Body-focused repetitive behaviors in children and adolescents, clinical characteristics, and the effects of treatment choices on symptoms: a single-center retrospective cohort study

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Abstract

Introduction: This study determines clinical features of body-focused repetitive behaviors (BFRBs) among a sample of Turkish children and adolescents, evaluates the naturalistic treatments selected in this sample, and determines the effects of those treatments during a 3-month follow-up.

Methods: The study included a cohort of 67 patients 9 to 17 years old attending a tertiary center between March and June 2013 with complaints of nail biting, skin picking, and/or hair pulling. The patients completed psychometric scales and were evaluated for symptom severity, improvement, and adverse effects after an initial interview and at control visits during the 4th and 8th weeks.

Results: The most common BFRB was nail biting. In nail-biting behavior, subjective awareness was higher, and urges prior to the behavior and release after the behavior were found to be higher. Although there was no significant difference between the choice of treatment and the course of psychiatric measures, significant improvement was found in functionality after treatment.

Conclusions: Despite its limitations, the small number of studies on BFRBs increases the importance of studies in this area.

Keywords: body-focused repetitive behaviors, children, adolescents, trichotillomania

Introduction

Trichotillomania (also known as hair-pulling disorder), excoriation disorder (or skin-picking), and some severe forms of onychophagia (nail biting) are classified under obsessive-compulsive and related disorders in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) (1). The close relationships between trichotillomania, excoriation disorder, onychophagia, and lip chewing have led to their classification as body-focused repetitive behaviors (BFRBs) or pathological grooming disorders (2, 3). BFRBs are defined as repetitive, intentional acts of habitual grooming behaviors that result in physical harm and social avoidance due to inability to control the behaviors (4, 5). Most studies conducted on BFRBs involve preclinical studies, epidemiological surveys, and case reports or series. Information about evidence-based treatments is still accumulating (5, 6).

BFRBs have serious dermatological effects such as resistant paronychia, tissue damage, and alopecia (7, 8), and growing research suggests that BFRBs are relatively common. The prevalence of excoriation disorder has been estimated as 1.4 to 5.4% among adults (9–11) and 2 to 4% among children and adolescents (12, 13). Excoriation disorder may display a tri-modal distribution, with increased incidence in childhood, adolescence to young adulthood, and middle adulthood (9, 11, 14). It is common among females and is associated with elevated rates of comorbidity and distress (9, 11, 14). Available evidence suggests that the prevalence of trichotillomania may vary between 0.5 and 2.0% among adults (15), and for children and adolescents the prevalence may vary between 0.5 and 3.9% (12, 16). Onset is typically in late adolescence and may be earlier among females (15, 17). Onychophagia has received relatively little attention compared to other BFRBs, and it is grouped with lip-biting and cheek-chewing in the DSM-5 (1, 18).

Available research on this disorder, although dated, suggests that rates may be as high as 50% in childhood, with a slight female preponderance, and that rates may decrease with age to 4.5% in late adulthood (18).

The neurobiology of BFRBs may involve various neurotransmitter systems, with noradrenaline, serotonin, dopamine, opiates, and glutamate being implicated in various studies (19–21). However, a recent review of available psychopharmacological treatments reported that the treatment armamentarium for BFRBs is still disappointing and that the need for further studies as well as integration of pharmacological agents with psychotherapeutic interventions still has not been met (19). Moreover, in a recent review of psychosocial therapies for pediatric BFRBs, none of the therapeutic methods met the criteria for well-established status (8). Regardless of the limitations of available therapies, recent surveys emphasize the ubiquity and deleterious effects of those symptoms (22).

Therefore this study seeks to a) determine the rates of BFRBs among a sample of Turkish children and adolescents referred for treatment or evaluation at a tertiary center focusing on child and adolescent psychopathology during a 3-month period, b) determine the clinical features in this sample, c) evaluate the naturalistic treatments selected by clinicians in this sample, and d) determine the effects of those treatments during a 3-month follow-up.

Methods

This retrospective cohort study was conducted at the Abant İzzet Baysal University Medical Faculty between March and June 2013. Patients 9 to 17 years old presenting at the center during the study...
period (n = 1,800) were screened for complaints of nail biting (n = 250), skin picking (n = 70), hair pulling (n = 98), and obsessive-compulsive behavior (n = 120). Body dysmorphic disorder (n = 16), psychotic disorder (n = 7), and mental retardation or intellectual disability (n = 325) were criteria for exclusion. The complaints must not have started after receiving treatment (n = 195) or using a drug or other substance (n = 158). Intellectual disability was excluded based on the Wechsler Intelligence Scale for Children–Revised (WISC-R).

Institutional review board approval for the study was procured from the Ethics Committee of the Faculty Medicine at Gaziantep University (2020, no. 266).

The baseline interview for children and parents included current and lifetime nail biting, hair pulling, and skin picking; the duration and frequency of BFRBs; the most frequent occasions for BFRBs; whether children were aware of those behaviors; the presence of urges prior to the behavior as well as release after the behavior; subjective distress due to behavior; the impact of BFRBs on peer relations, academic achievement, and family relations; bodily changes and social avoidance due to BFRBs; previous attempts at quitting the behaviors; and methods used in previous such attempts. The duration of behaviors, degree of control, and resistance to behaviors were evaluated with items modified from the Children’s Yale–Brown Obsessive Compulsive Scale (CYBOCS) (23). Semi-structured clinical interviews (i.e., iddie Schedule for Affective Disorders and Schizophrenia for School-Age Children: Present and Lifetime Version; K-SADS-PL) were used to determine psychiatric comorbidities. The severity of symptoms after the initial interview was evaluated with the Clinical Global Impression–Severity scale (CGI-S). Current functioning, functioning in the past year, and functioning at the time of maximum severity of symptoms were evaluated with the Children’s Global Assessment Scale (CGAS). BFRBs leading to distress or impairment were determined at the baseline interview.

Children completed the Children’s Depression Inventory (CDI), Screen for Child Anxiety and Related Disorders (SCARED), and Maudsley Obsessional Compulsive Inventory (MOCI) after the initial interview. Clinicians were free to choose behavioral therapy, antidepressants, antipsychotics, or other agents based on their clinical judgement, and the selections of children and their families. Control visits were conducted at the 4th and 8th weeks of treatment. At each visit, the clinicians evaluated symptom severity, improvement, and adverse effects with CGI-S, CGI-Improvement (CGI-I), and CGI–Adverse Effects (CGI-AE), and the children completed the CDI, SCARED, and MOCI.

Seven measures were used in this study:

1) The Wechsler Intelligence Scale for Children–Revised (WISC-R) was initially developed to evaluate intellectual functioning of children 5 to 15 years old. This was revised in 1974, and the age range of the revised version was changed to 6 to 16 years. The revised version was translated, and its reliability and validity were established by Savasir and Sahin (24). The WISC-R was used to exclude intellectual disability in this study.

2) CGI was developed by Guy (25) to allow mental health clinicians to rate the severity of symptoms in their patients as well as their treatment responses and adverse effects. The symptoms are rated on a seven-point Likert-type scale from 1 (not ill) to 7 (among the most extremely ill patients). Total improvement of symptoms with treatment is rated from 1 (very much improved) to 7 (very much worse). Side effects are rated from 1 (none) to 7 (significantly interfere with functioning). The clinicians rated the severity of symptoms, improvement, and adverse effects of patients at baseline and at control visits in this study.

3) CGAS was developed by Schaffer et al. (26) to provide a global measure of functioning in children and adolescents. It provides a single, global rating between 0 and 100. The clinicians rated the children in this study with CGAS at baseline, within the past year, and in the past when the severity of symptoms was maximum.

4) SCARED was developed by Birmaher et al. (27) to evaluate anxiety disorder symptoms. There are parent and child forms. The Turkish reliability and validity were established previously (28). Children completed the SCARED at baseline and at control visits in this study.

5) CDI is a 27-item, self-report, Likert-type scale. It was developed to evaluate subjective symptoms of depression among children 6 to 17 years old (29). The Turkish version was previously found to be valid and reliable (30). Children completed the CDI at baseline and at control visits in this study.

6) MOCI is a 30-item, true-false type, self-report scale to assess the presence and severity of obsessive-compulsive symptoms (31). The original form has subscales for “control,” “cleaning,” “slowness,” and “doubt.” In the translation process, seven items from the Minnesota Multiphasic Personality Inventory (MMPI) were added to the original scale to bring the total to 37 items. The Turkish version was previously found to be reliable and valid (31). Factor analysis revealed a three-factor structure for the Turkish version (i.e., cleaning-punishingness, obsessive thoughts / rumination, and slowness/checking). No cutoff score was calculated for the Turkish version. In this study, the MOCI was completed by children at baseline and at control visits.

7) K-SADS-PL was developed by Kaufman et al. (33) to evaluate present and life-time psychopathology among children 6 to 18 years old according to a semi-structured interview conducted along DSM-IV-TR criteria. The Turkish version was previously found to be valid and reliable (34).

Statistical analysis

The data were analyzed with SPSS for Windows™, Version 17 (SPSS Inc., Chicago, IL, 2008). Nominal variables were summarized as counts and frequencies, and quantitative variables were summarized as either means and standard deviations or medians and inter-quartile ranges (IQR), depending on assumptions of normality and the presence of outliers. Multivariate analysis of variance controlling for covariates (MANCOVA) or repeated measures was used to compare changes in measures according to treatment choices. P was set at 0.05 (two-tailed).

Results

A total of 185 patients (10.3%) presented at the center with complaints of BFRBs during the study period. Of these, 91 (49.2%) were under 9 years old, 12 (6.5%) refused to participate in the study, eight (4.3%) did not complete the forms, and seven (3.8%) did not attend follow-up visits. Therefore, 67 patients (56.9% male) with a mean age of 12.5 years (SD = 2.3) were eligible for enrollment in the study. The rate of chronic (> 3 months) BFRBs leading to distress or impairment among children ≥ 9 years old was 3.8%. The rates of trichotillomania and excoriation disorder according to DSM-5 criteria were 0.6% and 1.2%, respectively. Approximately one-
third of the patients (30.9%) had at least one comorbid disorder. The most common psychiatric comorbidities were attention deficit hyperactivity disorder (n = 21, 30.9%), followed by disorders related to conduct (n = 1, 1.6%), obsessive-compulsive behavior (n = 1, 1.6%), and the autism spectrum (n = 1, 1.6%).

The mean duration of BFRBs in the study sample was 44.7 months (SD = 23.0). Subjective distress, impairment, social avoidance, and bodily changes observable by others were most frequently reported for onychophagia. Similarly, urges prior to the behavior and release after the behavior were most frequently reported for onychophagia, followed by skin picking and trichotillomania. Watching television and a feeling of emptiness were the most frequent aggravating situations (Table 1).

The patients attempted quitting BFRBs a mean of 1.4 (SD = 2.3) times. The modes of past attempts to quit were 1.0 (n = 6, 9.0%) and 3.0 (n = 6, 9.0%). Past attempts to quit involved the use of negative reinforcement for onychophagia (n = 25, 80.0%), whereas behavioral methods were used for hair pulling (n = 1, 1.6%). Past attempts to quit skin picking involved pharmacological treatments (n = 1, 1.6%) and behavioral interventions (n = 1, 1.6%) at equal rates. The evaluation of current BFRBs in terms of frequency, duration, control, and resistance is illustrated in Table 2.

At the study center and within the specified timeframe, the most common treatment choice was use of atypical antipsychotics (n = 34, 50.0%), followed by selective serotonin reuptake inhibitors (SSRIs; n = 22, 32.4%). In 10 of the patients, SSRIs and atypical antipsychotics were prescribed at the same time (14.7%). Only two of the patients (2.9%) received habit reversal training. Baseline CGAS scores of children with onychophagia (67.9 ± 3.9), hair pulling (67.3 ± 3.2), and skin picking (67.9 ± 4.0) did not differ significantly (Kruskal–Wallis test, p = 0.90). The groups also did not differ for CGAS scores within the past year (onychophagia, 77.4 ± 4.0; hair pulling, 77.5 ± 2.9; skin picking, 78.3 ± 3.8; p = 0.60) as well as for scores at the maximum severity of symptoms (onychophagia, 61.2 ± 4.4; hair pulling, 61.3 ± 4.8; skin picking, 62.0 ± 2.6; p = 1.00). The baseline and control evaluations of patients are presented in Table 3.

One-way analysis of variance (ANOVA) for repeated measures was used to evaluate the interaction of time and diagnostic groups. Box’s M for the CDI, SCARED, MOCI, and CGI-S indicated that the covariance matrices are equal (F[6, 1452.9] = 0.9, p = 0.48; F[6, 1452.9] = 0.9, p = 0.50; F[6, 1150.3] = 0.6, p = 0.74; and F[6, 1452.9] = 0.6, p = 0.77, respectively). Sphericity could not be assumed (p < 0.001), and the Greenhouse–Geisser correction was used for within-subject effects. Analysis revealed that, except for the CGI, no interaction was present between the diagnostic group and visit time in terms of psychometric measures. For the CGI both time (F = 136.0, p < 0.01, partial

Table 1 | Current and life-time rates and phenomenological features of body-focused repetitive behaviors (BFRBs) reported among Turkish children 9 to 17 years old.

<table>
<thead>
<tr>
<th>Behavior, n (%)</th>
<th>Onychophagia</th>
<th>Trichotillomania</th>
<th>Skin picking</th>
</tr>
</thead>
<tbody>
<tr>
<td>Current</td>
<td>63 (92.6)</td>
<td>4 (5.9)</td>
<td>10 (14.7)</td>
</tr>
<tr>
<td>Past</td>
<td>45 (66.2)</td>
<td>1 (1.6)</td>
<td>2 (2.9)</td>
</tr>
<tr>
<td>Subjective awareness</td>
<td>50 (76.9)</td>
<td>4 (5.9)</td>
<td>9 (13.2)</td>
</tr>
<tr>
<td>Urges prior to behavior</td>
<td>36 (55.4)</td>
<td>3 (4.4)</td>
<td>7 (10.3)</td>
</tr>
<tr>
<td>Release after behavior</td>
<td>28 (43.1)</td>
<td>3 (4.4)</td>
<td>10 (14.7)</td>
</tr>
<tr>
<td>Subjective distress</td>
<td>34 (52.3)</td>
<td>2 (2.9)</td>
<td>4 (5.9)</td>
</tr>
<tr>
<td>Impairment</td>
<td>11 (16.9)</td>
<td>1 (1.6)</td>
<td>2 (2.9)</td>
</tr>
<tr>
<td>Past attempts at quitting</td>
<td>31 (47.7)</td>
<td>1 (1.6)</td>
<td>5 (7.4)</td>
</tr>
</tbody>
</table>

Table 2 | Frequency, duration, control, and resistance features of body-focused repetitive behaviors (BFRBs) reported among Turkish children 9 to 17 years old.

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Onychophagia (n = 63)</th>
<th>Trichotillomania (n = 4)</th>
<th>Skin picking (n = 10)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frequency</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Daily, most of the time</td>
<td>18 (27.7)</td>
<td>0 (0.0)</td>
<td>2 (2.9)</td>
</tr>
<tr>
<td>Daily, a few times</td>
<td>37 (56.9)</td>
<td>1 (1.6)</td>
<td>3 (4.5)</td>
</tr>
<tr>
<td>Weekly, a few times</td>
<td>10 (15.4)</td>
<td>4 (5.8)</td>
<td>6 (8.8)</td>
</tr>
<tr>
<td>Duration (per day)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 1 hr</td>
<td>52 (80.0)</td>
<td>5 (7.4)</td>
<td>9 (13.2)</td>
</tr>
<tr>
<td>1–3 hrs</td>
<td>13 (20.0)</td>
<td>0 (0.0)</td>
<td>2 (2.9)</td>
</tr>
<tr>
<td>4–8 hrs or more</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Control over behavior</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Full, adequate</td>
<td>4 (6.2)</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Sometimes</td>
<td>18 (27.7)</td>
<td>2 (2.9)</td>
<td>1 (1.6)</td>
</tr>
<tr>
<td>Rarely</td>
<td>34 (52.3)</td>
<td>3 (4.4)</td>
<td>5 (7.4)</td>
</tr>
<tr>
<td>None</td>
<td>13 (19.1)</td>
<td>0 (0.0)</td>
<td>5 (7.4)</td>
</tr>
<tr>
<td>Resistance to behavior</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Always</td>
<td>3 (4.4)</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Frequently</td>
<td>4 (5.9)</td>
<td>0 (0.0)</td>
<td>1 (1.6)</td>
</tr>
<tr>
<td>Sometimes</td>
<td>23 (35.8)</td>
<td>3 (4.4)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>No control, subjective distress</td>
<td>22 (32.4)</td>
<td>2 (2.9)</td>
<td>4 (5.9)</td>
</tr>
<tr>
<td>No control and distress</td>
<td>16 (23.5)</td>
<td>0 (0.0)</td>
<td>6 (8.8)</td>
</tr>
</tbody>
</table>
Excoriation disorder in college students was reported to be 3.8%, and the current rate of clinical BFRBs is 3.2%, and the current rate of clinical BFRBs is 12.3% met the criteria for a BFRB disorder (35). In previous studies, a total of 59.6% met the criteria for subclinical BFRB, and 34 to 64% for nail biting, 9.7 to 10.5% for hair pulling, 20 to 92% for skin picking, 42 to 43% for cheek biting, and 15 to 31% for diurnal tooth grinding. Pathological forms of BFRBs (i.e., those that occur frequently despite attempts to stop, causing physical impact and distress and/or functional impairment) are thought to be considerably less common. However, the prevalence of pathological forms of BFRBs cannot be determined clearly due to difficulties in diagnosis and the reluctance of patients to show their problematic area. Recently, Houghton et al. (35) showed that BFRBs are more prevalent than thought. In a large undergraduate sample, a total of 12.3% met the criteria for a BFRB disorder (35). In previous studies, the current rate of clinical trichotillomania in college students was reported to be 3.2%, and the current rate of clinical excoriation disorder in college students was reported to be 3.8% (36, 37).

In this study, the rate of chronic BFRBs leading to distress or impairment was 3.8%, and the rate of trichotillomania was 0.6%, and the rate of excoriation disorder was 1.2%. However, BFRBs were observed quite frequently in children and adolescents with mental retardation during sampling, but these cases were not included in the study because mental retardation was an exclusion criterion. Coexistence of mental retardation and BFRB seems to be a subject worth investigating, and it is important to examine this coexistence in the future studies.

Studies have found that trichotillomania, onychophagia, and skin-picking behavior are often associated with attention deficit hyperactivity disorder, obsessive compulsive disorder, and Tourette syndrome (2, 38). There are studies showing attention deficit hyperactivity disorder comorbidity, with a very high frequency of 44.4% (39). In our study, approximately one-third of the patients had at least one comorbid disorder, and, similar to the literature, the most common psychiatric comorbidities were attention deficit hyperactivity disorder, obsessive compulsive disorder, and autism spectrum disorder. Impulsivity has been thought to be one of the main factors underlying BFRBs (40). Furthermore, in DSM-4, trichotillomania was defined under the heading of impulse control disorders. High comorbidity with disorders such as attention deficit hyperactivity disorder and behavioral disorders in which impulsivity is at the forefront suggests that impulsivity may be an important factor.

Studies show that mood and anxiety disorders are also high in BFRBs (41). Embarrassment and a decrease in self-confidence may occur due to the appearance of the hair, nails, or skin. On the other hand, high levels of anxiety can also lead to BFRBs. In our study, although it was not at a clinically pathological level, anxiety and depression scores were high and scores decreased during the treatment process.

The most common BFRB in our sample was nail biting. In nail-biting behavior, subjective awareness was higher, and urges prior to the behavior and release after the behavior were found to be higher. In addition, impairment was higher in onychophagia. Onychophagia can be a simple habit, or it can be severe enough to cause significant tissue damage, resistant paronychia (42), dental malocclusion, and gingival damage. The fact that this study was conducted on a clinical sample and that the frequency of behavior was highest in onychophagia may have led to higher impairment.
Clinical characteristics and treatment of trichotillomania

Focused and automatic subtypes are mentioned for BFRBs. In the focused subtype, the person is aware of the behavior and it is often accompanied by high emotional tension. In the automatic subtype, the person is unaware of the behavior. It has been reported that most patients with trichotillomania are of the automatic type, whereas most patients with excoriation disorder are of the focused type (43). In our study, a scale was not used for subtyping but, based on the answers given to subjective questions, 63% of the cases reported that they were aware of the behavior and 50% did not have control over the behavior. The highest awareness rate was found in onychophagia.

Various conditions have been reported to trigger BFRBs. It has been reported that, in some patients, sensory triggers related to the hair or scalp, emotional triggers such as anxiety, distress, and anger, and cognitive triggers related to hair and appearance may initiate the behavior, whereas in other patients the behavior may begin during calming activities such as watching TV or reading a book (44, 45). The participants stated that the situations that increase the behaviors the most are watching TV, a feeling of emptiness, and studying. Considering that attention deficit hyperactivity disorder is accompanied by a high rate of BFRBs, distraction and/or stress during lessons may be a trigger for BFRBs. Identifying triggers is important in planning behavioral interventions and preventing relapses.

Treating BFRBs can be challenging because currently there are no FDA-approved drugs for treatment. Studies show that several pharmacological agents from various drug classes can be used for treatment. Antidepressants, atypical antipsychotics, opioid agonists, glutamate modulators, anticonvulsants, and cannabinoid agonists are the most commonly used drugs (46–49). Because of the relationship between obsessive compulsive disorder and BFRBs, treatment options may have similarities with those for obsessive compulsive disorder. One randomized controlled trial shows that fluoxetine at a mean dosage of 55 mg/day improved excoriation disorder symptoms more than placebo, and another randomized controlled trial shows that 20 mg/day of citalopram decreased excoriation disorder symptoms more than placebo (50, 51). On the other hand, two double-blind crossover studies have shown poor efficacy of fluoxetine in the treatment of trichotillomania (52, 53). Atypical antipsychotics, lamotrigine, N-acetylcysteine, and naltrexone have been tested in BFRBs, but the results have been inconsistent (46).

In this study, the most commonly used treatments were SSRIs, atypical antipsychotics, a combination of these, and habit reversal training. Considering the variables related to treatment choice, it is noteworthy that the use of SSRIs and atypical antipsychotics is higher in children with higher baseline anxiety levels. Although there was no significant difference between the choice of treatment and the course of psychiatric measures, significant improvement was found in functionality after treatment. Although studies conducted to date have not shown consistent results on the efficacy of psychopharmacological agents, alleviation of underlying anxiety, impulsivity, and other psychiatric problems may decrease BFRBs and increase functionality.

Our study has several limitations. The most important limitation is the retrospective character of the study design and the small sample size. In this study, improvement was only demonstrated on the CGI and not shown by psychometric scales. If there had been a control group receiving evaluation interviews only, it could have shown a similar trend in improvement. This is also a limitation and may be a point for future research.

Despite these limitations, the use of structured interview methods and the objective evaluations with psychometric measurements during follow-up may be strengths of the study. Despite these shortcomings, the small number of studies on BFRBs increases the importance of studies in this area.

Conclusions

Based on the results of this retrospective cohort study, the most common BFRB is nail biting, and the most common comorbidities are attention deficit hyperactivity disorder, bipolar disorder, obsessive compulsive disorder, and autism spectrum disorder. The most frequent aggravating situations are watching TV, a feeling of emptiness, and studying. Although there is no defined treatment algorithm for children and adolescents that present with BFRB complaints, physicians frequently prescribe pharmacological agents, and the most commonly used pharmacological agents are atypical antipsychotics and SSRIs. Although there was no significant difference between the choice of treatment and the course of psychiatric measures, significant improvement was found in functionality after treatment. Despite not being clinically pathological, anxiety and depression scores were high, and scores decreased during the treatment process.

References


