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Evidence-Based Psychosocial Treatments for Pediatric Body-Focused Repetitive Behavior Disorders

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Abstract: Habits, such as hair pulling and thumb sucking, have recently been grouped into a category of clinical conditions called body-focused repetitive behavior disorders (BFRBDs). These behaviors are common in children and, at extreme levels, can cause physical and psychological damage. This article reviews the evidence base for psychosocial treatment of pediatric BFRBDs. A review of academic databases and published reviews revealed 60 studies on psychosocial treatments for pediatric BFRBDs, 23 of which were deemed suitable for review. Based on stringent methodological and evidence base criteria, we provided recommendations for each specific BFRBD. Individual behavior therapy proved probably efficacious for thumb sucking, possibly efficacious for several conditions, and experimental for nail biting. Individual and multicomponent cognitive-behavioral therapy was named experimental for trichotillomania and nail biting, respectively. No treatment met criteria for well-established status in the treatment of any BFRBD. Recommendations for clinicians are discussed. Reasons for the limitations of existing research in children and adolescents are explored. Several recommendations are presented for future pediatric treatment research on BFRBDs.

Body-focused repetitive behaviors (BFRBs) are habits directed at one's own body. When such behaviors occur at a high frequency or intensity, they can produce physical and/or psychosocial problems. If BFRBs result in impairment, they can be considered BFRB

disorders (BFRBDs). BFRBDs are currently classified under various diagnostic labels in the Obsessive-Compulsive and Related Disorders category of the *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.; American Psychiatric Association, 2013). Examples include chronic hair pulling (trichotillomania [TTM]), skin picking (excoriation disorder [ExD]), nail biting (onychophagia), cheek biting, and thumb sucking. These conditions typically manifest during childhood or adolescence (Bohne et al., 2005; Woods & Miltenberger, 1996 Woods, D. W. & Miltenberger, R. G. (1996), and evidence suggests that these conditions are similar in many ways, despite topographical differences (Teng, Woods, Twohig, & Marcks, 2002; Woods, Miltenberger, & Flach, 1996).

In most cases, BFRBs are common, harmless habits. For example, research has found high rates of body-focused habits in college populations: 64% for nail biting, 38% for knuckle cracking, and 30% for finger tapping (Hansen, Tishelmian, Hawkins, & Doepke, 1990). Other studies have found rates of occasional skin picking in college students between 78% and 90% (Bohne et al., 2002; Keuthen et al., 2000). Woods et al. (1996) surveyed 426 college students and asked how often they engaged in many repetitive habits. Rates of these behaviors were high, such as 11% for hair pulling, 34% for nail biting, 43% for chewing on parts of the mouth, 34% knuckle cracking, and 15% for teeth grinding. However, when a more stringent cutoff was applied (i.e., engaging in the behavior five times per day), rates dropped considerably (e.g., 3.2% for hair pulling and 10.1% for nail biting). Evidence also suggests that habit behaviors are relatively common in young children and tend to decrease in prevalence with age. Evans et al. (1997) showed that rates of compulsive and habitual behaviors in 2- to 4-year-olds were significantly higher than in older children (5–6 years of age), and Foster (1998 Foster, L. G.) found that teachers reported significant decreases in BFRBs with age.

As opposed to the occasional and benign BFRBs present in many individuals, BFRBDs can lead to substantial physical and medical consequences. TTM can lead to scarring and hair loss, and for the minority (13%) who ingest pulled hairs (Grant & Odlaug, 2008), masses of undigested hair (known as trichobezoars) can form and lead to significant medical complications such as bowel obstruction, intestinal bleeding, acute pancreatitis, obstructive jaundice, or a perforated bowel (Bouwer & Stein, 1998; Muller, 1987). Other BFRBDs can result in repetitive strain injuries, dental malocclusions, permanent scarring, infections, and excessive bleeding (Bohne et al., 2005; Jones, Swearer, & Friman, 1997; Silva & da Fonseca, 2003; Snorrason & Woods, 2014; Wilhelm et al., 1999).

The negative psychosocial effects of BFRBDs also can be substantial. Peers view hair pulling negatively (Woods, Fuqua, & Outman, 1999), and pulling appears to result in

emotional consequences (Franklin et al., 2008; Soriano et al., 1996). This may lead some, particularly children, to deny engaging in the behaviors (Foster, 1998). In addition, children with TTM report disruption in their ability to maintain social relationships and, as a result, may avoid social events (Franklin et al., 2008);; Walther et al., 2014; Woods et al., 2006), possibly because hair pulling and its affects might become less socially acceptable as one matures. Several large-scale surveys have found moderate psychological difficulties (e.g., depression, anxiety, and stress) in children and adolescents with hair pulling and skin picking (Franklin et al., 2008; Tucker et al., 2011; Walther et al., 2014).

SUMMARY OF EVIDENCE-BASED PSYCHOSOCIAL INTERVENTION STUDIES FOR PEDIATRIC BFRBS

Various pharmacological and psychosocial treatments for BFRBDs have been explored, particularly for TTM and thumb sucking, but the literature is sparse. Several recent meta-analyses have evaluated the efficacy of interventions for adults with TTM (Bloch et al., 2007; McGuire et al., 2014) and ExD (Gelinas & Gagnon, 2013), but similar analyses have not been performed for pediatric populations.

In line with the goal of producing evidence base updates of psychosocial treatments for pediatric psychiatric disorders (Southam-Gerow & Prinstein, 2014), this review summarizes the empirical status of psychosocial treatments for BFRBDs in children and adolescents. Although little research exists on this topic, the article can serve as the foundation for future evidence base updates, spur more basic research and rigorous clinical trials, and serve as a source of *empirically informed approaches* for clinicians who encounter BFRBDs in pediatric populations.

To facilitate the review, a comprehensive search of the psychosocial treatment literature for child and adolescent BFRBs was conducted. It is important to note that although some forms of medication have been shown to be at least partially effective for treating BFRBs (Bloch et al., 2007; Gelinas & Gagnon, 2013; McGuire et al., 2014), such studies are not reviewed in this text.

Evidentiary criteria described by Southam-Gerow and Prinstein (2014) were used to evaluate the state of the literature. These criteria were based upon the American Psychological Association evidence base guidelines (Chambless & Hollon, 1998). As shown in Table 1, these criteria are formulated on a five-level system, including *well-established* (Level 1), *probably efficacious* (Level 2), *possibly efficacious* (Level 3), *experimental* (Level

4), and *of questionable efficacy* (Level 5). The guidelines specify a number of methodological criteria that are to be used to evaluate the literature and determine a treatment's appropriate level of empirical support (see Table 1).

TABLE 1 *Journal of Clinical Child & Adolescent Psychology* Evidence Base Updates—Evidence-Based Treatment Evaluation Criteria

Methods Criteria

M.1. Group design: Study involved a randomized controlled design

M.2. Independent variable defined: Treatment manuals or logical equivalent were used for the treatment

M.3. Population clarified: Conducted with a population, treated for specific problems, for whom inclusion criteria have been clearly delineated

M.4. Outcomes assessed: Reliable and valid outcome measures gauging the problem targeted (at a minimum) were used

M.5. Analysis adequacy: Appropriate data analysis were used and sample size was sufficient to detect expected effects

Level 1: Well-Established Treatments

Evidence Criteria

1.1. Efficacy demonstrated for the treatment by showing the treatment to be either:

1.1.a. Statistically significantly superior to pill or psychological placebo or to another active treatment

OR

1.1.b. Equivalent (or not statistically significant) to an already well-established treatment in experiments

AND

1.1.c. In at least two independent research settings and by two independent investigatory teams demonstrative efficacy

AND

1.2. All five of the Methods Criteria

Level 2: Probably Efficacious Treatments

Evidence Criteria

2.1. There must be at least two good experiments showing the treatment is superior (statistically significant) to a waitlist control group

OR

2.2. One or more good experiments meeting the Well-Established Treatment level with the exception of having been conducted in at least two independent research settings and by independent investigatory teams

AND

2.3. All five of the Methods Criteria

Level 3: Possibly Efficacious Treatments

Evidence Criteria

3.1. At least one good randomized controlled trial showing the treatment to be superior to a waitlist or no-treatment control group

AND

3.2. All five of the Methods Criteria

Level 4: Experimental Treatments

Evidence Criteria

4.1. Not yet tested in a randomized controlled trial

OR

4.2. Tested in one or more clinical studies but not sufficient to meet level 3 criteria

Level 5: Treatments of Questionable Efficacy

5.1. Tested in good group-design experiments and found to be inferior to other treatment group and/or waitlist control group, that is, only evidence available from experimental studies suggests the treatment produces no beneficial effect.

Note. Adapted from Southam-Gerow and Prinstein (2014).

For this review, several reference sources for treatment studies of BFRBDs were utilized. Authors made the decision to exclude self-biting and self-mouthing behaviors (in the self-injurious and stereotypic sense), as these are almost invariably associated with autism spectrum disorders and intellectual disability, whereas BFRBDs that commonly manifest in typically developing children are reviewed in the present article. Studies were identified through searches of PsycINFO, ISI Web of Science, and Google Scholar (keywords: *trichotillomania, hair pulling, skin picking, excoriation, dermatillomania, nail biting, onychophagia, nail picking, cheek biting, thumb sucking* and *treatment* or *therapy* and *child* or *adolescent* or *pediatric*). In addition, the authors examined previously identified review articles, meta-analyses, and their reference sections in order to screen for other published trials.

Based on these search criteria, 60 initial papers were identified. After an extensive examination of each study, 23 studies were deemed suitable for an evidence base review. These studies are summarized in Tables 23456, and Table 7 details the methodological criteria met by each study. To be included for review, studies had to (a) utilize group-designs or controlled and methodologically rigorous single-subject designs, (b) include at least one experimental condition using a psychosocial therapeutic technique, (c) include (at least in part) children and adolescents younger than age 18, and (d) be written in English. Reasons for ruling out 37 studies included not clearly containing adolescents or children within the sample ($n = 2$), lack of a control condition ($n = 21$), lack of psychosocial therapy ($n = 2$), and lack of reliable assessment methodology ($n = 12$). According to guidelines provided by Southam-Gerow and Prinstein (2014), summarizations of treatment efficacy were collapsed across type of treatment and format (i.e., individual behavior therapy), rather than naming specific therapies (i.e., habit reversal training). However, specific clinical trials and "brand-name" therapies are described in text.

Studies are described in text and in summary tables. Within the text and tables, the studies are ordered according to evidentiary base.

TABLE 2 Summarization of Reviewed Studies of TTM

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Franklin, Edson, Ledley, and Cahill (2010)	TTM	24 children and adolescents	HRT vs. Minimal Attention Control	RCT	NIMH-TSS	Significant improvement for treatment group, no significant group effects for control	Not reported	No significant difference from Posttreatment for follow-up
Tolin et al. (2007)	TTM	46 children and adolescents	Open label CBT	Open trial	NIMH-TSS, NIMH-TIS, CGI-S, CDI, MASC	Significant reductions on all outcome measures during acute treatment	NIMH-TSS – partial $\eta^2 = .389$, NIMH-TIS – partial $\eta^2 = .353$, CGI-S – partial $\eta^2 = .336$, CDI - partial $\eta^2 = .251$, MASC – partial $\eta^2 = .254$	Treatment effects maintained on NIMH-TSS and CDI. Partial relapse on NIMH-TIS, CGI-S, and MASC
Azrin, Nunn, & Frantz (1980)	TTM	Four children among 34 persons	HRT vs. MNP	RCT	Self-reported frequency	Significant advantage for HRT	91% reduction at 4-month	87% at 22-month
Altman et al. (1982)	TTM	3-year-old girl	Assorted Behavioral Techniques (Directed at thumb sucking)	Reversal	Hair count with reliability	Reduced to near-zero rates	N/A	Results maintained at 20 months
Blum et al. (1993)	TTM	Two children	Assorted Behavioral Techniques	Multiple baseline and Reversal	Observed pulling (with reliability)	Substantial reductions when treatment administered	N/A	Gains maintained through 6 and 12 months
Rapp et al. (1998)	TTM	Three adolescents	Simplified HRT	Multiple Baseline	Videotaped observation and coding (with reliability), and photographic measures	Reductions in time spent pulling hair, and significant improvements in hair appearance	N/A	Gains maintained at follow-up for two out of three participants

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Massong et al. (1980)	TTM	3-year-old boy	Assorted Behavioral Techniques	Reversal	Direct Observation with reliability	Reduced to zero	N/A	Results maintained at 2 months

Note: TTM = trichotillomania; RCT = randomized controlled trial; CBT = cognitive-behavioral therapy; HRT = habit reversal training; MNP = massed negative practice; NIMH-TSS = National Institutes of Mental Health Trichotillomania Severity Scale; NIMH-TIS = National Institutes of Mental Health Trichotillomania Improvement Scale; CGI-S = Clinical Global Impressions–Severity; CDI = Childhood Depression Inventory; MASC = Multidimensional Anxiety Scale for Children.

TABLE 3 Summarization of Reviewed Studies of ExD

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Christensen and Sanders (1987)	Thumb sucking	30 children	Habit reversal and DRO	RCT	Clinical observation with reliability	Habit Reversal and DRO equivalent but better than waitlist	Not reported	Results maintained at 3 months
Friman and Leibowitz, (1990)	Thumb sucking	22 children	Aversive taste & reward system	RCT	Parent recording (with reliability)	Significant reduction compared to wait-list control	Not reported	Unclear
Azrin, Nunn, & Frantz-Renshaw (1980)	Thumb sucking	18 children and adolescents	Habit reversal (behavior therapy)	RCT	Parent-reported frequency	Positive results of for habit reversal on mean percentage reduction in thumb sucking	N/A	Effects maintained for 20 months
Houten and Rolider (1984)	Thumb sucking	10 children	Response prevention and reward system	Multiple baseline	Parent recording with reliability	Reduced to near-zero rates for all children	N/A	Results maintained at varying follow-ups
Friman et al. (1986)	Thumb sucking	Three families with seven children	Aversive taste conditioning	Multiple baseline	Parent recording with reliability	Reduced to near-zero rates for all children	N/A	Zero rates maintained at 3- and 6-month follow-ups

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
		and adolescents						
Friman and Hove (1987)	Thumb sucking	Two young boys	Aversive taste treatment (behavior therapy)	Multiple baseline	Video observation of behavior (with reliability)	Substantial reduction in thumb sucking (and hair pulling)	N/A	Treatment effects maintained through 12 months
Rapp et al. (1999)	Thumb sucking	Two 5-year old fraternal twin boys	Simplified HRT	Multiple baseline	Video observation checked for interobserver agreement	Substantial reductions in thumb sucking for both participants	N/A	Near-zero levels for both participants at 6-months
Watson, Dittmer, & Ray (2000)	TTM and thumb sucking	18-month-old boy	Attention reflection and aversive taste treatment	ABCAC	Parent observation with reliability	Behaviors reduced in attention reflection, reduced further in aversive taste treatment	N/A	No follow-up
Friman (1990)	Thumb sucking	8 children	Aversive taste conditioning and positive reinforcement	Multiple baseline	Parent recording	Reduced to near-zero rates for all children	N/A	Zero rates maintained at 3- and 6-month follow-up
Watson and Allen (1993)	TTM and thumb sucking	5-year-old girl	Combination of behavioral techniques	Alternating treatment with reversals	Parent recording and alopecia measurement	Complete elimination of both behaviors	N/A	Symptom free at 30-month follow-up

Note: TTM = trichotillomania; RCT = randomized controlled trial; HRT = habit reversal training; MNP = massed negative practice; DRO = differential reinforcement of other.

TABLE 4 Summarization of Reviewed Studies of Nail Biting

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Ergun et al. (2013)	Nail biting	103 third-grade children	"Healthy Nails Program" or CBT	Quasi-experimental design	Clinician-rated measure and photographs	Significantly more treated clients showing total remission and total bitten nails	Not reported	Gains maintained at follow-up
Nunn and Azrin (1976)	Nail biting	13 clients (two children)	HRT	RCT	Photographs	Reduced to near-zero	N/A	Results maintained at 16 weeks
Woods et al. ([1999](#))	Nail biting and thumb sucking	26 children and adolescents (four with nail biting)	HRT	RCT	Home observation and rating	Habit reversal outperformed control	N/A	Effects maintained for 6 weeks

Note: RCT = randomized controlled trial; CBT = cognitive-behavioral therapy; HRT = habit reversal training.

TABLE 5 Summarization of Reviewed Studies of Cheek Biting

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Azrin et al. (1982)	Destructive oral habits (including cheek biting)	Three children out of 10 total participants	HRT vs. MNP	RCT	Self- and parent-monitoring	60% reduction in MNP vs. 99%–100% reduction in HRT	N/A	HRT gains maintained at 6 months and one patient slightly remitted at 22 months
Jones et al. (1997)	Cheek biting	15-year-old male	Abbreviated habit reversal (behavior therapy)	ABAB (reversal)	Blood spots on handkerchief	Substantial reduction in cheek biting (near zero levels) during administration of treatment	N/A	Maintained at 2 months

Note: HRT = habit reversal training, MNP = massed negative practice; RCT = randomized controlled trial.

TABLE 6 Summarization of Reviewed Studies of Thumb Sucking

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
Christensen and Sanders (1987)	Thumb sucking	30 children	Habit reversal and DRO	RCT	Clinical observation	Habit Reversal and DRO	Not reported	Results maintained

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
					with reliability	equivalent but better than waitlist		d at 3 months
Friman and Leibowitz, (1990)	Thumb sucking	22 children	Aversive taste & reward system	RCT	Parent recording (with reliability)	Significant reduction compared to wait-list control	Not reported	Unclear
Azrin, Nunn, & Frantz-Renshaw (1980)	Thumb sucking	18 children and adolescents	Habit reversal (behavior therapy)	RCT	Parent-reported frequency	Positive results of for habit reversal on mean percentage reduction in thumb sucking	N/A	Effects maintained for 20 months
Houten and Rolider (1984)	Thumb sucking	10 children	Response prevention and reward system	Multiple baseline	Parent recording with reliability	Reduced to near-zero rates for all children	N/A	Results maintained at varying follow-ups
Friman et al. (1986)	Thumb sucking	Three families with seven children and adolescents	Aversive taste conditioning	Multiple baseline	Parent recording with reliability	Reduced to near-zero rates for all children	N/A	Zero rates maintained at 3- and 6-month follow-ups
Friman and Hove (1987)	Thumb sucking	Two young boys	Aversive taste treatment (behavior therapy)	Multiple baseline	Video observation of behavior (with reliability)	Substantial reduction in thumb sucking (and hair pulling)	N/A	Treatment effects maintained through 12 months
Rapp et al. (1999)	Thumb sucking	Two 5-year old fraternal twin boys	Simplified HRT	Multiple baseline	Video observation checked for interobserver agreement	Substantial reductions in thumb sucking for both participants	N/A	Near-zero levels for both participants at 6-months
Watson, Dittmer, & Ray (2000)	TTM and thumb sucking	18-month-old boy	Attention reflection and aversive taste treatment	ABCAC	Parent observation with reliability	Behaviors reduced in attention reflection, reduced	N/A	No follow-up

Study	Disorder	Sample	Treatment	Trial Type	Measures	Results	Effect Size	Follow-Up
						further in aversive taste treatment		
Friman (1990)	Thumb sucking	8 children	Aversive taste conditioning and positive reinforcement	Multiple baseline	Parent recording	Reduced to near-zero rates for all children	N/A	Zero rates maintained at 3- and 6-month follow-up
Watson and Allen (1993)	TTM and thumb sucking	5-year-old girl	Combination of behavioral techniques	Alternating treatment with reversals	Parent recording and alopecia measurement	Complete elimination of both behaviors	N/A	Symptom free at 30-month follow-up

Note: TTM = trichotillomania; RCT = randomized controlled trial; HRT = habit reversal training; MNP = massed negative practice; DRO = differential reinforcement of other.

TABLE 7 Methods Criteria Checklist for Included Studies

Study	Condition	M1	M2	M3	M4	M5
Franklin, Edson, Ledley, & Cahill (2010)	TTM	X	X	X	X	X
Tolin et al. (2007)			X	X	X	X
Azrin, Nunn, & Frantz (1980)		X	X	X		X
Altman et al. (1982)		X	X	X	X	
Blum et al. (1993)		X	X	X	X	
Rapp et al. (1998)		X	X	X	X	
Massong et al. (1980)			X	X		X
Cavalari et al. (2013)	ExD		X	X	X	X
Ergun et al. (2013)	Nail biting		X	X		X
Nunn and Azrin (1976)		X	X	X		X
Woods et al. (1999)		X	X		X	X
Jones et al. (1997)	Cheek		X	X		X
Azrin et al. (1982)	biting	X	X	X		X
Christensen and Sanders (1987)	Thumb	X	X	X	X	X
Friman and Leibowitz (1990)	sucking	X	X	X	X	X
Azrin, Nunn, & Frantz-Renshaw (1980)		X	X	X		X
Houten and Rolider (1984)			X	X	X	X
Friman et al. (1986)			X	X	X	X
Friman and Hove (1987)			X	X	X	X
Rapp et al. (1999)			X	X	X	X
Watson, Dittmer, & Ray (2000)			X	X	X	X
Friman (1990)			X	X		X
Watson and Allen (1993)			X	X		X

Note: M1 = group design; M2 = independent variable defined; M3 = population clarified; M4 = outcomes assessed; M5 = analysis adequacy.

Based on this review, the only psychosocial treatments meeting criteria for *probably efficacious, possibly efficacious*, or *experimental* levels of evidence were behavioral or cognitive-behavioral. The theoretical foundation of behavioral therapies is based on operant and respondent learning principles. Human behavior is viewed as a function of its antecedents (e.g., discriminative stimuli and establishing operations) and consequences (e.g., reinforcers or punishers). Antecedents signal the availability of rewarding or punishing consequences for a given behavior, and consequences maintain that behavior. The behavioral model posits that BFRBDs are habitual behaviors originally acquired by and periodically maintained through alleviating aversive mental or emotional states (e.g., stress, anxiety) and/or providing tactile stimulation (e.g., pleasurable sensations).

Although early methods of BFRBD treatment relied on nonconstructive approaches, such as slapping the hands or applying solutions with unpleasant taste to the skin (Friman & Hove, 1987; Vargas & Adesso, 1976), more recent methods have sought to disrupt the maintaining variables through several different methods. Response prevention and stimulus control techniques, procedures designed to increase the effort involved in performing the behavior or to attenuate the sensory stimulation created by the behavior, are also used. Examples include wearing mittens (Deaver, Miltenberger, & Stricker, 2001) or a special orthodontic retainer (Silva & da Fonseca, 2003) and limiting time spent in situations associated with increased frequencies of the behavior. Behavior therapists also often provide or encourage parents to apply verbal praise and reinforcement for successful completion of nonhabitual or competing behaviors—a practice known as social support. Thus, behavior therapy can include aversive conditioning methods, response prevention, and stimulus control, as well as social reinforcement and reward systems. Current behavioral treatments have integrated several of these empirically supported methods into therapeutic packages, such as Habit Reversal Training (HRT; Azrin & Nunn, 1973). HRT includes three main components: awareness training, competing response training, and social support. Awareness training involves teaching the participant to become more aware of when they engage in or are about to engage in the behavior. Competing response training involves teaching the participant to do a behavior that is physically incompatible with the BFRB when he or she becomes aware that the BFRB is about to occur. The participant is then asked to hold that behavior for a fixed period (e.g., 1–3 min). Social support involves identifying a support person (typically the parent) to provide the child with praise and other reinforcement for engaging in therapeutic exercises. Recently, cognitive techniques have been implemented alongside HRT, in a format similar to

standard cognitive-behavioral therapy (CBT; Ninan et al., 2000; Tolin et al., 2007). CBT can include behavioral components but also involves techniques aimed at helping the individual cope with or mindfully accept maladaptive thoughts, emotions, and bodily sensations that are thought to trigger BFRBDs.

REVIEW OF THE PSYCHOSOCIAL TREATMENT LITERATURE IN PEDIATRIC BFRBS

Trichotillomania

Seven studies were identified that used group-design or controlled single-subject design methodologies for pediatric TTM. Two other studies (Watson & Allen, 1993; Watson, Dittmer, & Ray, 2000) measured reductions in hair pulling, but because the primary target of treatment was thumb sucking they were reviewed in the thumb-sucking section. HRT was tested in three of the seven studies, assorted behavioral techniques (e.g., attention reflection, stimulus control, aversive conditioning, response prevention) in three studies, and CBT in one study. CBT for pediatric TTM is a heterogeneous treatment package that shares several components with HRT, such as awareness training and competing response training, but also includes stimulus control and several cognitive therapy techniques, such as cognitive restructuring and covert modeling (Franklin & Tolin, 2007). Studies that used group-designs contributed the most to the evidence base for pediatric TTM and are reviewed first.

Franklin, Edson, Ledley, and Cahill (2011) tested HRT against a minimal attention control condition (e.g., psychotherapy placebo) in 24 children and adolescents with clinical hair pulling. The study met all five methodological criteria. Improvement was assessed using two psychometrically sound, clinician-rated outcome measures (National Institutes of Mental Health Trichotillomania Severity Scale and Clinical Global Impressions-Severity Scale), with masked independent evaluators conducting all assessments. Results showed significant improvement in the behavior therapy group as compared to no significant improvements in the control condition. In addition, those in the behavior therapy condition showed maintenance of gains at a 16-week follow-up. Tolin et al. (2007) performed an open trial of CBT in 46 children and adolescents with TTM. Therapy consisted of HRT along with cognitive techniques, such as cognitive restructuring, relapse prevention, and covert modeling. The researchers used several psychometrically sound assessment methods, measuring hair pulling along with comorbid depression and anxiety. Large and significant reductions on all measures were found between baseline and posttreatment (all partial η^2

effect sizes >.25). Treatment effects were maintained at follow-up on severity (National Institutes of Mental Health-Trichotillomania Severity Scale) and depression indices (Childhood Depression Inventory), but partial relapse occurred on measures of therapeutic response (e.g., National Institutes of Mental Health–Trichotillomania Improvement Scale, Clinical Global Improvement–Severity Scale) and anxiety (Multidimensional Anxiety Scale for Children). In the final randomized controlled trial (RCT), Azrin, Nunn, and Frantz (1980) tested HRT against Massed Negative Practice (MNP). MNP involves coaching the child to actively perform the symptom repetitively and was thought to paradoxically promote a process called "reactive inhibition." Among the 34 participants in the trial, only four were children, and the study used self-reported frequency of hair pulling as its primary outcome variable. These methodological problems prevent the study from being included in summary recommendations. However, it is worthy to note that all children and 91% of the total participants in the HRT condition achieved significant reductions in symptoms, as compared to negligible effects in the MNP condition. Gains were maintained in 87% of HRT individuals at 22-month follow-up.

In the first of several studies using single-subject designs, Altman, Grahs, and Friman (1982) used attention reflection (verbally praising appropriate behavior and ignoring hair pulling) and aversive taste treatment (by applying a bad tasting substance to the thumb) to treat a 3-year-old girl with TTM and thumb sucking. The authors suspected that thumb sucking and hair pulling were covarying behaviors, meaning they are linked in a behavioral sequence in which one does not occur without the other. Using a reversal design, the authors showed that the strategies substantially reduced hair pulling, and gains were maintained at a 20-month follow-up. Blum, Barone, and Friman (1993) used a multiple baseline with an embedded reversal design to test parent nurturing (reinforcement for positive, playful behaviors), hair-pulling-contingent punishment (time-out or verbal reprimand), and response prevention (gloves, sitting on hands, gripping a pencil) in two children. Results showed substantial reductions in hair pulling when treatment was administered, and gains were maintained through a 12-month follow-up in both children. Rapp and colleagues tested HRT in a multiple baseline design (Rapp, Miltenberger, Long, Elliot, & Lumley, 1998). Three adolescents were provided simplified HRT and were assessed through video observation and coding (reliability checks were conducted). All participants showed substantial reductions in time spent pulling hair, and independent evaluators rated significant improvements in hair appearance over the course of therapy. Gains were maintained at follow-up in two of three participants. Massong, Erwards, Range-Sitton, and Hailey (1980) tested attention reflection (parent verbally reinforcing appropriate play behavior and ignoring hair pulling) and response prevention (cutting the hair close to the scalp) in a 3-year-old boy. Using an ABAC design (Barlow,

Nock, & Hersen, 2008), in which each treatment was introduced between baseline reversals, the behavior was reduced to near-zero levels and maintained at 2-month follow-up. It should be noted that the two of the previously mentioned studies used 3-year-old children with TTM as participants (Altman et al., 1982; Massong et al., 1980), and some have suggested that very young children with hair pulling might represent a distinct group from those who begin hair pulling later in childhood, such that they have a more time-limited course and favorable prognosis (Lewin et al., 2009; Santhanam, Fairley, & Rogers, 2008; Swedo et al., 1992; Tay, Levy, & Metry, 2004). This caveat may limit the generalizability of results from those two studies.

In critically reviewing treatment trials for pediatric TTM, it appears that individual behavior therapy (e.g., HRT and/or other behavioral techniques) possesses the most positive empirical evidence, making it a *possibly efficacious* treatment. Individual CBT should be considered an *experimental* treatment. Massed negative practice, however, must be labeled as having *questionable efficacy*, given that it showed no significant effects (Azrin, Nunn, & Frantz, 1980). See Table 8 for a summary of recommendations. No treatment, not even behavior therapy, can be said to be probably efficacious or well-established because of the limited state of the current literature. Although two RCTs evaluated individual behavior therapy for pediatric TTM, one (Azrin, Nunn, & Frantz, 1980) suffered from several methodological limitations, including reliance on a mixed sample with children and adults and lack of reliable and valid outcome assessment measures. As a result, only one study satisfied all five of the stated methods criteria necessary for the *possibly efficacious, probably efficacious*, or *well-established* levels.

TABLE 8 Evidence-Base Level for Pediatric BFRBDs

BFRBD	Level 1: Well-Established	Level 2: Probably Efficacious	Level 3: Possibly Efficacious	Level 4: Experimental	Level 5: Of Questionable Efficacy
Trichotillomania	—	—	Individual behavior therapy	Individual cognitive-behavior therapy	Individual massed negative practice
Excoriation	—	—	—	Individual behavior therapy	—
Nail biting	—	—	—	Individual behavior therapy and multicomponent cognitive-behavior therapy	—
Cheek biting	—	—	—	Individual behavior therapy	—
Thumb sucking	—	Individual behavior therapy	—	—	—

Note: BFRBDs = body-focused repetitive behavior disorders.

Excoriation Disorder

There currently exist no group-design studies for pediatric skin picking, but one single-subject trial has been conducted. Cavalari, DuBard, and Luiselli ([2013]) tested a simplified version of HRT, consisting of competing response training and differential reinforcement (i.e., social support), with a 17-year-old girl with autism spectrum disorder using an ABAB reversal and fading design. Implementation of all therapeutic components reduced skin picking to near-zero levels, and the gains were maintained through 4-month follow-up. Yet, the fact that the participant from Cavalari et al. ([2013]) was autistic limits the generalizability of their findings to typically developing children.

Based on the limited evidentiary support for psychosocial treatments for ExD, individual behavior therapy possesses *experimental* status (see Table 8). Clearly, more research is needed on the topic, and clinicians who are in search of additional empirical guidance should refer to treatments such as individual CBT and behavior therapy, which have shown efficacy in adults (reviewed in a meta-analysis by Gelinas & Gagnon, [2013]).

Nail Biting

Three group-design studies have evaluated behavioral or cognitive-behavioral psychosocial treatments for pediatric nail biting. Ergun, Toprak, and Sisman ([2013]) evaluated a "healthy nails" program in 103 third-grade Turkish schoolchildren, which through examination of therapeutic methods, appears to be a multicomponent (individual, family, school) version of CBT. In a quasi-experimental design using a clinician-rated measure and independently rated photographs of nail beds, treated children showed significant nail picking remission (56%) and improvements in nail bed hygiene as compared to those in the waitlist control group. Gains were maintained in 64% of children at the 8-week follow-up.

Two other studies tested behavior therapy for pediatric nail biting but contained significant methodological flaws that prevent them from being considered for summary recommendations. Nunn and Azrin ([1976]) tested HRT in a waitlist-controlled RCT with 13 participants, two of whom were adolescents and the rest adults. Although the pediatric sample was too small to enable between-group statistics, and separate results for adolescents were not reported, the findings are still noteworthy. Using self-reported nail biting frequency and photographs of nail length for reliability checks, results showed that participants in the waitlist condition did not reduce their nail biting, whereas participants who underwent treatment reduced biting to near-zero levels (99% reduction in

frequency). Results were maintained for 16 weeks in all but two participants, each of whom reported a single lapse. Woods et al. ([1999](#)) evaluated HRT in four children with nail biting. This trial was part of a larger effort to evaluate HRT for oral-digit habits, and because most participants presented with thumb sucking as their BFRB, the sample of nail biters was small (four treatment vs. one control). Data were not reported specifically for nail biting, creating the same problems as studies with both adults and children. As such, the Woods et al. ([1999](#)) study will not contribute to summary recommendations but is described briefly. Home observation and ratings of behavior frequency were used as outcome measures, and reliability checks were performed. In the analysis of total effects of treatment across both nail biting and thumb sucking, the authors found that HRT outperformed the waitlist control condition, and effects were maintained at 6-week follow-up.

In evaluating the evidentiary support for behavioral and cognitive-behavioral treatments for pediatric nail biting, one quasi-experimental study meeting three of five methodological criteria demonstrated positive effects of multisystemic CBT versus waitlist control (Ergun et al., [2013](#)). Two studies demonstrated positive effects of individual behavior therapy versus waitlist control (Nunn & Azrin, [1976](#); Woods et al., [1999](#)), but the aforementioned methodological limitations prevent them from contributing largely to the pediatric nail biting evidence base. As such, individual behavior therapy was designated as *experimental* and multicomponent CBT as *experimental*. See Table 8.

Cheek Biting

Only two studies were reviewed for the psychosocial treatment of pediatric cheek biting, both testing versions of HRT. Jones et al. ([1997](#)) used a reversal design to test HRT in a 16-year-old boy. The experimenters gave the participant a handkerchief with which he was instructed to blot on his inner cheeks after biting and save the blotted handkerchief in a plastic bag at the end of each day. The number of blood spots on the handkerchief was counted daily as the primary outcome measure. Over the course of treatment phases, the participant showed substantial reductions in cheek biting that approached zero levels, and effects were maintained at 2-month follow-up. Azrin, Nunn, and Frantz-Renshaw (1980) compared HRT to MNP in 10 individuals, including three children, with destructive oral habits. Specifically, the three children in the study all repetitively pushed the tongue against the teeth and licked the lips—technically not cheek biting but arguably functionally equivalent oral habits. Again, the lack of valid group statistics on child participants is a significant limitation. Using both self- and parent monitoring to assess results, the study

reported 60% reductions in problem behaviors in MNP versus 99%–100% reductions in HRT, gains that were largely maintained in the HRT condition at 6 months.

Because the two studies for pediatric cheek biting are limited, in that the first involved only one participant and the other did not involve children with cheek biting specifically, individual behavior therapy techniques meet the *experimental* level of evidentiary support. See Table 8.

Thumb Sucking

Eleven studies were reviewed that tested individual behavioral therapies for pediatric thumb sucking, including four RCTs, five multiple baseline experiments, and two reversal designs. The group-design studies on treatments for pediatric thumb sucking are reviewed first.

Christensen and Sanders (1987) randomized 30 children to receive HRT, differential reinforcement of other behavior (i.e., lack of thumb sucking), or waitlist control. The study met all five methodological criteria. Both active treatments significantly reduced thumb sucking compared to control, as measured by independent observation. No significant differences were seen between treatment groups. Thus, both HRT and differential reinforcement of other behavior (DRO) can be considered superior to control. Friman and Leibowitz (1990) randomized 22 children to a waitlist control or a behavioral treatment involving aversive taste treatment via Stop-zit© (a bad-tasting substance applied to the thumb) plus a reward system. This study also met all five methodological criteria. Using parent observation of thumb sucking, which was checked for reliability, results showed a significant positive effect for the treatment condition at posttreatment. At 1-year follow-up, the parents of all but one child reported complete remission of the behavior. Azrin, Nunn, and Frantz-Renshaw (1980) evaluated HRT compared to aversive taste treatment. Using parent-reported frequency of thumb sucking, which was not checked for reliability, 94% of patients reduced frequency of thumb sucking as compared to 44% in the control condition. Treatment effects were maintained at 20-month follow-up. Woods et al. (1999), described earlier, evaluated HRT for pediatric thumb sucking (along with nail biting) in 22 children and adolescents. Results showed significant positive results for behavior therapy in comparison to the waitlist control, but the researchers failed to report data specifically for thumb sucking.

Other studies used single-subject designs to evaluate the efficacy of HRT, response prevention, differential reinforcement, and aversive taste treatment. Houten and Rolider

(1984) compared response prevention (e.g., wearing boxing gloves) to a reward system (e.g., verbally praising lack of sucking and punishing sucking by loss of playtime) in 10 children and showed that response prevention but not the reward system substantially reduced and nearly eliminated the behavior. Friman, Barone, and Christopherson (1986) used aversive taste treatment (Stop-zit) with seven children and adolescents in a multiple baseline experiment and achieved near-zero rates of thumb sucking for all children. Results were maintained at 3 and 6 months. Using a multiple baseline design, Friman and Hove (1987) tested aversive taste treatment (foul-tasting oil on thumbs) in two male adolescents. Using reliability-checked video observation, substantial reductions in thumb sucking were found when treatment was applied, and remission was maintained through a 12-month follow-up. One study (Rapp, Miltenberger, Galensky, Roberts, & Ellingson, 1999) tested a simplified version of HRT in two 5-year-old fraternal twins using a multiple baseline design. The investigators video recorded the children and coded for thumb sucking, which was checked for reliability. Thumb sucking was substantially reduced in both participants, and gains were maintained at 6 months. Watson, Dittmer, and Ray (2000) tested attention reflection and aversive taste treatment in an 18-month-old with co-occurring thumb sucking and hair pulling. Using a reversal design, they found that attention reflection produced reductions in both behaviors but aversive taste treatment eliminated hair pulling and substantially reduced thumb sucking. Friman (1990) also tested aversive taste conditioning, as well as positive verbal reinforcement, in eight children using a multiple baseline design and parent recording. Results yielded near-zero rates of thumb sucking that maintained through 6-month follow-up. Watson and Allen (1993) tested three behavioral techniques in a 5-year-old girl using a reversal design: aversive taste treatment (Stop-zit), an alarm that activated whenever the thumb was placed in the mouth, and response prevention (wearing an orthotic device that prevented thumb sucking). All forms of treatment reduced thumb sucking, but only response prevention was able to nearly eliminate the behavior. Also of note, the child substantially reduced co-occurring hair pulling throughout the course of treatment.

Despite the fact that thumb sucking has received the most empirical attention among pediatric BFRBDs, and evidence suggesting that behavioral methods are indeed effective, the existing literature contains several flaws that limit individual behavior therapy from receiving well-established status. Of the two group-design studies that evaluated individual therapy for pediatric thumb sucking and met all five of the methods criteria (Christensen & Sanders, 1987; Friman & Leibowitz, 1990) both found behavior therapy superior to waitlist control. However, one found that habit reversal was equivalent to differential reinforcement (Christensen & Sanders, 1987). Also, the third group-design study (Azrin, Nunn, & Frantz-Renshaw, 1980) found significant differences between HRT

and aversive taste treatment but failed to meet all five methods criteria because it lacked reliable assessment methods. Thus, the evidence base lacks an experiment showing that individual behavior therapy is statistically superior to a psychological placebo or other active treatment, but HRT, DRO, and aversion therapy have been shown as superior to control. We therefore designate these three types of individual behavioral therapies with probably efficacious status for pediatric thumb sucking. See Table 8.

EVALUATIVE CONCLUSIONS

Between the five types of BFRBs assessed in this review, the strongest empirical support exists for individual behavior therapy of thumb sucking. For the remaining BFRBDs, the existing evidence for individual behavior therapy can be labeled only as possibly efficacious or experimental. Furthermore, individual and multi-component CBT were only found to have experimental status for TTM and nail biting, respectively. The results of this critical evidence base review of psychosocial treatments for pediatric BFRBs reflect the disappointing state of the current literature. Although the evidence for treatments in adult populations has grown enough to merit meta-analyses (Bloch et al., 2007; Gelinas & Gagnon, 2013; McGuire et al., 2014), treatments for pediatric populations remain on tenuous empirical footing.

Despite the lack of certainty that can be gleaned from pediatric treatment trials, this review has clinical utility. Behavioral approaches do not possess equivalent evidence bases across BFRBDs, but three different types of behavior therapy (HRT, DRO, and aversion therapy) were deemed *probably* efficacious for pediatric thumb sucking. Moreover, BFRBDs share many phenomenological characteristics and are generally considered functionally equivalent (Azrin & Nunn, 1973). This suggests that the core components of behavior therapy, operant learning techniques, may be well suited for pediatric BFRBDs and are currently the best supported option. With greater empirical attention and fewer methodological limitations in future studies, many of these behavioral methods might show greater evidentiary support. Therefore, an *empirically informed approach* to pediatric BFRBs should favor behavioral techniques. Although there is considerable room for other approaches to be proven efficacious, the current standard of care should be individual behavior therapy.

This review also points to the need for more rigorous tests of psychosocial interventions for pediatric BFRBDs. RCTs are the current gold standard for assigning an evidence base for clinical interventions, and several have been conducted for these

populations. However, several notable limitations prevented individual behavior therapy from potentially being given a higher evidentiary standing. First, psychometrically sound measurement instruments should always be employed. Several studies in this review (Azrin, Nunn, & Frantz, 1980; Azrin, Nunn, & Frantz-Renshaw, 1980; Jones et al., 1997) failed to use instruments with strong reported psychometric properties. Second, while single-subject design experiments can be useful for piloting treatments in small samples, many of these papers failed to use clear treatment protocols, making it unclear exactly how treatments were administered. Future trials should always cite treatment manuals or describe their logical equivalents in detail, making it clear how to compare results to similar trials and replicate successful pilot studies via group-design RCTs. Finally, several studies (Azrin, Nunn, & Frantz, 1980; Azrin et al., 1982; Nunn & Azrin, 1976) employed samples consisting of both adults and children, and results were contained in sample sizes that are too small for child and adult samples to be analyzed separately, making it unclear whether results in adult and pediatric populations are mutually generalizable. Future studies targeting pediatric populations should do so exclusively to avoid such issues and contribute more effectively to this underserved literature.

Given the fact that BFRBs commonly manifest during childhood (Bohne et al., 2005), the lack of quality empirical attention is particularly problematic. One possible reason for this neglect might be that pediatric BFRBs are considered "normal" in children and are believe to remit through maturation. Indeed, this seems to be the case as it would be considered fairly normal for a child to tug at his hair, suck her thumb, or pick at his skin, but the same behavior in adults would yield greater scrutiny. Some support for this notion comes from several surveys on young children, children and adolescents, and adults with TTM, which collectively show that hair pulling-related impairment progressively increases throughout development (Franklin et al., 2008; Walther et al., 2014; Woods et al., 2006). However, future research might take several future directions to ensure that children and adolescents with impairing BFRBDs are not continually underserved by the state of the psychosocial literature.

First, researchers should focus their efforts on ways to better predict for whom such behaviors will become problematic in adulthood. One review noted that many adults with TTM reported onset during childhood or adolescence (Snorrason, Belleau, & Woods, 2012), suggesting that some individuals whose hair pulling onsets during childhood may spontaneously remit without significant consequence while another group develops a chronic condition. Perhaps longitudinal studies could be conducted to track pediatric hair pulling from onset and to identify factors that predict classification into each subgroup, making early intervention in the latter individuals feasible. Second, if it holds true that

many children and adolescents with BFRBDs show relatively benign prognoses, existing evidence still suggests that, at least for TTM, these conditions are still associated with significant functional impairment (Franklin et al., 2008; Walther et al., 2014), and interventions are still needed. Accordingly, research should develop brief, manualized treatment protocols that can be implemented in school and community settings. The Ergun, Toprak, & Sisman (2013) study testing multicomponent CBT for pediatric nail biting in the school system could be seen as a promising step in this direction, and the relative simplicity of behavioral interventions (HRT, DRO, and aversion therapy) make them good candidates for school psychologists and social workers who may have limited BFRBD intervention experience. These simple and highly accessible treatments might make for highly efficient and effective options for those who might rarely seek outside professional care. Finally, as this literature matures, it is critical that researchers pay attention to mediators and moderators of treatment effectiveness. Regrettably, despite having Level 2 support, no data exist regarding mediation and moderation in individual behavior therapy for pediatric thumb sucking. The only study to perform such analyses was Franklin, Edson, and Freeman (2010); these authors studied the effect of age on the Franklin et al. (2010) trial of HRT for pediatric TTM. That study found no significant age-related differences in treatment response. In adults with TTM, McGuire et al. (2014) found that the inclusion of mood-related components (i.e., CBT vs. behavior therapy), as well as increased number of treatment sessions significantly increased the effectiveness of behavior therapy. Although the Tolin et al. (2007) trial of CBT for pediatric TTM provided that treatment *experimental* status, future trials should determine whether cognitive components are incrementally effective for pediatric BFRBDs relative to standard behavioral approaches. Finally, although treatment studies on individual BFRBDs are valuable, future efforts should be directed across diagnoses and use designs that enable comparisons across groups. These studies, alongside basic research, might begin to more clearly elucidate the similarities and peculiarities associated with BFRBDs.

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