Functional Communication Training in Rett Syndrome: A Preliminary Study

Breanne J. Byiers, Adele Dimian, and Frank J. Symons

Abstract
Rett syndrome (RTT) is associated with a range of serious neurodevelopmental consequences including severe communicative impairments. Currently, no evidence-based communication interventions exist for the population (Sigafoos et al., 2009). The purpose of the current study was to examine the effectiveness of functional assessment (FA) and functional communication training (FCT) methods for teaching 3 individuals (ages 15–47 years) with classic RTT novel communicative behaviors. Using single-case experimental designs, functional reinforcers were identified (FA) and each participant quickly learned to activate a voice-output switch to obtain a reinforcer (FCT). These results suggest that individuals with classic RTT can learn novel communicative responses, which has important implications for future intervention research.

Key Words: Rett syndrome; functional communication training; functional analysis

Rett syndrome (RTT) is a devastating neurodevelopmental disorder affecting almost exclusively females. It is caused, in most cases, by mutation or deletion of the MECP2 gene, which encodes the epigenetic regulator methyl-CpG binding protein 2 (MeCP2). Although our understanding of the full range of functions of the MeCP2 protein is still being established, it has been implicated in synapse development and maintenance, and is therefore essential for normal neurological function (e.g., Tao et al., 2009). Following apparently normal perinatal development, girls with RTT show a loss of communication and motor skills, and the development of stereotypical hand movements (Hagberg, Hanefeld, Percy, & Skjeldal, 2002). A number of health problems, including seizures, scoliosis, and apnea are common in this population (e.g., Williamson & Christodoulou, 2006).

Due to the pervasive nature of the effects of the syndrome, girls and women with RTT typically require life-long assistance in all areas of their lives. The disorder results in a complete loss of expressive language for most affected individuals, aside from those with a milder preserved speech variant (Zapella, Gillberg, & Ehlers, 1998). As a result, most individuals with RTT have no formal recognized method of communication (von Tetzchner, 1997), although parents and other caregivers often report the use of unconventional gestures, facial expressions, and eye gaze to communicate (e.g., Lavas, Slotte, Jochym-Nygren, van Doorn, & Engerstrom, 2006). Currenty, the degree to which individuals with RTT retain sufficient motor control to allow the reliable production of nonverbal communicative cues is unclear. To date, two studies (Sullivan, Laverick, & Lewis, 1995; Watson, Umansky, Marcy, & Repacholi, 1996) reported the results of interventions using nonsocial environmental contingencies (e.g., toy activation) as reinforcers for motor behaviors (e.g., arm reaching and leg kicking) for single participants. Although both studies reported increases in motor behavior, neither study used an internally valid experimental design. More recently, Lancioni and colleagues (2014) built on these previous studies by demonstrating that a young woman with RTT showed an increase in the frequency of micro-switch activations when preferred videos were provided.
Evidence of communicative behaviors, both nonverbal and verbal, has been observed among individuals with RTT (combined N = 41). Evidence of communicative behaviors was found for some participants in each of the studies, but the variability in methods made it impossible to draw firm conclusions regarding the range of communicative behaviors and abilities in this population. The primary issue in interpreting the results of the studies is the nature of the potential communicative acts among individuals with RTT. Because most of the participants engaged in behaviors that might be communicative, but might also be interpreted as more basic physiological responses (e.g., changes in breathing, body posture), descriptive changes in the rate of the behaviors are not conclusive evidence of communicative acts. Studies with procedures that experimentally assess the sensitivity of the behaviors to environmental consequences would provide a more conclusive answer to the question, but have not yet been conducted.

Overall, the existing studies suggest that individuals with RTT engage in a variety of nonverbal behaviors that appear to be sensitive to environmental conditions and that caregivers tend to interpret as communicative. Reducing the ambiguity surrounding idiosyncratic behavior displayed by individuals with RTT and determining whether it may function or have the potential to function as a communicative act is a logical, important, and needed next step. One approach to doing so may be to rely on well-established behavioral assessment technology (i.e., functional analysis [FA]; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994) and functional communication training (FCT; Carr & Durand, 1985).

There is a strong body of literature focused on FA-FCT as a strategy for reducing problem behaviors (e.g., Durand, 1990). A newer, but growing body of work is supporting the techniques used in FCT as a potential method for improving the interpretability of prelinguistic behaviors among children and adults with severe disabilities (Keen, Sigafoos, & Woodyatt, 2001). Although the concepts of function-based communication interventions have been suggested by researchers for individuals with RTT dating back to the late 1990s (e.g., von Tetzchner, 1997), they have yet to be tested in any empirical studies.

In a second systematic review paper, Sigafoos and colleagues (2009) reported that of nine communication intervention studies with individuals with RTT, only one study (Van Acker & Grant, 1995) was sufficiently well designed to provide any conclusive evidence. In the study by Van Acker and Grant, two of the three participants showed clear increases in responding (touching a computer screen to request food) as a function of the computer-based intervention. Among the other studies, six lacked a recognizable experimental or quasi-experimental research design, and two included single-subject designs that
were compromised by a lack of replication across participants (Sigafoos, Laurie, & Pennell, 1996), or improvements during baseline among several participants (Hetzroni, Rubin, & Konkol, 2002). The studies were a mix of various types of interventions, including operant conditioning paradigms, music and sensory therapies, and other approaches. None of the studies, however, incorporated the participants’ existing potential communicative acts or used function-based methods. Despite the variability in the strategies and research methods, positive results in communication were reported for a majority of the participants involved suggesting, at least tentatively, that individuals with RTT can learn to communicate. Nevertheless, a literature search reveals that no further communication intervention studies in RTT have been reported since the publication of the review paper in 2009.

Collectively, the literature reviews conducted by Sigafoos and colleagues (2009; 2011) reveal two important gaps in the RTT communication literature: (a) studies confirming the communicative functions of potentially communicative behaviors, and (b) studies employing recognized research designs and reliable measurement of dependent variables to generate experimental evidence of positive responses to communication interventions. The purpose of the current preliminary study was to begin addressing both of these gaps by assessing the capacity of a small clinical sample of individuals with RTT to learn and produce novel, functionally communicative behaviors evaluated using well-established FA-FCT procedures and methodologically rigorous, single-subject experimental designs.

Methods

Participants and Setting
Following Institutional Review Board approval, three individuals with RTT and their families were recruited for the study through a statewide RTT parent group in collaboration with a regional RTT Clinic. All three participants had clinical diagnoses of classic RTT and were dependent on others for all aspects of their daily lives. All analyses took place in quiet locations within the participants’ primary residences.

Jen was 15 years old, could walk with some assistance, and had minimal hand function, although she could eat some finger foods independently. She was taking several medications at the time of the study including anti-convulsants and melatonin. She had no functional communication skills and her receptive language skills were unclear. She had some experience using voice output switches for communication at home and at school, but no information on the degree to which she used the switches independently was available, and she was not actively using any kind of augmentative communication system at the time of the study. Jen had an identified MECP2 mutation.

Tammy was 27 years old and was nonambulatory. She could hold and drink from a lidded cup independently, but had no other functional hand use. She was on several medications at the time of the study, including anti-convulsants and prescription medications for sleep. She was nonverbal and did not use any augmentative communication system, although her caregivers reported that she had used voice-output switches for communication in the past. No information on Tammy’s language or cognition functioning was available. Although Tammy had a clinical diagnosis of classic RTT, no genetic testing had been conducted to confirm the presence of a MECP2 mutation.

Rose was 47 years old and could walk unassisted. She could feed herself independently with a spoon with some spilling. She was not taking any medications as the time of the study. She was nonverbal and had no experience using any augmentative communication systems. No standardized assessments of language or cognitive functioning were available. Rose had a clinical diagnosis of classic RTT, but no genetic testing had been conducted.

Functional Assessment
The functional assessments included semistructured interviews, unstructured observations, and analog functional analyses. Interviews were conducted with the participants’ primary caregivers (Jen’s mother, Tammy’s grandmother, and the manager of Rose’s group home). Interview questions were taken from the Functional Assessment Interview (FAI; O’Neill et al., 1997), and the Inventory of Potential Communicative Acts (IPCA; Sigafoos et al., 2006). After describing the purpose of the interview, the researchers asked the caregivers to identify nonverbal behaviors used by the participants that were believed to be potentially communicative. If the caregivers were
unable to list any potentially communicative behaviors, the interviewers prompted them to consider idiosyncratic behaviors such as eye gaze, vocalizations, and changes in facial expression. For each behavior identified, caregivers were asked when, where, with whom, and during which activities the behavior was most likely to occur. They were also asked about preferred objects and leisure activities, and nonpreferred activities or tasks to be used in the functional analysis conditions.

Following the interview, the research team conducted at least 1 hour of unstructured observations of each participant during her typical daily routine to confirm the occurrence of the potentially communicative behaviors identified in the interview. During the observations, trained observers recorded all instances of potentially communicative behaviors, including those identified by Sigafoos et al., (2000) as well as challenging behaviors including aggressive, destructive, or self-injurious behaviors. The potentially communicative behavior that occurred most frequently during the observation was selected as the target behavior for the subsequent analyses.

Experimental FA were conducted with each participant. All FA sessions were 5 min in length and were presented in an adapted multi-element format with the conditions with the highest and lowest rates of behavior condition repeated at least twice. All FA sessions were conducted in a single visit for each of the participants. Rate per minute of the target behavior was measured using an event count by one of the researchers present during the sessions. Interobserver agreement (IOA) was calculated for a minimum of 25% of sessions (randomly selected from all sessions) for each participant by an independent observer using video recordings of the sessions. IOA for Jen’s sessions ranged from 83% to 100% (M = 94%), Tammy’s sessions ranged from 83% to 100% (M = 94%), and Rose’s sessions ranged from 84% to 100% (M = 92.7%).

**Conditions.** Free play, escape, and attention conditions based on the procedures described by Iwata et al. (1982/1994) were conducted with each participant. Additionally, based on information from the caregiver interview, tangible conditions, designed to test the effects of access to preferred television shows (Jen), food (Tammy), and head and shoulder massage (Rose) were also conducted.

The free play condition was designed as a control condition, during which it was expected that the participants would engage in few, if any communicative behaviors because preferred activities and attention were provided noncontingently. During these sessions, preferred activities were available and positive social attention (comments, praise) was provided every 10–15 s, regardless of the participant’s behavior. There were no programmed consequences for any behaviors.

In the escape condition, the participant was prompted to engage in an activity of daily living (folding laundry for Jen, activating augmentative devices for Tammy, and vacuuming for Rose), using physical prompts to complete the activity. For Rose, selection of the activity was based on caregiver report that it was nonpreferred. Tammy’s and Jen’s caregivers, however, were able to identify specific activities that were believed to be non-preferred, as the participants were rarely, if ever, required to engage in chores or academic activities in the home setting, and caregivers reported that no activities of daily living could be completed independently. Therefore, activities were selected for which materials were available and physical prompting was possible. Contingent on the occurrence of the target behavior, the participant was told “It’s ok, you can take a break,” and all materials and people were removed from the area for 10 s.

In the attention condition, the participant was provided with preferred objects or activities. After 10 s of interaction, the researcher told the participant “I need to do some work now. I’ll be back in a few minutes.” The researcher moved at least 5 feet away from the participant and avoided eye contact or verbal interactions. Contingent on the occurrence of the target behavior, the researcher provided 10 s of verbal (e.g., “It’s ok,” or another comment typically used by the caregivers) and physical attention (e.g., a pat on the back or arm).

The tangible conditions varied across the three participants, depending on the common consequences the caregivers reported providing for the target behaviors. For Jen the consequence provided for the target behavior was 10 s of access to a preferred television show, for Tammy the consequence was a bite of preferred food, and for Rose the consequence was 10 s of head and shoulder massage. For all of the participants, the session began with a brief period of access to the reinforcer, after which the researcher said that the reinforcer was no longer available and withdrew it but continued providing verbal
interaction every 10–15 s. Contingent on the occurrence of the target behavior, a brief period of access was provided.

Functional Communication Training

Switch placement and training. Before the experimental sessions began, assessment sessions were conducted to identify an appropriate alternative communicative response for each participant. This process involved placing the switch and physically prompting the participant to press it with the appropriate body part. All prompted switch-presses were followed by praise and a brief period of access to a preferred food or activity. A three-step most-to-least prompting hierarchy (e.g., Wolery, Ault, & Doyle, 1992) was used to promote independent switch-pressing. If the researcher was unable to reduce the level of physical prompting used after five consecutive trials, a new switch type or location was tried. Once the participant demonstrated at least one independent switch activation using a specific switch type and location, a formal training session was implemented. At the beginning the training session, the participant was told to press the switch to obtain the reinforcer. Independent switch presses resulted in praise and a brief period of access to the functional reinforcer identified during the functional assessment. If the participant did not independently activate the switch within one minute, the minimum level of physical prompt necessary was used. All prompted trials were reinforced. Training was considered complete when the participant produced the response independently within 1 min on five consecutive opportunities.

For Jen and Rose, the FCT sessions took place with the participants sitting on the couch in the living rooms of their homes; the switches were placed on the couch beside the participant next to her dominant hand. For Tammy, the FCT sessions took place with her lying on a blanket on the living room floor and the switch was placed on the floor beside her dominant hand.

Experimental design. Once appropriate switch placement had been identified, 5 min FCT sessions were conducted in A-B-A-B reversal designs with each participant. In the A, or baseline sessions, the occurrence of the participant’s target behavior resulted in 15 s of reinforcement (according to the function identified during the FA) and switch activations were ignored. In the B, or intervention sessions, the contingencies were reversed such that switch activations resulted in reinforcement and target behaviors were ignored. At the beginning of the first session of each new phase, the contingencies were explained verbally to the participant (e.g., “If you want to watch TV, you need to tell me with the switch”). Between four and 10 FCT sessions were conducted per visit, depending on the participant’s schedule. The number of visits required to complete all of the FCT sessions ranged between two (Rose), and three (Jen and Tammy).

Data analysis. Frequency counts for the target behaviors and independent switch activations were converted to rate per minute and a line graph was created for each participant. The results were analyzed using visual inspection and Non-Overlap of All Pairs (NAP; Parker & Vannest, 2009), a quantitative measure of the degree of change between conditions for each dependent. NAP was designed as an indicator of performance differences between conditions or phases, as indexed by data overlap. NAP scores range from 0.0 to 1.0. According to Parker and Vannest (2009), a NAP score can be interpreted as “the probability that a score drawn at random from a treatment phase will exceed (overlap) that of a score drawn at random from a baseline phase” (p. 359), or more simply, the proportion of non-overlapping data between the two phases. Therefore, a score of 1.0 would indicate no data overlap between phases, a score of 0.5 would indicate half of the points from phase A exceed those in phase B, and a score less than .5 would indicate deterioration in performance with the intervention.

Interobserver agreement. The frequency of both the target behaviors and switch activations was recorded by the primary observer during all sessions. Interobserver agreement was calculated for a randomly selected subset of at least 25% of the FCT sessions in each phase for each participant. Agreement ranged from 87% to 100% for Jen’s sessions (M = 98%), 80% to 100% for Tammy’s sessions (M = 96%), and 92% to 100% for Rose’s sessions (M = 96%).

Results

Functional Assessment

The results of all of the participants’ functional analyses are presented in Figure 1. For all participants, the target behavior identified via
interviews and observations demonstrated clear sensitivity to environmental conditions.

Jen. Jen’s mother reported that whining was Jen’s most frequently used form of communication. It was reported that she whined primarily when she was bored, especially when the TV was turned off, or turned to a nonpreferred program. This was supported by the results of Jen’s FA, in which whining occurred exclusively in the tangible (TV) condition.

Tammy. Tammy’s grandmother reported that she frequently engaged in self-injurious behaviors (SIB), including hitting herself in the abdomen and side of the head with a closed fist, and scratching her arms and legs with her fingernails. Although Tammy’s grandmother reported that she did not believe that SIB served any social functions, these behaviors were selected as the target behaviors because Tammy’s grandmother considered them to be problematic. The results of the FA indicate that Tammy’s SIB was sensitive to reinforcement in the form of access to attention, as the behavior occurred exclusively in the attention condition.

Rose. The manager of Rose’s group home reported that she would frequently grab the arms or hands of other people in the environment and she believed that this behavior was Rose’s way of requesting head and shoulder massages, which were reported to be highly preferred. This hypothesis was supported by the results of Rose’s FA because, although the behavior occurred occasionally in all of the sessions with the exception of the free play condition, it occurred at the highest rates in the tangible (massage) conditions, during which the behavior occurred over four times per minute.

Functional Communication Training

Although the participants showed some variability, all three participants demonstrated their ability to vary their communicative behaviors between switch-presses and vocalizations according to the changes in the reinforcement contingencies (see Figure 2).

Jen. Overall, there was clear evidence of experimental control over the rates of Jen’s whining, as there was very little overlap between the experimental conditions, and there were clear and immediate changes in level after phase changes (NAP = .96). Changes in Jen’s rates of switch pressing, were more delayed in phases 2 and 3 and as a result there was more overlap between the phases (NAP = .74). In the final phase, however, a clear and immediate increase in switch pressing was observed.

Tammy. Overall, evidence for experimental control over Tammy’s rates of switch activation was very strong, as there was little overlap between the phases and clear and immediate changes in level occurred following every phase change (NAP = .92). Although there were not always immediate changes in the levels of Tammy’s SIB following phase changes, there were clear trends in the expected directions in phases 2 and 3, and there was an immediate decrease in the final phase (NAP = .64).

Rose. Rose had clear and immediate changes in her rates of both grabbing and switch activations following phase changes, and as a result, there was no overlap between experimental conditions. In addition, rates of switch pressing reduced to near-zero levels when reinforcement was provided for grabbing only, although
grabbing remained at moderate levels throughout the B phases (NAP = 1.0 for both behaviors).

**Discussion**

The purpose of this preliminary study was twofold: (a) to determine whether the potentially communicative behaviors exhibited by three individuals with RTT were sensitive to reinforcement contingencies, and (b) to examine the capacity of the participants to activate voice-output switches and in order to access functional reinforcers. Initial interviews with the caregivers resulted in the identification of a potentially communicative behavior for each participant. Subsequently, specific reinforcers were identified for each participant’s target behavior (i.e., a putative communicative function was established). That these behaviors were functional was further supported by the results of the FCT intervention, which showed that all
three participants were less likely to produce the target behavior in the second and fourth phases, when reinforcement was available only for activating the voice-output switch. Based on these results it seems likely that, for these participants, the identified target behaviors represented nonconventional, but functional methods of communication.

These results are promising, as they suggest that individuals with RTT are capable of learning to communicate with their caregivers. Nevertheless, based on these results alone, it would remain unclear whether this process requires months or even years of learning to establish. During the FCT sessions, however, all three participants quickly learned (during a single session) to activate the voice-output switch to obtain the reinforcer. This observation has important implications for the development of efficient and effective communication interventions for individuals with RTT in real-world contexts including home and school.

Our results are consistent with the very few previous studies demonstrating that some behaviors exhibited by individuals with RTT can be increased through the use of operant conditioning, with some evidence of “intentionality” in these behaviors (e.g., Piazza, Anderson, & Fisher, 2008; Watson et al., 1996). Evidence regarding intentional communicative behaviors, however, has been harder to obtain. Although several studies have demonstrated that the idiosyncratic behaviors of individuals with RTT vary in frequency and intensity in different social contexts (e.g., Oliver, Murphy, Crayton, & Corbett, 1993; Sigafoos et al., 2000), the research designs used in these studies did not allow for the demonstration of intentionality. More generally, the relation between the terms and concepts of “intentional communication” as used in the developmental psychology and language development literature and “functional communication” as used in the applied behavioral psychology literature is not clear. Evidence for one may not necessarily be evidence for the other.

Many developmental psychologists draw a distinction between intentional behavior, in which an individual produces a direct outcome from the environment, and intentional communication, which requires that the individual understand the role of the listener in a communicative exchange (e.g., Bretherton & Bates, 1979). Previous studies have suggested that individuals with RTT may not reach the developmental level necessary for intentional communication (Woodyatt & Ozanne, 1994). Although the current study was not specifically designed to assess intentional communication, the results provide some preliminary evidence that individuals with RTT may be capable of developing intentional communication, as the ability to vary communicative signals when the message is not understood is one indicator of intentional communication (e.g., Wetherby & Prizant, 1989). Clearly, further work explicitly designed to test this idea would be necessary. Regardless of whether the behaviors exhibited by the participants are better described as intentional behaviors or intentional communication, these results have important clinical and educational implications for individuals with RTT. Given the speed and ease with which the participants acquired the new responses, the results suggests that more effort should be made in providing individuals with RTT appropriate ways of affecting their environment.

Several limitations of the current study should be noted. First, the participants represent three consecutive cases, and likely do not represent the full range of functioning among individuals with RTT. Future studies should include larger samples with individuals of different ages and physical functioning. Additionally, although all three participants met the clinical criteria for classic RTT, no genetic confirmation of MECP2 mutations was available for the two older participants. Therefore it is possible that those two participants had different genetic mutations from those with confirmed genetic mutations (although RTT is a behaviorally diagnosed disorder [i.e., one can be diagnosed with RTT but fail to find a definitive MECP2 mutation]). Second, the current study was designed as a preliminary proof-of-concept study only. The duration of the intervention was very brief (under 6 hours with each participant), and some phases included only 3 or 4 data points, rather than the recommended 5 (e.g., Kratochwill et al., 2010). Additionally, there was no planned follow up with the families to determine whether there was continued use of the switches, so no information about the maintenance and generalization of FCT procedures for communication intervention in the sample was available. Third, only one communicative function was selected for each participant although each participant likely had a number of different communicative behaviors and functions. Future studies should
further explore the range of communicative behaviors and functions in this population and examine the degree to which multiple discriminable responses can be taught. Finally, the relative response effort associated with the two behavioral options (e.g., target behavior vs. switch activation) was not considered directly in the current study. Future studies should ensure that the responses being taught require less effort than the responses being replaced to facilitate generalization and maintenance.

Given the paucity of evidence regarding learning and communication in RTT, studies that further examine the capacity of individuals with RTT to learn novel skills are critical to the quality of life of affected individuals and their families. This study provides preliminary evidence that at least some individuals with RTT use idiosyncratic behaviors to communicate with their caregivers, and that they can quickly learn to switch their communicative signals in response to change in environmental contingencies.

References


Received 9/26/2012, accepted 11/17/2013.

Preparation of this report was supported, in part, by NICHD Grants No. 44763 & 47201 to the University of Minnesota, the Minnesota Medical Foundation, and the University of Minnesota Dissertation Fellowship.
Our sincere thanks to the Minnesota Rett Syndrome Family Research Association - a committed group of parents - and their children for their support.

Authors:
Breanne Byiers (e-mail: byier001@umn.edu), University of Minnesota, Department of Educational Psychology; Adele Dimian, University of Minnesota, Department of Educational Psychology; and Frank J. Symons, University of Minnesota, Department of Educational Psychology.

Correspondence concerning this article should be addressed to Breanne Byiers, Department of Educational Psychology, Education Science Building, 56 River Road, University of Minnesota, Minneapolis, MN, 55455 (e-mail: byier001@umn.edu).