Pathologic Hair-Pulling: A Review of the Literature and Case Reports

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Pathologic hair-pulling (trichotillomania) has been described in the dermatologic and psychiatric literature for the last century, but has become the focus of increased attention in the last few years. Once thought to be either a wholly benign condition or a symptom of obsessive-compulsive disorder, more recently hair-pulling has been noted in the context of numerous types of psychopathology, and has been reported to respond to several different types of intervention, both psychological and somatic. Estimates of its prevalence have varied widely. Because of the disparate conclusions of the literature on this condition, a more careful assessment of diagnostic validity is recommended.

PATHOLOGIC HAIR-PULLING was first described by Hallopeau[1] in 1889. He coined the term trichotillomania, which he defined as a self-inflicted alopecia resulting from avulsion of hair. Since that time, this behavior has been reported and discussed in both the psychiatric and dermatologic literature. Since the 1940s, when only five references to trichotillomania were made in the literature,[2] there has been a gradual increase in interest in the condition. Recently, attention to this condition has become more marked, as witnessed by the publication of four reports on hair-pulling in English-language psychiatric journals in the first 4 months of 1991 alone.[3-6]

Today, trichotillomania is classified as an impulse disorder—not elsewhere classified in the DSM-111-R.[7] Popkin,[8] reviewing this group of diagnoses, notes that "... individually they have been the subject of much theorizing and speculation, but distressingly little systematic study."

Obviously, this situation makes decisions about patient evaluation, treatment and management problematic. The purpose of this report is to review the literature on pathologic hair-pulling with respect to demographics, diagnostic assessment and validity, natural history, and treatment. The cases presented illustrate some of the variety of presentations and responses to treatment of this condition. It is our contention that neither the evidence of the literature nor our own case reports support the diagnostic validity of trichotillomania as currently defined. Finally, we discuss our own proposals for future clinical investigation.

DEMOGRAPHICS

There are no epidemiologic studies of trichotillomania in the world literature. The incidence of the syndrome has been variously estimated: Mannino and Delgado[2] reported that in a 10-year period, only seven of 1,368 children seen at the Mental Health Study Center presented with hair-plucking as a chief complaint. Their review of the literature showed a similarly low incidence of reported cases. Krishnan et al.[9] also found a low incidence, from three patients in 500 to five in 10,000 children with psychiatric disorders. However, Greenberg and Sarner,[10] Swedo et al,[11] and Christenson et al[3] voice the suspicion that this symptom is more common than would be expected from such studies. Azrin et al[12] have estimated that as many as 8 million Americans may have this condition. The dermatologic literature offers some evidence for a high incidence of trichotillomania: Mehregan,[13] through a questionnaire sent to 100 dermatologists, found that the average clinician could expect to see two to three cases of trichotillomania per year. Mueller[14] found that, between 1968 and 1977, 145 patients with this symptom presented to the dermatology service at the Mayo Clinic, and thereafter an average of 15 to 20 cases per year were seen.

The incidence of trichotillomania by sex among reported contacts overwhelmingly indicates a predominance of female over male patients. In the several descriptive studies of small patient samples published since the mid-1960s, only Mueller and Winkelmann[15] found

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more male (n = 15) than female (n = 9) patients. In his later study, Mueller[14] found a female to male ratio of greater than 2:1. Christenson et al[3] suggest males may have more ability to conceal their hair-plucking by masking it as male pattern baldness and by shaving mustache and beard sites.

The incidence by age is more problematic. Most investigators, especially in the dermatologic literature, observed that the symptom occurs more commonly among preadolescent children than among adolescents or adults: Mehregan[13] estimates the ratio of children to older patients to be 7:1. On the other hand, Greenberg and Sarner[10] found retrospectively that 13 of their 19 contacts had onset of hair-plucking between 11 and 16 years of age, while only three had onset at an earlier age. Similarly, most case reports in the psychiatric and psychologic literatures involve patients in their late teens to mid-30s.[16-19] These differences may reflect the belief often stated by both dermatologic[20] and psychiatric[21] investigators that the degree of psychopathology among children with trichotillomania is generally much less than in older populations. Consistent with this is the recent report of Christenson et al[3] in the psychiatric literature of 60 hair-pullers with mean age of onset being 13 years and mean duration 21 years.

SYMPTOMATOLOGY AND PHYSICAL FINDINGS

Patients are reported to complain at times of pruritis in the area of alopecia, but rarely complain of other types of irritation or pain as a justification for their hair-plucking. In fact, a significant portion of patients observed denied the self-induced nature of the symptom.[14] In another, largely self-referred, sample,[3] many found the act itself painful.

In all the studies under review, alopecia was found predominately on the scalp, but was also noted in some cases on the eyebrows, eyelids, pubes, face, trunk, and extremities. In cases involving the scalp, peripheral portions of the scalp are typically spared, often resulting in a tonsured appearance. The pattern of alopecia is generally ill-defined and irregular, with an admixture of both short and long hairs. In Mehregan's study,[13] the distinguishing feature of this type of alopecia was the incomplete nature of the hair loss and the absence of inflammation, atrophy, and scarring.

Histopathologic studies have shown as typical of trichotillomania follicular plugging and the presence of catagen hairs. The prognosis for hair regrowth was "less favorable" than previously suspected in one study.[15] Other investigators do not address this problem, the assumption being that in the absence of other kinds of scalp pathology the hair will grow back without difficulty.

The most medically serious consequence of trichotillomania is the development of trichobezoars when patients eat the avulsed hair. Bezoars can result in intestinal obstruction, gastrointestinal bleeding, bowel perforation, acute pancreatitis, and obstructive jaundice. Although only a few cases of this complication have been reported, suggesting that it is rare, thorough evaluation should include questioning the patient about trichophagy and obtaining a medical workup as needed.[15]

The primary dermatologic diagnosis to consider in cases of alopecia is alopecia areata, with tinea capitis and other less common fungal infections also in the differential. In two studies[10,14] it was found that at least 50% of patients had received the diagnosis of alopecia areata before the self-induced nature of the alopecia was discovered.

DIAGNOSIS AND NATURAL HISTORY

The diagnostic and prognostic significance of trichotillomania has been discussed by several investigators. Phillipopoulos,[22] writing in 1961, claimed that most psychiatrists recognized pathologic hair-plucking as a symptom of obsessive-compulsive neurosis.

Greenberg and Sarner[10] found that the psychiatric diagnoses in their sample were more varied, including "schizophrenic reaction," "borderline state," "psychoneurosis, mixed type," as well as "psychoneurosis, obsessive-compulsive." Since that time, several different views on the psychopathologic significance of trichotillomania have been put forward. Galski,[23] reviewing the psychoanalytic literature, emphasizes the role of hair as a symbol of sexual conflict and as a transitional object through which the patient works out feelings of abandonment. Sticher et

al.[24] similarly argue that "the cause of hair plucking is purely psychological," again stressing the symbolic importance of hair.

Azrin et al[25] have described this symptom as a habit, similar to nail-biting or nose-picking. Wolf[26] notes that a behavioral analysis of such habits will reveal their importance as a means of reducing stress. Case reports in the psychiatric literature, concerned more with the treatment of individual patients and less with theoretical explanations of the symptom, have emphasized obsessive-compulsive disorder,[17] major depression,[16] and psychosis[27] as likely diagnoses in people with pathologic hair-plucking.

A recently published descriptive study of 60 subjects[3] addresses the question of diagnosis. The authors found that, while all subjects experienced either increasing anxiety before pulling out hair or gratification afterward, 28% of the subjects did not experience both and so did not meet DSM-III-R criteria for trichotillomania, suggesting that the criteria are "overly restrictive." This study also found a high rate of comorbid psychiatric diagnoses either concurrently with the trichotillomania or by history. In particular, 65% of the subjects received a diagnosis of mood disorder. Similarly, 57% were diagnosed with an anxiety disorder. Surprisingly, only 15% of the study population met criteria for obsessive-compulsive disorder. Butterworth[28] reports on the high frequency with which the symptom is found among an institutionalized population of the mentally retarded.

As previously mentioned, trichotillomania in preadolescent children is often thought to be much less indicative of severe psychopathology than in adolescents and adults. Both dermatologic and psychiatric literatures describe the occurrence of hair-plucking in childhood as "a form of frustration."[21] Investigators emphasizing family dynamics as the major contributing cause of trichotillomania in children note that the symptom resolves quickly with improvement in parent-child relations.[21,29] Mehregan[13] found that most dermatologists felt confident that the symptom could be relieved simply by discussing it with the child and his parents. However, Mueller and Winkelmann[15] observed in their case sample that the longer the symptom had persisted, regardless of age of onset, the more likely psychiatric consultation would be required.

TREATMENT

Psychoanalytic case reports, the earliest case studies of treatment for trichotillomania in the literature, have reported moderate success in the treatment of the symptom, but these studies have not included follow-up or objective measures of improvement.[22,30]

The first report of hair-plucking treated with behavioral methods involved the successful use of thought-stopping to prevent eyebrow-plucking in a 40-year-old woman with a 30-year history of the symptom.[31] In the 1970s and 1980s, several individual case reports documented success in the use of various behavioral techniques for this problem.[18,33-34] The only controlled study of behavioral therapy for hair-pulling[25] compared the utility of habit reversal, a technique of behavioral therapy developed by Azrin and Nunn,[35] with negative practice training. The two procedures were taught to similar populations of chronic hair-pullers in one 2-hour session. Subjects were followed by telephone. Those who received negative practice training had a 52% to 68% reduction in hair-pulling over a 3-month follow-up period, while for the subjects who were taught habit reversal, the reduction of symptoms was 91% at 4 months and 87% at 22 months. Greenberg and Marks,[36] in an uncontrolled study of seven patients, expressed the opinion that habit reversal will be the treatment of choice for the behavior in the future. Other studies report successful treatment using hypnosis.[37]

Except for the study of Swedo et al[11] using clomipramine, the pharmacotherapy literature on trichotillomania has been limited to case reports. Childers[27] reported success using chlorpromazine in two middle-aged women with long histories of unremitting psychosis who had been in an institutional setting for several years before treatment. Snyder[17] used amitriptyline to successfully treat a 26-year-old women with obsessive-compulsive disorder and trichotillomania, but without evidence of major depression. Krishnan et al[16] used isocarboxizide in the treatment of a 32-year-old man with trichotillomania of several years' duration and major depression of 2 months' duration. Both the

depression and the hair-plucking resolved with this therapy. A recently reported chart review[4] suggests that lithium carbonate may be helpful in controlling chronic hair-pulling. Winchel (personal communication) reported success in the amelioration of compulsive hair-pulling during a 16-week open trial of fluoxetine.

Swedo et al[11] compared clomipramine and desipramine in a population of hair-pluckers using a double-blind, cross-over method of control. They found that clomipramine was significantly more efficacious than desipramine in this population, and that changing from the more effective medication to the comparison drug resulted in an exacerbation of the symptom. This study is noteworthy for having excluded patients with diagnoses other than trichotillomania that might explain the symptom, including schizophrenia, major depression, and obsessive-compulsive disorder. However, their method of selecting patients for participation in the study may have introduced a bias toward the inclusion of obsessional patients: nine of the 20 patients originally included had referred themselves to the investigators after watching a television documentary on obsessive-compulsive disorder; also, the inclusion criteria for the study required a description of the hair-plucking very similar in some particulars to the DSM-III-R criteria for obsessive-compulsive disorder, ie, "repetitive, compulsive pulling out of the hair that the patient considered unreasonable and undesirable. . . . The symptom had to be . . . sufficiently severe to interfere with the patient's daily life because of the time lost to hair-pulling episodes." While this does not negate the significance of the finding, it does draw into question its generalizability to all patients with the symptom.

CASE REPORTS

These three cases illustrate some of the clinical situations in which pathologic hair-pulling may occur. Each case has some feature that is contrary to received ideas about pathologic hair-pulling. In the first case, the patient has spontaneous remission of hair-pulling in childhood, but has recurrence of the behavior in conjunction with obsessive-compulsive disorder in late adolescence, thus bringing into question the reported dichotomy between hair-pulling in childhood and in adolescence.

Both the first and second cases show a problematic response to behavior therapy, with abrupt initial improvement, but subsequent relapse of hair-pulling. In the second case, a primary diagnosis of obsessive-compulsive disorder could not be made until late in the course of his clinical follow-up due to his reluctance to discuss his other compulsive behaviors. In the third case, hair-pulling is seen in association with substance abuse and impulsive behaviors, without any evidence of obsessive or compulsive pathology.

In all these cases, hair-pulling is found in conjunction with other psychiatric diagnoses. The response to therapy of each patient suggests that hair-pulling is best treated as part of the primary diagnosis, rather than as a separate and isolated diagnosis.

Case 1

Ms A is a 42-year-old married white woman who presented with a chief complaint of being unable to control her hair-plucking. She reported that she had started pulling her hair out at 4 years of age, but had stopped spontaneously within 1 year. When she was 16 years old, she heard her older sister complain of a college roommate who had lice. Her sister told her that lice accumulate on "bumpy hairs." In an effort to assure herself that she did not have lice, the patient began pulling individual strands of hair looking for "bumpy" ones. She recalls that she experienced mounting anxiety as she sought these hairs: if a hair was straight, she threw it away, but if it was "bumpy" she felt an immediate sense of relief. Though she eventually stopped looking for lice, Ms A continued to follow this same pattern of looking for "bumpy" hairs whenever she pulled her hair. Over the years she developed a more elaborate ritual involving hair-plucking, biting her nails, and pinching pimples and other "imperfections" in her skin. The amount of time spent on this ritual varied with the degree of stress the patient was experiencing at a given time and with the amount of time available to her to perform the ritual in private. With her marriage at 19 years of age, she increased her hair-pulling to an average of at least 1 hour per day, and within a few years her hair-pulling had progressed across the calvarium, making it necessary for her to wear a wig to hide her baldness. When initially seen, Ms A had near-total baldness of the left side of her head except for a fringe of hair suggesting a tonsure, and a less complete but clearly evident lack of hair over the right side of her head. The patient was emphatic in stating that her compulsion to pull her hair was undesirable and uncontrollable. Her mental status was most remarkable for a tendency to ruminate about topics only tangentially related to the subject at hand and for her overinclusive narration of events.

The patient denied episodes of major depression, although she admitted to feeling chronically tired and sleeping greater than 8 hours per day. She initially denied other compulsive behaviors, but noted that hair-plucking was often linked to an inability to clean her house adequately. Ms A's description of household tasks revealed her need to spend several hours cleaning the bathtub every time it was used. In completing the Yale-Brown Obsessive Compulsive Scale (Y-BOCS) symptom checklist,[38,39] Ms A also noted a compulsion to explain things and an undesirable tendency to spend hours daily ruminating about numerous topics ("I have all this garbage running through my head all the time").

The patient's initial workup included a Y-BOCS (score, 13 on obsessions, 13 on compulsions) and a behavioral analysis. In this way, it was found that hair-plucking relieved the anxiety associated with all Ms A's obsessions and cleaning compulsions. Based on these findings, Ms A was started on low-dose clomipramine, which was increased over several months to 225 mg/d. At the same time, she was given progressive relaxation training and then taught exposure and response prevention techniques. She was instructed to keep a record of her compulsive behaviors and soon reported an almost complete cessation of hair-plucking lasting several weeks. However, she also noticed a concurrent increase in other compulsive behaviors, including washing her hands for an hour or more per day and taking very long showers. Exposure therapy proved difficult because the patient was able to avoid the anxiety induced by the exposures by performing them only when she was extremely tired or when she was too busy to perform them adequately. Over the first several weeks, as these problems were addressed, the hair-plucking returned to baseline levels, but the patient was able to minimize her compulsive behaviors and her avoidance of the exposures. After 9 months of therapy, including 3 months of clomipramine 225 mg/d and more adequate performance of behavioral therapy, the patient was pulling no more than 10 hairs per week. She had also stopped biting her nails and checking her skin, and reported decreased ruminative thinking. She was observably less ruminative, overinclusive, and anxious on examination. Her other cleaning compulsions had also diminished considerably.

Case 2

Mr B is a 25-year-old single man who presented to the psychiatric clinic with the chief complaint of pulling hair for 18 years. He started pulling hair from his eyelashes, eyebrows, and shoulders at age 7, and by age 12 he was also pulling hairs from his upper extremities and pubic area. He would pull his hair in private while ruminating about events in his life and how he could have better handled them. He describes pulling his hair in a ritualistic fashion that involves bringing his hand up to his mouth, taking a single hair between his teeth and biting it out. He would play with the piece of hair in his mouth, eventually spitting it out. He never swallowed or ate his hair. He performed this ritual one to two times per day for 30 minutes to 2 hours. Often he was not aware of this activity and was surprised when he looked down at the dashboard of his car while driving and saw large amounts of hair. Although this ritual seemed senseless and time-consuming, he felt compelled to continue until it was completed.

After several therapy sessions, Mr B admitted to various other compulsive behaviors. As a child he would spend hours organizing rock collections and placing *National Geographics* in chronological order, and copying and alphabetizing items. As an adolescent he would mow the family's lawn in a manner that entailed dividing the yard into nine sections and tending to each section, including mowing the grass, trimming the bushes, and organizing the rocks, before he could go on to the next section. Because of this elaborate ritual it would take him all week to mow the lawn once. Despite these behaviors, he successfully completed high school and college.

However, since graduating he has done little to pursue his career as a musician. He spends much of his day attending various support groups and diligently follows their programs in an attempt to "rebuild" his life. He prepares exhaustive schedules of daily activities and ways to improve himself. Although he constantly writes his thoughts down in a notebook that he carries with him, he continues to ruminate a great deal about his past. He is preoccupied with self-doubt and achieving perfection. He describes having frequent thoughts about sexual violence, but is reluctant to give details about these. However, he admits that they are senseless and upsetting.

Mr B was treated while in college with psychotherapy, but terminated it after 1 week. He has had several short periods of abstinence from hair-pulling since childhood. He reports good health and is not on any chronic medications. His family history is significant for his mother pulling her eyebrows out when she was younger, but not now. His sister pulled her hair out with her teeth in a manner similar to the patient, and also pulled hair from her head. She has been diagnosed with bipolar affective disorder and prescribed lithium. Many members of his father's family, including his father, abuse alcohol.

On exam, Mr B initially exhibited no evidence of affective disorder, psychosis, or motor abnormalities. His appearance was notable for a lack of hair on his arms and hands, which he did not attempt to hide. He was mildly anxious in early sessions, but this lessened with time. At the time of his initial evaluation, he had read some literature about habit reversal therapy and with further education and support he showed a marked improvement after 2 weeks of treatment. However, within 1 month, his hair-pulling had returned to baseline. He was reluctant to try antiobsessional medication because of his fear of side effects. He discontinued therapy after 1 month, stating he felt he could pursue the techniques on his own, though he admitted to the lack of improvement in his condition.

Case 3

Ms C was a 22-year-old, single, white woman employed as a bartender at the time she presented for admission via a state hospital emergency room. She had called a local suicide prevention hotline and reported that she was having thoughts of shooting herself. Shortly before this, she had had at least six drinks and an argument about her recent DWI with her mother. Police were notified of her threats and Ms C was brought to the emergency room. Her chief complaint at presentation was: "I shouldn't have called that Suicide Hotline."

Ms C reportedly began to use drugs at the age of 10. She used marijuana, cocaine, phenylpropanolamine, and psilocybin between the ages of 10 and 14. She quit using the latter three drugs at age 14 and would only use marijuana occasionally. She did continue to have difficulties with alcohol abuse. She had been fired from a job for arriving at work intoxicated. She had arrests for driving while intoxicated and purchasing alcohol for minors. She would get tremulous at times with abstinence and has had blackouts with heavy use.

At the time of initial presentation, Ms C had a 4 cm × 4 cm bald spot on the crown of her head, which she reported as very embarassing to her. She later said that she had begun to pluck her eyebrows and eyelashes at 4 years of age and that this had continued through the present. Her mother initially took her to a pediatrician, who told them that it was "just a phase" and that she would grow out of it. She began to pull hair from the top of her head at the age of 14 years. She would typically spend 30 minutes per day pulling out her hair. She would only do this while alone. She could occasionally resist the urge to pull out her hair.

Ms C denied any obsessions or compulsions. She denied psychotic symptoms. She reported that she has always "been depressed," but lacked the symptoms required to meet criteria for an affective disorder. She was first seen by a psychiatrist at the age of 15 and diagnosed with borderline personality disorder. She had her first psychiatric hospitalization at age 16 when she cut her wrists in a suicide attempt. She was not treated with medication at that time.

Ms C has reportedly always had difficulties with control of her behavior with frequent temper tantrums as a child. She initially did fairly well in school, but performance began to decline in junior high school. She had truancy, school suspensions, and fighting. Her only arrests were the alcohol-related episodes noted above.

During her hospitalization, she gave no evidence of affective disorder, psychosis, or obsessive-compulsive disorder and refused fluoxetine for her hair-pulling. She was discharged to an alcohol treatment program, which she completed. She eventually sought psychiatric care as an outpatient and was treated with fluoxetine 20 mg/d. She initially reported that she had increased ability to resist the urge to pull out her hair. However, she decided within a few weeks that medication was not helpful. During that period she continued to abuse alcohol episodically. After 6 weeks, she discontinued fluoxetine and was lost to follow-up.

DISCUSSION

Our review and case presentations have shown that the validity of trichotillomania as a unique diagnosis is questionable. Consequently, generalizations about demographics, natural history, and treatment cannot be reliably made. Reports of the prevalence of the behavior range from extremely rare[2] to fairly common,[12] and are largely guesswork. However, given the deficiencies in our understanding of pathologic hair-pulling, epidemiologic study to better ascertain the demographics of this condition would be premature.

Similarly, the natural history of pathologic hair-pulling requires further exploration in the context of diagnostic assessment. Although Christenson et al[3] devote a large part of their report to the behavioral characteristics of their study subjects, they do not analyze these characteristics with respect to the high frequency of comorbid diagnoses they found in this population. Such an analysis could have been helpful in explaining the relation between hair-pulling and other, seemingly discrete, diagnoses. It is also interesting to note that the rationale for treating chronic hair-pullers with clomipramine has been that this is a compulsive behavior,[11] while the rationale for using lithium carbonate has been that the behavior is impulsive in nature.[4] This discrepancy and the fact that both therapies are reported to be successful suggests how far we are from understanding the nature of this behavior.

The treatment modalites reported to be successful in ameliorating pathologic hair-pulling include a large part of the armamentarium of psychiatry and clinical psychology. Again, diagnostic issues would seem to dictate the type of therapy used. While this seems obvious in some cases (eg, the use of antipsychotics in institutionalized schizophrenics), it is not always so. A possible selection bias in the study of Swedo et al on drug treatment has already been pointed out: if obsessive-compulsive disorder rather than hair-pulling per se was the reason for the success of clomipramine in this population, then the utility of clomipramine as a treatment of choice for trichotillomania would be arguable. Similarly, our experience with patients A and B showed a problematic response to behavioral therapy, while Azrin et al[25] had phenomenal success using habit reversal in only one 2-hour session. A thorough diagnostic assessment would be helpful in distinguishing those patients who might benefit from behavioral therapy alone.

Obviously, pathologic hair-pulling is not the first behavioral disorder in which diagnosis, natural history, treatment, and prognosis have been ambiguous. In an attempt to address similar problems in the diagnosis of schizophrenia, Robins and Guze[40] recommend five phases of investigation for assessing diagnostic validity. These are (1) clinical description, including both a description of signs and symptoms as well as demographic data, precipitating factors, and other information that may "define the clinical picture more precisely"; (2) laboratory studies, including chemical, physiological, radiological, anatomical, and psychological—an area that the authors recognize has not been consistently reliable in the study of idiopathic psychiatric

syndromes; (3) delimitation from other disorders, i.e., the establishment of exclusion criteria to assure a homogeneous patient population in the study of a given disease; (4) follow-up studies in order to reassess earlier conclusions about diagnosis, and to learn the prognostic significance of a diagnosis; and (5) family studies, since many psychiatric diseases run in families, and the finding of a higher prevalence of similar syndromes in families would strongly support the validity of a diagnosis. Referring to these principles of investigation, our review has shown that the clinical description of pathological hair-pulling has been largely anecdotal with a wide range of conclusions drawn by various investigators. Pathological hair-pulling has been variously described as a unique syndrome and as coexistent with several other psychiatric disorders. Laboratory study, especially in the absence of a clear clinical description of pathological hair-pulling, seems particularly unpromising. There have been no follow-up studies of pathological hair-pulling in the literature. While there are anecdotes of this behavior occurring in families, there have been no family studies.

Given the growing interest in pathologic hair-pulling in the psychiatric literature, a systematic investigation such as that described by Robins and Guze will be necessary to improve evaluation and treatment of this behavior.

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