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Body focused repetitive behavior disorders: Significance of family history Sarah A. Redden, Eric W. Leppink, Jon E. Grant*

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Abstract

Background: The significance of family history in body-focused repetitive behavior disorders (BFRBs) (i.e. trichotillomania and skin picking) has received scant research attention. We sought to understand the clinical and cognitive impact of having a first-degree relative with a BFRB or a substance use disorder (SUD).

Methods: 265 participants with BFRBs undertook clinical and neurocognitive evaluations. Those with a first-degree relative with a BFRB or an SUD were compared to those without on a number of clinical and cognitive measures.

Results: 77 (29.1%) participants had a first-degree family member with a BFRB and 59 (22.2%) had a first-degree family member with an SUD. In terms of clinical severity, the amount of time spent picking or pulling per day in the past week was higher among those with a first-degree relative with an SUD. There were a higher rate of ADHD and higher HAM-D scores among those with a positive family history of an SUD. There were no significant cognitive differences based on family history.

Conclusions: These results indicate that among those with BFRBs, having a first-degree family member with an SUD may be associated with a unique clinical and cognitive presentation. Whether family history also is associated with differential response to treatments awaits further research.

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1. Introduction

Trichotillomania is an often debilitating psychiatric condition characterized by recurrent pulling out of one's own hair, leading to hair loss and marked functional impairment [1,2]. Skin picking disorder is characterized by the repetitive and compulsive picking of skin which causes tissue damage [3]. Trichotillomania and skin-picking appear to have substantial clinical and possibly even neurobiological similarities, and based on available evidence, have been described as body-focused repetitive behavior disorders (BFRBs) [4].

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Although both trichotillomania and skin picking may seem like simple behaviors, research has demonstrated that BFRBs are complex, highly individualistic disorders [5]. The clinical utility of identifying potential subtypes of BFRBs, therefore, has been examined in the literature, including focused versus automatic behaviors [6], early versus late age at onset [7–9], and comorbidity patterns [10]. One area that has received little attention is the possible importance of family history.

BFRBs have long been thought to have a familial basis [11–17]. Several family studies have reported elevated rates of BFRBs in first-degree relatives of probands with a BFRB [14,18,19]. The suggestion that BFRBs may be familial seems consistent with existing twin studies which suggest that the disorders are heritable in addition to being familial [20,21].

Although studies have examined the familial aspects of BFRBs, the family history issue is perhaps more complex than initially thought. One family history study of trichotillomania that included a control group found that the first-degree relatives of subjects with trichotillomania were significantly more likely to have substance use disorders (21.6% alcohol and 14.7% drug use disorders) than relatives of non-ill comparison subjects (7.7% alcohol use disorders and 2.2% drug use disorders) [14]. Similarly, a study of 34

patients with skin picking disorder found lifetime alcoholism in 37.5% of first-degree relatives [22].

Thus, the existing literature suggests that BFRBs run in families where we also see high rates of BFRBs and SUDs. Existing data, however, do not provide information as to what, if anything, these types of familial associations may mean for the person with trichotillomania or skin picking disorder. Therefore, understanding the clinical and neurocognitive aspects of BFRBs and how these factors differ between individuals with different types of family histories may be important in order to identify potential clinical and cognitive subtypes, improve neurobiological models, and optimize treatment. The purpose of this study is to investigate whether adults with BFRBs with a first-degree relative with a BFRB or an SUD have a different clinical presentation than those without, and whether analysis of different families' histories has any clinical relevance.

2. Materials and methods

2.1. Subjects

Data from a total of 265 participants taking part in neurocognitive, neuroimaging, or pharmacotherapy trials for the treatment of skin picking or trichotillomania were pooled together for the purposes of this study. Inclusion criteria included male and female outpatients between the ages of 18 and 65 years with a primary diagnosis of either trichotillomania or skin picking disorder (all earlier diagnoses were later reassessed in light of DSM-5 criteria for both disorders). Exclusion criteria included current psychotic disorders, bipolar disorder, or past three-month history of substance use disorders, and an inability to understand study procedures and provide written informed consent. All measures of assessment were taken at baseline prior to the implementation of any treatments. Data were collected from September 2006 through January 2015.

2.2. Assessments

Participants were asked about the presence of lifetime BFRBs and SUDs (which included alcohol and drug use disorders, but not nicotine) in all first-degree relatives. As some picking and pulling behavior may occur in family members without rising to the level of a disorder, only severe picking resulting in chronic lesions and pulling resulting in noticeable alopecia met the definition of skin picking disorder or trichotillomania in a family member. Substance use disorders were defined as the chronic use of drugs or alcohol resulting in either noticeable social and occupational dysfunction or the need for a twelve-step program or formal treatment. All information about relatives came from the proband. No direct evaluations of the first-degree relatives were performed.

Current and lifetime psychiatric comorbidity was assessed using the Structured Clinical Interview for DSM-IV (SCID) disorders [23] and valid and reliable

SCID-compatible modules for impulse control disorders using the Minnesota Impulse Disorders Interview (MIDI) [24]. The diagnosis of ADHD was based on self-report history by the participant during a semi-structured interview.

Participants completed measures regarding the severity of their hair pulling or skin picking. For purposes of this combined dataset, we extracted minutes per day pulling or picking as this single measure could be combined across disorders whereas the disorder-specific scales could not be. In addition, participants were asked if they had previously received any treatment for their picking or pulling behavior.

In terms of formal measures, all participants completed the following at baseline:

- Clinical Global Impression- Severity (CGI) [25]. The CGI is a valid and reliable, 7-item scale used to assess symptom severity. It uses a Likert-scored scale with 1 = "not ill at all" to 7 = "among the most extremely ill." The scale was used to assess only the severity of the BFRB symptoms.
- Sheehan Disability Scale (SDS) [26]. The SDS is a valid and reliable, three-item, self-report scale assessing psychosocial functioning in three areas of life: work, social or leisure activities, and home and family life. Scores on the SDS range from 0 to 30.
- Quality of Life Inventory (QoLI) [27]. The QoLI is a valid and reliable 16-item, self-report positive psychology scale assessing areas of life such as health, love, work, recreation, home, friendships, self-esteem, and standard of living.
- Current depressive and anxiety symptoms were assessed using the 17-item *Hamilton Depression Rating Scale* [28] and the *Hamilton Anxiety Rating Scale* [29], respectively.

2.3. Cognitive testing

2.3.1. Stop-signal task (SST)

The Stop-signal task from the CANTAB is a well-validated task quantifying the ability to suppress impulsive responses [30]. This task provides a sensitive estimate of the time taken by the participant's brain to stop a prepotent response, referred to as the 'Stop-signal reaction time' (SSRT).

2.3.2. Intra-dimensional/Extra-dimensional Set Shift task (IDED) [31]

The IDED task in the CANTAB includes aspects of rule learning and behavioral flexibility, and was derived from the Wisconsin Card Sort Test [32]. There are nine stages to the task, requiring different components of set acquisition, reversal, and flexibility.

2.4. Data analysis

Based on the family history, participants were categorized into the following groups: family history positive/negative for a BFRB and family history positive/negative for an SUD. We used one-sample Kolmogorov–Smirnov Tests to test for

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