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Using stimulus fading without escape extinction to increase compliance with toothbrushing in children with autism

Michele R. Bishop [a,*], Amy L. Kenzer [a], Christine M. Coffman [a,1], Courtney M. Tarbox [b], Jonathan Tarbox [a], Taira M. Lanagan [a,2]

[a] *Center for Autism and Related Disorders, Inc., 19019 Ventura Blvd., Tarzana, CA 91356, United States*
[b] *FirstSteps for Kids, Inc., 2447 Pacific Coast Highway, Suite 111, Hermosa Beach, CA 90254, United States*

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ABSTRACT

Routine toothbrushing is an essential part of good oral hygiene. This study investigated the use of stimulus fading without escape extinction to increase compliance with toothbrushing with three children with autism spectrum disorder (ASD). A 30-step stimulus fading hierarchy was implemented; gradually increasing the proximity of the toothbrush to the child's mouth, the duration of exposure to the toothbrush, and finally the duration of toothbrushing. Results demonstrated increased compliance with clinician implemented toothbrushing and generalization to caregivers for all participants. Systematic probes, conducted throughout the intervention, eliminated approximately 50% of the stimulus fading steps for each participant, with the specific steps omitted varying across participants. This study extends the generality of stimulus fading without escape extinction to oral hygiene practices for children with ASD.

1. Introduction

In 2007 the Centers for Disease Control reported that 42% of children aged 2–11 years old have had a cavity in their primary teeth and 21% of children aged 6–11 years old have had a cavity in their permanent teeth (Dye et al., 2007). Daily oral hygiene practices, including toothbrushing with fluoride toothpaste and flossing, reduces the risk of developing cavities and periodontal diseases (Alaluusua & Malmivirta, 1994; USDHHS, 2000). Furthermore, researchers suggest that children with cavities in their primary teeth are at increased risk for developing cavities in their permanent teeth (Helfenstein, Steiner, & Marthaler, 1991; Li & Wang, 2002; Steiner, Helfenstein, & Marthaler, 1992); making it essential for parents to establish a regular oral hygiene schedule for their children at home.

Current research on toothbrushing is restricted to teaching independent toothbrushing skills (Abramson & Wunderlich, 1972; Bouter & Smeets, 1979; Horner & Keilitz, 1975; Poche, McCubbrey, & Munn, 1982; Snell, Lewis, & Houghton, 1989; Wolber, Carne, Collins-Montogomery, & Nelson, 1987) and maintenance of regular toothbrushing (Blount & Stokes, 1984; Blount, Baer, & Stokes, 1987; Iwata & Becksfort, 1981; Lattal, 1969; Swain, Allard, & Holborn, 1982), with no studies, to date, examining compliance with caregiver implemented toothbrushing at home. While independent toothbrushing is an important skill, oftentimes it remains necessary for caregivers to brush their child's teeth to maintain a high level of oral

* Corresponding author at: Center for Autism and Related Disorders, Inc., 1620 N. 48th Street, Phoenix, AZ 85008, United States. Tel.: +1 602 325 2485x106.
E-mail address: m.bishop@centerforautism.com (M.R. Bishop).
[1] Now at: Advanced Child Behavior Solutions, LLC, United States.
[2] Now at: FirstSteps for Kids, Inc., United States.

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hygiene. Compliance with regular oral hygiene may be difficult for parents of children with ASD due to problem behavior associated with routine dental care (Brickhouse, Farrington, Best, & Ellsworth, 2009; DeMattei, Cuvo, & Maurizio, 2007; Farmer & Aman, 2011; Lai, Milano, Roberts & Hooper, 2012). In a survey on unmet dental needs for children with ASD, Lai et al. (2012) found that more than half of the families indicated uncooperative behavior as a barrier to dental care for their child. Similarly, DeMattei et al. (2007) reported that about half of participants with ASD displayed uncooperative behavior during an oral assessment by a dental hygienist.

When problem behavior occurs in reaction to oral hygiene procedures (e.g., toothbrushing) dental professionals may elect to use more restrictive techniques, such as sedation or physical restraint (Adair, Waller, Schafer, & Rockman, 2004; Davila, 1990; Kamen & Skier, 1985); while caregivers responsible for maintaining daily oral hygiene are more likely to delay toothbrushing or avoid it all together. This practice allows the child to avoid the aversive task, even if only temporarily, and increases the likelihood of plaque accumulation and subsequent tooth decay. One method used to increase compliance with aversive tasks is stimulus fading, which involves gradually increasing exposure to the aversive stimulus by increasing the proximity of the stimulus, the intensity of the stimulus, or the duration of exposure to the stimulus (Cooper, Heron, & Heward, 2007; Miltenberger, 2008).

Stimulus fading has been used to increase compliance with blood tests (Shabani & Fisher, 2006), eating nonpreferred foods (Freeman & Piazza, 1998), swallowing medication (Ghuman, Cataldo, Beck, & Slifer, 2004; Schiff, Tarbox, Lanagan, & Farag, 2011), and medical and dental procedures (Birkan, Krantz, & McClannahan, 2011; Cuvo, Godard, Huckfeldt, & DeMattei, 2010; Cuvo, Reagan, Ackerlund, Huckfeldt, & Kelly, 2010). Researchers have successfully incorporated stimulus fading into treatment packages; the most common components being differential reinforcement, escape extinction, video modeling, and noncontingent reinforcement. The aforementioned procedures are relatively benign with the exception of escape extinction. When escape extinction is used to establish compliance with toothbrushing there is a risk of injury when attempting to place a toothbrush in the mouth of a child who is actively resisting. Two studies demonstrate that stimulus fading without escape extinction results in increased compliance (Schiff et al., 2011; Shabani & Fisher, 2006), suggesting that escape extinction may not always be a necessary component of treatment.

One consideration in stimulus fading is how much each stimulus fading step differs from the previous step. Generally, researchers describe stimulus fading hierarchies that consist of small steps toward a goal (Cuvo, Godard, et al., 2010; Schiff et al., 2011), rather than listing distinct skills for each step (Luscre & Center, 1996). For instance, Cuvo, Godard, et al. (2010) presented a stimulus fading hierarchy for compliance with the placement of a dental mirror in a person's mouth by initially decreasing the distance between the mirror and the individual, then increasing the amount of time the mirror touched the individual's lips, and finally, increasing the amount of time the mirror was in the individual's mouth. In contrast, the hierarchy of steps for a dental exam described by Luscre and Center included one step for compliance with the dental mirror, one step for compliance with the dental tool, and one step for compliance with the suction tool. Researchers report success with interventions using a variety of stimulus fading hierarchies. However, it may be possible to minimize problem behavior and eliminate the need for escape extinction by beginning at a point where the stimulus is not aversive and progressing through steps that have only slight variations from each other, making the change from one step to the next unnoticeable.

Another variable to consider is how rapidly to advance through a stimulus fading hierarchy. Some researchers gradually move through each step in the hierarchy (Birkan et al., 2011; Cuvo, Regan, et al., 2010); however, there needs to be a balance between slowly moving through the stimulus fading steps and ensuring that treatment is not needlessly extended by teaching unnecessary steps. Other researchers periodically probe the terminal goal during treatment (Freeman & Piazza, 1998; Shabani & Fisher, 2006) or use pre-instructional probes to determine the initial stimulus fading step (Ghuman et al., 2004), with few using systematic probes throughout treatment to inform decisions about which stimulus fading steps are required (Cuvo, Godard, et al., 2010; Schiff et al., 2011). Prior to beginning treatment it is impossible to determine if an individual will need to be exposed to all, or only some, of the steps in a stimulus fading hierarchy. A study by Schiff et al. (2011) found that systematic probes, conducted three steps ahead of the current mastered step, eliminated about 25% of the stimulus fading steps. The inclusion of systematic probes allows a clinician to make client specific decisions about which steps are necessary during treatment.

The purpose of the present study was to extend research on stimulus fading without escape extinction to compliance with clinician implemented toothbrushing with children with ASD. Generalization to caregiver implemented toothbrushing was assessed. Regular probes were performed to determine if any stimulus fading steps could be skipped throughout the intervention.

2. Method

2.1. Participants and setting

Three children with ASD who physically avoided caregiver implemented toothbrushing participated. John was 4 years old, and Todd and Holly were 5 years old. Sessions were conducted in the child's home in a bathroom where toothbrushing typically occurred (John and Holly) or in a room where the child received therapy services (Todd). A standard toothbrush, electric toothbrush, toothpaste, and preferred stimuli were present for each session. Stimulus fading was implemented using a standard toothbrush (John and Todd) and an electric toothbrush (Holly). Generalization to an electric toothbrush was probed for John and Todd. 1–12 sessions were conducted daily, 1–5 days per week depending on participant availability.

2.2. Experimental design and data analysis

A multiple baseline across participants design was used. The percentage of trials each participant complied with toothbrushing and the percentage of trials with physical avoidance were recorded. Compliance was defined as the participant tolerating the current stimulus fading step with no physical avoidance. Physical avoidance included turning head away from the toothbrush, pushing the toothbrush away, not opening mouth for the toothbrush, and leaving the toothbrushing area. Vocal protests were ignored and included saying "No," "I don't want to," and crying. Additional data were collected on the number of stimulus fading steps eliminated through probes and length of intervention.

2.3. Procedure

2.3.1. Baseline

During baseline sessions the clinician said "I am going to brush your teeth" and attempted to brush the child's teeth for 60 s. There were no programmed consequences for compliance. Physical avoidance resulted in termination of toothbrushing for 30 s and vocal protests were ignored. Each baseline session consisted of one trial.

2.3.2. Stimulus fading

A 30-step stimulus fading hierarchy was used to establish compliance with clinician implemented toothbrushing. See Table 1. Prior to each session, the child was asked to select one item from an array of three to five preferred foods, toys, or activities identified by their caregiver. The item selected was delivered contingent on compliance with the current stimulus fading step. Physical avoidance resulted in termination of the stimulus fading trial for 30 s and vocal protests were ignored. Each stimulus fading session consisted of three trials. Participants advanced to the next stimulus fading step following 100% compliance for one session. When a participant was unable to achieve a minimum of 66% compliance for two consecutive sessions, training on the current step ceased and training on the previous step was repeated (Todd).

The stimulus fading hierarchy began with increasing the proximity of the toothbrush, moving from 61 cm away to next to the child's mouth, while also increasing the duration of exposure, starting with a brief flash and progressing to 5 s (steps 1–9). For these steps each trial began with the instruction "I am going to show you the toothbrush, but I am not going to brush yet." The instruction "Open your mouth" was added for step 9 because this step required the participant to tolerate the toothbrush next to his or her open mouth. The next four steps required the child to tolerate the toothbrush in his or her mouth for gradually longer durations, beginning with 0.5 s and increasing to 5 s (steps 10–13). The instruction for these steps was "I am

Table 1
Stimulus fading hierarchy for toothbrushing.

Steps	Description
1	**Flash toothbrush 61 cm away**
2	Show toothbrush for 1 s, 61 cm away
3	Show toothbrush for 3 s, 61 cm away
4	**Show toothbrush for 5 s, 61 cm away**
5	Show toothbrush for 5 s, 15 cm away
6	Show toothbrush for 5 s, 8 cm away
7*	**Show toothbrush for 5 s, 3 cm away**
8	Show toothbrush for 5 s, next to mouth
9	Show toothbrush for 5 s, next to mouth with mouth open
10*	**Put toothbrush in mouth for 0.5 s**
11	Put toothbrush in mouth for 1 s
12	Put toothbrush in mouth for 3 s
13	**Put toothbrush in mouth for 5 s**
14	Brush teeth for 1 s, counting aloud for each second
15	Brush teeth for 3 s, counting aloud for each second
16*	**Brush teeth for 5 s, counting aloud for each second**
17	Brush teeth for 7 s, counting aloud for each second
18	Brush teeth for 10 s, counting aloud for each second
19*	**Brush teeth for 12 s, counting aloud for each second**
20	Brush teeth for 15 s, counting aloud for each second
21	Brush teeth for 20 s, counting aloud for each second
22	**Brush teeth for 25 s, counting aloud for each second**
23	Brush teeth for 30 s, counting aloud for each second
24	Brush teeth for 35 s, counting aloud for each second
25*	**Brush teeth for 40 s, counting aloud for each second**
26	Brush teeth for 45 s, counting aloud for each second
27	Brush teeth for 50 s, counting aloud for each second
28*	**Brush teeth for 55 s, counting aloud for each second**
29	Brush teeth for 60 s, counting aloud for each second
30	**Brush teeth for 60 s**

Note: Steps in bold text indicate when intermediate probes were conducted. An asterisk indicates when terminal goal probes may be performed.

going to put the toothbrush in your mouth, but I am not going to brush yet. Open your mouth.'' The final 17 steps focused on toothbrushing for progressively longer durations, starting with brushing teeth for 1 s while counting aloud for each second and ending with the terminal goal of brushing teeth for 60 s (steps 14–30). These trials began with the instruction ''I am going to brush your teeth. Open your mouth.''

2.3.3. Probes

Four types of probes were conducted: caregiver probes, electric toothbrush probes, intermediate probes, and terminal goal probes. Each probe consisted of one trial; physical avoidance resulted in the termination of the probe for 30 s, and vocal protests were ignored. There were no programmed consequences for compliance during terminal goal, electric toothbrush, and caregiver probes; however, social praise was delivered contingent on compliance with intermediate probes. Caregiver and electric toothbrush probes were conducted during baseline and following mastery of the terminal goal to assess generalization.

Intermediate probes were performed following mastery of every third stimulus fading step (i.e., steps 1, 4, 7, 10, 13, 16, 19, 22, 25, 28, and 30) throughout treatment to determine which steps to target. Note that there are only two steps between step 28 and step 30 because step 30 is the terminal goal. The step subsequent to the last successful intermediate probe was targeted for treatment. That is, if a child passed the intermediate probe for step 7 and failed the intermediate probe for step 10, treatment began at stimulus fading step 8; then, intermediate probes resumed after the child mastered step 10.

Terminal goal probes were performed following a mastered intermediate probe at steps 7, 16, and 25. If a participant failed the intermediate probe for steps 7, 16, or 25 and mastered the subsequent intermediate probe for step 10, 19, or 28, a terminal goal probe was conducted. If the participant failed the intermediate probe for step 10, 19, or 28 no terminal goal probe was conducted. For example, if a participant failed the intermediate probe for step 7 and following stimulus fading passed the intermediate probe for step 10, a terminal goal probe was conducted. Alternatively, if the participant failed the intermediate probe for steps 7 and following stimulus fading failed the intermediate probe for step 10, a terminal goal probe was not conducted. An exception is noted for John, where terminal goal probes were not conducted following mastered intermediate probes for steps 19 and 25; instead, they were conducted following mastery of the intermediate probe for step 22 and mastery of stimulus fading step 28. Failure on a terminal goal probe resulted in the continuation of intermediate probes until the next teaching step was identified. For example, if a child passed the intermediate probe for step 16 and failed the subsequent terminal goal probe, an intermediate probe for step 19 would be conducted.

2.4. Interobserver agreement

Interobserver agreement (IOA) data were collected during 74% of probe sessions (including baseline, intermediate probes and terminal goal probes), 81% of stimulus fading sessions, and 71% of caregiver probe sessions. An agreement was scored if both observers recorded compliance with the probe or stimulus fading step, or if both observers recorded noncompliance with the probe or stimulus fading step. IOA was calculated by dividing the number of agreements by the total number of agreements plus disagreement and multiplying by 100%. IOA for probe sessions was 98% (range: 94–100%). IOA was 100% for stimulus fading sessions and caregiver probe sessions.

3. Results

Fig. 1 shows results for John, Todd, and Holly. During baseline for John and Todd, compliance with toothbrushing remained at 0% correct with standard and electric toothbrushes across clinician and caregiver probes. Physical avoidance occurred during 100% of sessions. Baseline data for Holly revealed an increase in compliance from 0% correct to 100% correct with a standard toothbrush during clinician and caregiver probes. When an electric toothbrush was introduced compliance decreased to 0% correct, with physical avoidance occurring during 100% of sessions. Stimulus fading was conducted with a standard toothbrush for John and Todd, and an electric toothbrush for Holly.

At the beginning of the stimulus fading phase, John, Todd, and Holly were exposed to intermediate probes to determine the initial stimulus fading step. All participants complied with probes for steps 1 and 4, with physical avoidance occurring during the probe for step 7. As a result, all participants began stimulus fading with step 5. For John, performance improved throughout stimulus fading with a standard toothbrush; 100% compliance and 0% physical avoidance occurred on the terminal goal probe following mastery of step 28. Compliance during a clinician implemented electric toothbrush generalization probe was observed. The inclusion of intermediate and terminal goal probes resulted in the omission of 13 potential stimulus fading steps, with compliance on 70% of intermediate probes and 57% of terminal goal probes. Compliance was consistently observed during the last four terminal goal probes. This intervention required a total of 33 sessions conducted over 10 days, excluding baseline.

During stimulus fading with a standard toothbrush, Todd initially displayed a moderate increase in compliance with physical avoidance observed during steps 5, 6, and 7. However, compliance increased to 100% correct and physical avoidance decreased to 0% on the intermediate probe for step 10 and subsequent terminal goal probe. Stimulus fading was temporarily suspended for 2 months due to illness. A terminal goal probe was conducted when sessions resumed and maintenance was not observed; physical avoidance tended to occur after about 30 s of toothbrushing. Intermediate probes were used to determine the next appropriate stimulus fading step; 100% compliance was observed for step 22 and physical avoidance

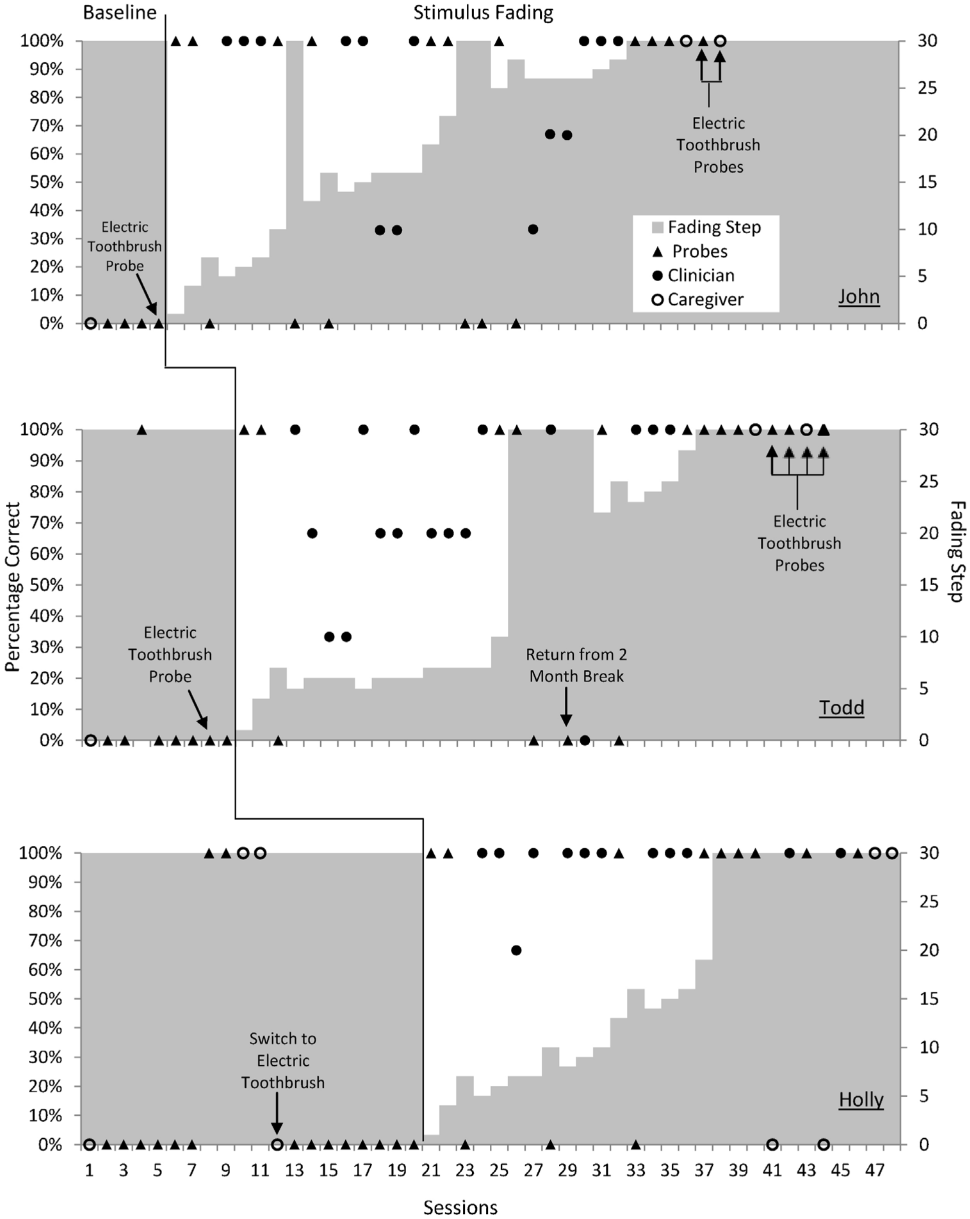

Fig. 1. Percentage of trials with correct responding during baseline, stimulus fading, and probe sessions for John (top panel), Todd (middle panel), and Holly (bottom panel). The step of the fading procedure being implemented during a session is depicted by the gray column (right *y* axis).

occurred for step 25; therefore, stimulus fading continued at step 23. Compliance improved, with Todd achieving 100% correct and 0% physical avoidance on the intermediate probe for step 28 and the subsequent terminal goal probe. Generalization to clinician implemented toothbrushing with an electric toothbrush was observed. A total of 16 potential stimulus fading steps were eliminated through the use of intermediate and terminal goal probes. Correct performance was scored on 71% of intermediate probes and 77% of terminal goal probes; with 100% compliance on the final six terminal goal probes. A total of 35 sessions, conducted across 23 days, were necessary to complete this intervention, excluding baseline and the break in treatment due to illness.

Holly exhibited an immediate increase in compliance and reduction in physical avoidance when stimulus fading with an electric toothbrush was initiated, with 100% compliance and 0% physical avoidance occurring on the intermediate probe for step 19 and the following terminal goal probe. The use of intermediate and terminal goal probes resulted in the removal of 16 potential stimulus fading steps; compliance was observed on 57% of intermediate probes and 100% of terminal goal probes. This intervention was completed in 27 sessions over 7 days, excluding baseline.

Generalization to caregiver implemented toothbrushing was observed for all participants. Results indicate 100% compliance and 0% physical avoidance on caregiver probes with standard and electric toothbrushes for John and Todd. Generalization was not immediately observed for Holly; with physical avoidance continuing to occur each time her caregiver attempted toothbrushing with an electric toothbrush. Two additional stimulus fading sessions at the terminal step (step 30) were necessary before generalization was observed.

4. Discussion

These findings extend existing research on stimulus fading by demonstrating an efficient method for establishing compliance with clinician implemented toothbrushing and generalization to caregivers. On average the intervention was completed in 32 sessions (range: 27–35 sessions) conducted over 13 days (range: 7–23 days). The inclusion of systematic probes eliminated about 50% of the potential stimulus fading steps for each participant; with the specific steps removed varying across participants. Furthermore, results indicate that stimulus fading combined with reinforced compliance was effective without escape extinction; which is consistent with data presented by Schiff et al. (2011) and Shabani and Fisher (2006).

Escape extinction continues to be a popular component of stimulus fading interventions (Birkan et al., 2011; Cuvo, Goddard, et al., 2010; Cuvo, Reagan, et al., 2010; Freeman & Piazza, 1998), with Kemp (2005) suggesting that it is a critical element when treating noncompliance with painful procedures. Even though escape extinction may be one part of a stimulus fading treatment package and is implemented only when necessary; there remains an increased risk of injury when attempting to complete medical and dental procedures with a child who is physically resisting. Additionally, parents often prefer not to use escape extinction (Kerwin, 1999). For these reasons, including increasing evidence that escape extinction may not always be necessary, it is reasonable to suggest that practitioners include escape extinction only when it is safe for the client and when reinforcement-based stimulus fading procedures have failed.

Results also support findings reported by Cuvo, Goddard, et al. (2010) and Schiff et al. (2011) by demonstrating that systematic probes avoided superfluous stimulus fading steps; reducing the chance of teaching unnecessary steps and potentially prolonging treatment. While some may argue that an alternative is to include fewer stimulus fading steps from the beginning (Luscre & Center, 1996), it is not possible to predict which steps to eliminate for a given individual. In the current study, systematic probes resulted in the removal of different teaching steps for each participant, suggesting that the stimulus fading steps needed to improve compliance are idiosyncratic. Therefore, a more conservative approach is to include enough steps such that advances from one step to the next are undetectable; relying on systematic probes to guide client specific decisions about which steps are necessary.

One limitation is that an extended baseline condition was not included for all participants. Holly displayed increased tolerance of a standard toothbrush within seven baseline sessions, with generalization to caregiver implemented toothbrushing. Similar increases may have been observed for other participants if they were exposed to more baseline sessions. The results for Holly are in contrast with findings presented by Cuvo, Godard, et al. (2010), which indicated minimal improvements in compliance with a dental assessment during an extended baseline condition. One possible explanation for the inconsistent results may be related to the general familiarity of the aversive task. In daily life, children are usually exposed to stimuli related to toothbrushing (e.g., toothbrush, toothpaste) more often than they are exposed to stimuli associated with a dental assessment (e.g., gloves, dental light, dental chair, dental mirror). Furthermore, participation in a dental assessment may be more aversive than toothbrushing, which could impact compliance during repeated exposure. Future research should examine these variables to determine the conditions under which repeated exposure to an aversive task will bring about sufficient improvement in compliance with that task.

Another limitation of this study is that two interventions were implemented simultaneously, stimulus fading and reinforced compliance; thus the relative contribution of each is unknown. Future research should evaluate the critical components of stimulus fading treatment packages; providing practitioners with vital information about how to best allocate resources during stimulus fading interventions. A third limitation is that data on plaque accumulation were not collected. There is an assumption that increased compliance with toothbrushing will result in decreased dental plaque. However, it is possible that caregivers were not adequately brushing all the teeth. Future research should investigate this, along with compliance with other oral hygiene tasks, such as flossing and use of a waterpik, that were not addressed in this study.

The stimulus fading procedure outlined in this investigation shares similar characteristics with dental desensitization methods discussed by Kemp (2005). Both techniques involve gradual exposure to dental stimuli in order to increase compliance; however, desensitization also incorporates relaxation strategies. While research suggests that both stimulus fading (Cuvo, Godard, et al, 2010; Cuvo, Regan, et al., 2010) and desensitization (Altabet, 2002; Conyers et al., 2004; Kemp, 2005; Luscre & Center, 1996; Neuman, Altabet, & Fleming, 2000) effectively increase compliance with dental procedures; it remains unclear if the addition of relaxation strategies expedites the learning process. Future research should examine the value of including relaxation strategies when implementing interventions to increase compliance with dental procedures.

Toothbrushing is an essential component of daily oral hygiene; with caregivers frequently responsible for maintaining the oral health of their children with ASD. Problem behavior associated with toothbrushing will impede parents' best efforts to establish a regular oral hygiene schedule. This study presents an effective procedure for increasing compliance with clinician implemented toothbrushing with generalization to caregivers for children with ASD. Compliance with caregiver implemented toothbrushing is the first step in a life-long commitment to oral hygiene. Future research should evaluate if compliance with caregiver implemented toothbrushing will promote the acquisition of independent toothbrushing skills.

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