi) lactobezoars (inspissated milk). In the case of trichobezoars, the cause why hair is collected in the stomach is not entirely understood. Due to its indigestibility and resiliency, however, it becomes entrapped within the mucosal folds acquiring more hairs and becoming larger in size. As hair continues to accumulate, peristalsis causes the formation of a ball that becomes even more matted together assuming the shape of the stomach, usually as a single solid mass. Bezoars' black color is explained by the acid content of the stomach that denatures the hair protein.[14] This condition is often life-threatening and, unless psychiatric counseling is provided, can recur.[15]

RS can be managed laparoscopically[16] or with endoscopic retrieval, which is less invasive compared to surgical removal.[17] Surgical treatment, however, is most often required when a patient presents with a massive gastric mass.[15,18,19]

Discussion

Except the psychogenic causes, already mentioned above, trichotillomania has been associated to CD.[7,8,9,10] However, the connection between the two conditions has not yet established and further attention should be paid by the physicians compiling the patient's history as supplementary laboratory tests may reveal a patient's predisposition to the disease. Pertinent to this, iron deficiency anemia and pica relating to celiac disease has been presented as a probable reason for presence of trichophagia and bezoars[20,9] showing the importance of more laboratory tests that may be proven of prognostic value. This view is further strengthened by articles relating to celiac disease and bezoar as extracted by a thorough search of medical literature databases showing the potential association between the two cases.[20] Furthermore, a compilation of case reports on trichotillomania and trichobezoar with celiac disease has been presented in a recent 2019 publication[20] allowing the assumption of such relationship.

Finally, it is worth of noting that failed endoscopic or open retrieval of the entire gastroduodenal bezoars may be the cause of future ileo-ileal intussusception, a serious and often life-threatening condition if untreated.[12] Thus, complications and/or recurrences may occur if the mass is not promptly excised or the patients remain psychiatrically unattended.

Disclosure

This is an opinion paper that adds information pertinent to diagnosis or potential side-effects associated with trichophagia and its surgical removal.

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