Original Research Article

A Systematic Review and Metaanalysis of Clinical Perspective in Trichotillomania

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Abstract

Trichobezoar is a mass of hairs found trapped in the gastrointestinal system. They are common in children under the age of 2 and in children and adults living in poverty. Complications such as gastric ulceration, bleeding and perforation and intestinal obstruction may occur. There is paucity of reports on trichobezoars in Psychiatric literature however it is well described in terms of surgical diagnosis and procedure. Here, we present a case of trichotillomania and pica leading to intestinal obstruction to stress the importance of considering trichobezoars as one of the important differentials in cases of patients presenting with abdominal pain in face of suggestive symptoms, even if signs of trichotillomania are not present.

Keywords: Bezoar, Obstruction, Trichobezoar, Trichotillomania, pica.

Introduction

Pica is described as the compulsive ingestion of substances not fit as food or of no nutritional value. The most common forms of pica include geophagia (soil-like), pagophagia (ice), trichophagia (hair), and amylophagia (starch like).[1] It commonly occurs in children under the age of 2 years who actually start perceiving the world through the oral cavity. The likely devastating complication of pica is the development of a bezoar, with subsequent gastrointestinal complications such as gastric ulceration, bleeding and perforation, and small bowel obstruction. “Bezoar” is an Arabic word refering to a kind of antidote “bazahr” or “badzehr”, meaning that stones obtained from the stomach or intestines of animals were thought to have medicinal properties. Bezoars can be categorized in four types: trichobezoar (hair); lactobezoar(milk/curd); phytobezoar (vegetables) and miscellaneous (sand, paper).[7] eyelashes, eyebrows and other parts of the body. The process results in an instant release of tension, a sense of relief and security. However, non-scaring alopecia is its clinical presentation. The development of trichobezoar following ingestion of the pulled hair is its salient complication in a few cases. Subsequently, it may cause symptoms pertaining to the gastrointestinal tract culminating in intestinal obstruction, perforation, pancreatitis and obstructive jaundice. The Rapunzel syndrome (trichobezoarTrichobezoar is the most common bezoar in the pediatric age group.[2]

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Trichobezoars were first reported by Baudomant in 1779, consisting of dense mass of hair, occupying the gastric cavity to a various extent. Patients frequently present with recurrent abdominal pain and a palpable abdominal mass. The mechanism underlying is thought to be that the smooth surface of hair does not propagate further in gut peristalsis, getting trapped in the mucosa. As more hair is added, the resulting mass causes the stomach to cease peristalsis completely. The stomach of normal individuals have capacity to clear even large foreign bodies in up to 80 to 90% of the cases, which may suggest that bezoar formation occurs in the presence of both altered gastric anatomy or physiology and continued ingestion of the offending substance.[11,12] When the trichobezoar is seen extending from the stomach to various lengths of the intestine, it is called "Rapunzel Syndrome", for its resemblance to a tail. [3]

Trichobezoars are usually described in the context of an underlying psychiatric disorder, trichotillomania is commonly considered as cause of it, and usually accompanied by characteristic features such as signs of alopecia. There is paucity of reports on trichobezoars in psychiatric literature however it is well described in terms of surgical diagnosis and procedure.[4]

5% to 30% of the patients with trichotillomania engage in trichophagia while 1% to 37.5% of these will develop a trichobezoar. The explanation for such disparities may be the fact that most patients of trichotillomania primarily present to paediatric OPD, dermatology OPD and surgery OPD due to the physical consequences of their disorder and diagnosis of trichotillomania is made at a later stage. Also these patients may not be identified by clinicians as having a psychiatric problem, they may simply be lost in psychiatric referral after surgical recovery owing to stigma or ignorance.

Previous studies distinguish between voluntary pica (eating in response to what is available) and involuntary pica (eating in response to a compulsion or addiction). The likelihood that some cases of pica may respond to selective serotonin reuptake inhibitors (SSRIs) has led some to view it as a form of compulsion and that pica belongs on the same compulsivity-impulsivity spectrum that includes trichotillomania, trichophagia, and OCD .[5]

Method

Literature Search

The Cochrane Central Register for Controlled Trials and databases listed in EBSCOhost were searched. Google scholar and the PsychFileDrawer.org archive was searched, a website that specialises in studies which fail to replicate and studies that report null results. Electronic search strategies were based on Cochrane recommendations, the "Cochrane Highly Sensitive Search Strategy" for identifying randomised trials in MEDLINE (Higgins & Green, 2011).

Abstracts were screened and the inclusion criteria applied. All records were saved and duplicates removed. The references of included studies were searched. Authors were contacted by email (see Appendix S1 for list of authors contacted). Reasons for exclusion were recorded.Full-text articles were quality assessed with a standardised tool (See Appendix S2 for the detailed search strategy).

Quality Assessment

The suitability of studies was assessed with the Quality Assessment Tool for Quantitative Studies (Higgins & Green, 2011). Two researchers assessed full-text studies independently. Overall scores and acceptance/rejection decisions were compared. Any substantial discrepancies were resolved by a third author (RM).

Acceptance Criteria

The acceptance criteria were discussed between study authors and finalised before commencement of the systematic search. Only studies which achieved an overall score higher than an average moderate rating (6/12) were included. Randomised controlled clinical trials including quasi-randomised controlled clinical trials and randomised cross-over trials were included. Studies were not excluded based on lack of useable data (Higgins& Green, 2011).

Participants

The target population was adults who have been diagnosed with trichotillomania, with or without comorbidity.

Interventions

Psychological treatments needed to include the use of BT techniques or BT-based treatments (e.g., movement decoupling). Any psychological intervention that is administered in conjunction with BT (e.g., cognitive therapy, ACT) could be included. Psychological interventions included any intensity, length, and type of therapy delivery (e.g., group, individual, tele-interventions, self- help). Pharmacological interventions included any dosage, delivery mode, frequency, and duration. Com- parison groups could be active or passive control groups. A passive control group is a group of participants on a wait list or with minimal attention; an active control group is a group of participants who were given a placebo pill, or supportive, not symptom-specific intervention (e.g., progressive muscle relaxation).

Outcomes

The primary outcome was a reduction in trichotillomania symptoms as measured by the Massachusetts General Hospital—Hairpulling Scale (MGH-HPS); a self-report measure of symptom severity (see Keuthen et al., 1995). Reduction of symptoms could be measured with a variety of measurement types, such as: self-report questionnaires, number of hairs pulled, number of episodes ofhair pulling, and clinician ratings.

Results

Included Studies

The flow of studies through the screening process is presented in the Preferred Reporting Items for Systematic Reviews and Meta-analysis flow chart (see Fig. 1). Four hundred and thirty-four search records were found through database searches, no studies were identified by contacting authors, and 23 publications were identified by reference searches. Twelve studies, with a combined total of 299 participants, were included in this systematic review.

	Age	Clinical Presentation	First Clinical Visit	Provisional Diagnosis	Site	Clinical Intervention	Psychiatric intervention
Iqbal MM, et al[4]	26 year old male	Forcible hair pulling from scalp and moustache. Progressive loss of interest and motivation	Primary care physician	Trichotillomania	Scalp, Moustache	6 weeks' trial of sertraline, risperidone and clonazepam By Primary Care Physician	Trifluoperazine 1mg (thrice daily), Trihexyphenidyl 2mg (half three times daily), and Alprazolam 0.5mg (once daily). Furthermore, he was also advised for regular sessions of behavioral therapy
MichaelKD [5]	21-year-old Woman	chronic and significant hair pulling, associated hair loss, concealment of damage secondary to hair pulling, body image disturbance, depression, anxiety, and low self-esteem.	Psychiatrist	Trichotillomania	Scalp		HRT, progressive muscle relaxation, Gestalt awareness training, positive practice of rubbing a string of beads between her fingers (most often used during hair-pulling events), and thought stopping
Görgülü SA et al[6]	43-year-old married woman	Complaints of hair pulling.	Psychiatrist	Trichotillomania and Post-Traumatic Stress Disorder (Delayed Expression)	Scalp		Sertraline 100 mg, Aripiprazole 5 mg

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Kumar PNS et al[7]	58-year-old, married Hindu,	pulling hair and eating the hair root for the last 5 years.	Psychiatrist	Schizophrenia and trichorhizophagia.	Scalp		Olanzapine, escitalopram 10 mg
Patkar P et al[8] Case Series	14-year-old girl	plucking her own hair	Psychiatrist	TM	Scalp		fluoxetine 80 mg and buspirone 20 mg. Jacobson's Progressive Muscular Relaxation (modified for children), deep breathing exercises, distraction techniques, response prevention, thought-stopping, and diary maintenance was
	10-year-old male illiterate child	patchy hair loss due to pulling out of hair by the child. severe level of retardation in intellectual functioning	Psychiatrist	Trichotillomania	Scalp		fluoxetine 10 mg and risperidone 1 mg
	6-year-old Hindu girl child	bald patch on the vertex of her head due to her pulling out her own hair.	Psychiatrist	autism spectrum disorder with moderate severity of symptoms	Scalp		syrup fluoxetine, and referred for occupational and sensory stimulation therapies
	4-year-old girl child, Hindu by religion	pulling out her hair from the head, especially by her left hand. Most of the time, she threw away the hair immediately after plucking it; however, sometimes, she was seen smelling it before throwing.	Psychiatrist	Children's Apperception Test (CAT) test to look for any depressive or anxiety features	Scalp		syrup fluoxetine 10 mg

Modi N et al[9] Case Series	30-year-old female patient	hearing of voices, persecutory ideas and fearfulness, suspiciousness, and pulling out her hair for the last 12 years. pulled hair from the scalp and developed patches in these areas	Psychiatrists	Schizophrenia with Trichotillomania	Scalp		6 mg of risperidone, 20 mg of fluoxetine
	18-year-old female patient	anger outbursts and self-harm behavior; pulled her scalp hair and hid this hair in her books and cupboards.	Psychiatrist	borderline personality disorder with depression and Trichotillomania	Scalp		300 mg of oxcarbazepine, 20 mg of fluoxetine, Schema-focused therapy
	40-year-old female patient	depressed mood without diurnal variation, feelings of worthlessness and hopelessness, sleep disturbance for 2 months along with scalp hair pulling behavior.	Psychiatrist	Bipolar affective disorder with Trichotillomania	Scalp		1000 mg of sodium valproate.

Behaviour Therapy versus Passive Control

Four studies were included in the analysis which addressed the question "how effective is BT when com- pared to a passive control condition?" Included studies were: Van Minnen, Hoogduin, Keijsers, Hellenbrand, and Hendriks (2003), who investigated BT verses wait listcontrol (effect size (ES) = 1.08); Woods, Wetterneck, et al. (2006), who investigated ACT combined with HRT versus wait list (ES = 1.71); Ninan et al. (2000), who investigatedCBT versus Medication placebo (ES = 5.09); and Keuthen et al. (2012), who investigated DBT enhanced CBT versus minimal attention control (ES = 1.78). The results indicated that BT was more effective than a passive control condition (ES = 1.85, 95% confidence interval (CI) = 0.97, 2.74). While significant differences betweenstudy effect sizes were found (I^2 = 71%), further analyses investigating this difference were not conducted due to thesmall sample size.

Discussion

Overview of Findings

Based on the available RCTs, the findings indicate that BT has the most evidence as an effective intervention for TTM. This is when BT is conducted by highly trained therapists in research conditions and is compared to passive control groups. However, a more complex picture emerges when BT is compared to an active control, with the analysis revealing that when BT is delivered in groupformat or via the Internet, it was no more effective than the active control conditions. Fluoxetine was found to have minimal to no efficacy for the treatment of TTM. Clomipramine, olanzapine, and N-acetyl cysteine showed potential efficacy for TTM treatment.

Pharmacological Interventions

When compared to a placebo, N-acetyl cysteine, olanzapine, and clomipramine all show potential for the treatment of TTM symptoms. This is in agreement with both Bloch et al. (2007) and Rothbart et al. (2013).

With respect to psychological therapies, there is some preliminary evidence for the potential efficacy of integrating cognitive and affective strategies in the treatmentof TTM, as well as the contribution of social support, self-help activities, and generic therapy factors in the treatment of TTM. The social support component of HRTis not currently the most common component used in therapy for TTM (Flessner et al., 2010). However, the potential efficacy of supportive group therapy found herein suggests that social support may be important in reducing underlying negative emotions and beliefs. When comparing the social support discussed in previous literature and the social support explored in the current review, previous literature suggested that social support consists of a loved one reminding the individual to use a competing response and offering praise when the competing response is used correctly. The current review indicates that non-technique orientated social support of others who have experience with TTM may be particularly important in the treatment of TTM, consistent with the operation of this mechanism more generally in group therapy (Bieling, McCabe, & Antony, 2013). Finally, it is also of note that the only RCT to compare single versus dual modality treatment provides preliminary support for the efficacy of combined BT and medication. Future research would need to further examine the elements of the specific intervention that may have contributed tothe change.

Conclusion

In summary, this review found qualified support for some of the pharmacological and psychological treatments, although the small number of studies is a notable caveat to the drawing of firm conclusions. Nonetheless, N-acetyl cysteine, clomipramine, and olanzapine show some efficacy for the treatment of TTM. However, evidence is lacking for the usefulness of fluoxetine. BT shows the greatest efficacy for the treatment of TTM when com- pared to a passive control, supporting previous research. Of significance, this review found preliminary evidence that when BT delivered in group format or via the Inter- net is compared to an active control group, BT, progressive muscle relaxation, and supportive therapy show similar efficacy. Future research to enable more firmcomparisons is needed, particularly given the poor treatment outcome in this area in routine practice, combined with the schism between research evidence and typical treatment practices.

Informed Consent: Written informed consent was taken from patients.

Ethical Approval: Ethical committee approval was taken from the Institutional Committee Of Ethics, GMCB (GMCB/2022/11-99) .

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