Functional Communication Training in Rett Syndrome

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Dedication

This dissertation is dedicated to all of the families of children and adults with Rett syndrome who have supported and participated in our research projects.

Abstract

Rett syndrome (RTT) is associated with a range of serious neurodevelopmental consequences including severe impairments in communication. Currently, no evidence-based communication interventions exist for the population (Sigafoos et al., 2009), and there is limited empirical evidence that individuals with RTT are able to demonstrate operant motor behaviors, (e.g., behaviors that are controlled by the individual in order to create and effect on the environment). The purpose of the current study was to examine the effectiveness of functional assessment (FA) and functional communication training (FCT) methods for teaching a clinical case series of girls and women (aged 4-47 years) with classic RTT to request preferred events or items using an augmentative communication device. Functional analysis (FA) was used to identify the communicative function of potential communicative acts (PCAs) identified during parent interviews and observations. Subsequently, each participant was taught to activate a voice-output switch to request the functional reinforcer identified in the FA. Using ABA and ABAB single case experimental designs, the degree to which each participant alternated between the PCA and switch activation according to changes in the environmental consequences (e.g., reinforcement vs. extinction for a particular response) was examined. Clear experimental effects of the intervention condition were observed on at least one response for six of the seven participants. The remaining participant did not complete the study. Overall, these results suggest that individuals with RTT can use intentional motor behaviors, and are responsive to environmental consequences. These results have important implications for the development of appropriate communication interventions for this population.

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Chapter 1: Introduction

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 spoken language for the most affected individuals. A minority of individuals diagnosed with RTT experience a milder preserved speech variant (Zapella, Gillberg, & Ehlers, 1998). Most individuals with RTT are not taught to use graphic or gestural mode communicative methods (e.g.,sign language, picture exchange systems, and voice-output devices) presumably because of severe manual motor deficits and continuous hand stereotypies. As a result, most individuals with RTT have no systematic communication system (von Tetzchner, 1997).

Very little empirical work has been implemented to identify the communicative functions of ambiguous, potentially communicative acts in this population. Consequently, there has been limited empirically based guidance on the identification of or implementation of strategies to teach communication targets for this population. One reason for the limited literature on communication in RTT may be that assessment of receptive and expressive language abilities has proven difficult because individuals with RTT often cannot produce the motor or language responses required for the administration of standardized assessments, and they may require extended response times (Demeter, 2000). To get around these issues, studies have been conducted that utilized structured or naturalistic observations of interactions between affected individuals and their caregivers to identify possible communicative behaviors.

Woodyatt and Ozanne (1992; 1993; 1994; 1997) used unstructured observations of small samples of individuals with RTT in which all of the participants reportedly engaged in idiosyncratic behaviors (e.g., changes in facial expression, vocalizations, hyperventilation) that appeared to serve communicative functions. These investigators concluded, however, that almost all of the participants functioned at a 'pre-intentional' level of communication. Similarly, Sigafoos, Woodyatt, Tucker, Roberts-Pennel, and Pittendreigh (2000) reported that, in their sample of three individuals with RTT, certain motor behaviors (e.g., eye gaze, facial expressions), represented potential communicative acts (PCAs), in that they were differentially sensitive to environmental conditions. The authors reported that, although caregivers and teachers interpreted the behaviors as communicative, it was unclear from the available information whether the behaviors were intentional communicative bids, or simply expressions of physiological arousal in response to environmental change (autonomic instability is a comorbid characteristic of the syndrome).

Sigafoos et al. (2011) subsequently reviewed the eight published studies that had explicitly examined communicative behaviors with individuals with RTT (combined N = 41). Evidence of communicative behaviors were 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 across the samples. The primary issue in interpreting study results was 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), changes in the rate of the behaviors were 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 idiosyncratic behaviors that appear to be sensitive to environmental conditions and that caregivers tend to interpret as communicative. Reducing the ambiguity surrounding idiosyncratic behavior(s) displayed by individuals with RTT and determining whether it functions or has the potential to function as a communicative act is a logical, important, and a needed next step. One approach leading to this determination involves relying on well-established behavioral assessment technology (i.e. functional analysis [FA]) and functional communication training (FCT).

There is a strong body of literature focused on FA-FCT as a strategy for reducing problem behaviors (e.g., Betz, Fisher, Roane, Mintz, & Owen, 2013; Derby et al., 1997; Dunlap, Ester, Langhan, & Fox, 2006; Durand, 1990; Harding, Wacker, Berg, Lee, & Dolezal, 2009). 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 (e.g., Keen, Sigafoos, & Woodyatt, 2001; Drasgow, Halle, Ostrosky, & Harbers, 1996). 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 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) met the criteria for conclusive evidence established by the authors of the review. In this study, 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 incorporated the participants' existing potential communicative acts, or used function-based methods. Despite the variability in the intervention strategies and research designs employed, positive communication outcomes were reported for a majority of the participants , providing tentative support that individuals with RTT can learn to communicate. Nevertheless, little research has been conducted since the review paper published in 2009. In fact, a systematic search of several psychology- and medicine-related databases using the search terms "rett syndrome", and "communication" revealed only several descriptive studies examining patterns of communication among individuals with RTT, but only one intervention study. This study (Bartolotta, & Remshifski, 2012) reports the effects of coaching communication partners on the communicative responses of four individuals with RTT. Although the authors reported an increase in communication bids for the participants, the case-study approach used, as well as the heterogeneity in intervention procedures across participants, mean that no clear conclusions can be drawn from this study.

The literature reviews conducted by Sigafoos and colleagues (2009; 2011) revealed two important gaps in the RTT communication literature that include an absence of studies: (a) confirming the communicative functions of potential communicative acts (PCAs), and (b) employing recognized research designs and reliable measurement of dependent variables to generate experimental evidence of positive responses to communication interventions. This investigation will address both of these gaps by assessing the capacity of a 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.

The specific research questions addressed in the current study are:

1. Do the participants with RTT increase the frequency of their PCAs in response to one of more types of positive and/or negative reinforcement contingencies (as compared to the control condition) within the context of experimental functional analysis (FA) sessions?
2. Will the participants learn to produce a novel motor response (activating a voice-output switch) to obtain the functional reinforcer associated with the highest rate of responding during the FA sessions?
3. Will the participants' rates of the two available responses (e.g., PCA and switch-activation) vary according to changes in the environmental contingencies (e.g., reinforcement vs. extinction)?

Chapter 2: Review of the literature

Rett Syndrome (RTT)

As advances in scientific technology lead to improved understanding of the genetic bases for a number of different syndromes, research into the behavioral phenotypes - profiles of behaviors, and strengths and weaknesses in functioning particular to a disorder - has increased (Dykens & Hodapp, 2001). Such research can have important impacts, including insights into the effects of genetic and molecular deviations on behavior and cognitive and adaptive functioning (Flint, 1996; Hodapp, 1997); the identification of subgroups of individuals within a particular syndrome with unique characteristics (Hagerman, 2002); improved understanding of biological and environmental reasons for within-syndrome variability in behaviors and functioning (Dykens & Hodapp, 2001), and the development of syndrome-specific recommendations for intervention and treatment (Dykens & Hodapp, 1997; Hodapp & Fidler, 1999).

Rett syndrome (RTT) is one such disorder for which behavioral phenotype research may have a profound impact. RTT is a progressive neurodevelopmental disorder that occurs almost exclusively in females. The prevalence of RTT is estimated to be 1 in every 10,000 females, making it one of the most common causes of profound disability in girls (Ellaway & Christodoulou, 2001). The identification of mutations in the MECP2 gene in a majority of individuals with RTT (Amir et al., 1999) has increased understanding of the genetic basis for the disorder, although, to date, such mutations have not been found in all individuals presenting with the classic RTT phenotype, and more than 60 different mutations have been identified (Dragich, Houwink-Manville, &

Schannen, 2000; Shahbazian, & Zoghbi, 2001). As a result, the diagnosis of RTT remains a clinical one, based on behavioral and physical characteristics, rather than genetic tests alone (Hagberg, 2002).

RTT is characterized by an apparently normal pre- and peri-natal period, followed by deceleration of head circumference growth, loss of purposeful hand use, regression in cognitive, language, and social abilities, and the appearance of stereotypic hand movements (Hagberg, 2002). Although a number of variants of the disorder have been described (Hagberg & Gillberg, 1993; Hagberg & Skjeldal, 1994), with the exception of a preserved speech variant, a severe deficit in expressive language, along with a complete or nearly complete lack of functional hand use are virtually universal among individuals with RTT (Hagberg & Witt-Engerstrom, 1986; Hagberg, Hanefeld, Percy, & Skjeldal, 2002).

The progression of RTT has been reported to occur in a sequence of four relatively stable stages (Hagberg, 1995; Hagberg, 2002). Stage I, or the early onset stagnation stage, typically begins between 6 and 18 months of age. It is marked by a deceleration of head growth, as well as a delay in developmental progress, although the pattern of development may not yet be obviously abnormal. Typically between 1 to 3 years of age, the individual enters Stage II, or the developmental regression stage. This stage is characterized by severe regression in communication, language, and fine motor skills, usually resulting in a complete or nearly complete loss of speech and functional hand use. Stage II lasts between a few weeks up to a year, and is followed by Stage III, or the pseudostationary period. This period has been described as a wake-up period, as it is often characterized by the reemergence of some communicative behaviors. The characteristic stereotypical hand movements emerge during this period. Onset of Stage IV, or the late motor deterioration period, is typically around age 10, although it may occur much later in some. During this period, mobility becomes severely limited and scoliosis worsens. Most individuals become completely dependent on wheelchairs. Although cognitive skills may decline during this stage, this is not the case for all individuals (Engerstrom & Hagberg, 1990), and some may continue to develop new skills (Piazza, Anderson, & Fisher, 1993).

Communication in RTT

Loss of communication and communication impairments are among the diagnostic criteria for RTT. During the initial regression phase, affected individuals typically lose any spoken language that they had previously acquired, although many individuals never acquire any speech, and some main retain a few words (Hagberg, 2002). Stereotyped hand movements, accompanied by the loss of purposeful hand movements, make most alternative modes of communication, such as sign language, picture exchange systems, and many augmentative communication devices, inaccessible to the majority of individuals with RTT. The degree to which cognitive impairments further contribute to communication deficits is currently unclear, as there are no reliable and valid assessments available for this population (Byiers & Symons, 2013).

Many of the studies examining communication in RTT have relied on parents' perceptions of the affected individuals' comprehension and production capabilities. Overall, results from these types of studies suggest that many caregivers believe that their daughters understand much of what is said and are capable of communicating. In one study, 38% of caregivers interpreted their daughter's behaviors as having communicative meaning, and 23% reported that they could follow simple commands (Demeter, 2000). In another, 76% of caregivers reported that their daughters understood at least words or simple sentences (von Tetzchner, 1997).

Many studies involving direct observation of communication skills in this population have lead to very different conclusions. Initial studies by Woodyatt and Ozanne (1992, 1993, 1994, 1997) included a combination of caregiver interviews, sensorimotor assessments, and naturalistic observations conducted with small samples of individuals with RTT. The researchers noted that the participants had limited behavioral repertoires, and almost all demonstrated no clear communicative intent. They concluded that most individuals with RTT lack the developmental level necessary for the development of communicative intent. Nevertheless, all of the participants engaged in behaviors, such as facial expressions, gestures, body movement or other behaviors that caregivers interpreted as meaningful.

Most definitions of intentional communication require the coordination of attention between the desired object and the communicative partner (e.g., Sugarman, 1984). Identifying intentional communication among individuals with severe disabilities is made more difficult, however, by the sensory or motor impairments that may interfere with the ability to produce the behaviors necessary for demonstrating intentionality using traditional definitions (Iacono, Carter, & Hook, 1998). As a result, judging the intentionality of behaviors in this population requires some level of subjectivity. In an attempt to assess the functions of potentially communicative behaviors among individuals with RTT using more objective measures, Sigafoos, Woodyatt, Tucker, Roberts-Pennell, and Pittendreigh (2000) conducted structured observations in conditions designed to elicit communication with three adolescents with RTT. Although none of the participants had any spoken language, caregivers reported several behaviors that they viewed as communicative for each of the participants, and the observations indicated that these behaviors were differentially sensitive to the environmental conditions. In addition, they compared agreement between caregivers on the form and function of each participant's potentially communicative behaviors. They found that, although most caregivers agreed on the forms of the behaviors used by each individual to communicate, agreement on the functions of the behavior was poor. The authors concluded that, although the behaviors appeared to be sensitive to environmental influences, suggesting that they may serve communicative functions for the participants, they could not rule out the possibility that the behaviors were simply physiological, attentional, or orienting responses.

At an even more basic level, little research has been conducted to determine the degree to which individuals with RTT retain sufficient motor control to allow the reliable production of operant responses that could be used to communicate. Aside from studies examining the effects of environmental variables on the frequency of stereotypic hand movements, only two studies (Sullivan, Laverick, & Lewis, 1995; Watson, Umansky, Marcy, & Repacholi, 1996) have examined the influence of environmental contingencies on the operant motor behaviors of individuals with RTT. Both studies reported the results of interventions in which non-social environmental contingencies (e.g., toy activation) were used to increase operant motor behaviors (e.g., arm reaching and leg kicking). Although both studies reported increases in motor behavior, the Sullivan, Laverick, and Lewis study provided only narrative descriptions of the results without any objective data, making it impossible to confirm the investigators' interpretations. Although the Watson, Umansky, Marcy and Repacholi reported some data, in the form of average numbers of behaviors produced over time, there was no experimental design in place to control for threats to internal validity such as history or maturation. In addition, each study was conducted with one young child (3-5 years of age), making it difficult to determine whether the results would be applicable to a broader age range.

Communication Interventions in RTT

Despite the paucity of research regarding operant behaviors in RTT, some researchers have suggested that it may be possible for individuals with RTT to compensate for deficits in expressive language through the acquisition of alternative modes of communication (Burford & Trevarthen, 1997). To date, however, no evidence-based communication interventions currently exist for this population. In their review paper, Sigafoos et al.(2009) identified 9 studies in which communication interventions were implemented with at least one individual with RTT. A total of 31 participants participated across the studies. Overall, at least some positive gains related to communication were reported for 84% of the participants across the studies. All but one of the studies, however, were found to be inconclusive due to a lack of rigorous experimental designs, unclear description of the methods, or insufficient reporting of the data. The one study deemed to have conclusive results involved a computer-based intervention designed to teach requests for food and drink with three girls with RTT (Van Acker & Grant, 1995). Two of the three participants reached criterion for all seven symbols taught, whereas the third participant acquired a single symbol. All three of the participants showed evidence of simple discrimination skills, in that they requested preferred food/drink more frequently than non-preferred items.

Despite the limits in experimental design of the available studies, the consistent reports of increased behaviors in response to the interventions across the operant behavior and communication studies provides preliminary evidence of learning among individuals with RTT. This suggests that affected individuals may be capable of producing operant motor behaviors that could be used to communicate. Further research is needed, however, to identify effective intervention strategies for this population and to establish evidence-based practices for use in clinical and educational settings.

Functional Assessment (FA) & Functional Communication Training (FCT)

Researchers have suggested that caregivers can encourage the development of communication among children with severe and multiple disabilities by recognizing and responding to nonintentional behaviors in consistent ways (Siegel-Causey & Guess, 1989). In support of this strategy, previous studies have demonstrated the importance of caregiver responsivity to child behaviors on the communicative development of both typically developing children and those with developmental disabilities (e.g., Hancock & Kaiser, 2002; McDuffie, & Yoder, 2010; Tamis-LeMonda, Bornstein, & Baumwell, 2001; Yoder & Warren, 1999), although no studies have specifically examined the role of responsivity in RTT. Previous studies with children with other severe or multiple disabilities have shown that teachers and other caregivers may not respond to communicative attempts if they are unclear or subtle (Houghton, Bronicki, & Guess, 1987), as may be the case of the potential communicative acts (PCAs) exhibited by individuals with RTT. Therefore, by teaching caregivers to recognize and respond to PCAs consistently, or by replacing vague or subtle behaviors with more recognizable or conventional forms of communication may make it more likely that the behaviors will result in reinforcement for the child, thereby promoting their continued use (Sigafoos et al., 2000).

In addition to identifying the forms of PCAs, identifying the function of the behaviors may be helpful in developing effective communication interventions for individuals with severe disabilities (Drasgow & Halle, 1995). Skinner (1957) stated that the function of a communicative act is defined by its effects on the communicative partner, or listener. Within this context, all communicative acts serve to access positive reinforcement, (attention or other preferred events or activities), or negative reinforcement (escape from unpleasant situations) through the response of the listener. Therefore, identifying the function of an individual's existing communicative behaviors can help identify objects or events that are likely to be effective reinforcers for new communicative behaviors. In this way, an intervention designed to teach a new communicative behavior that would result in access to an outcome for which the learner already has a communicative form may be an efficient strategy.

Finally, assessment of PCAs facilitates the identification important contextual variables that may affect the individual's motivation to engage in communicative acts (e.g., Halle, 1992). Specifically, identifying the conditions under which an individual is likely to emit communicative behaviors can provide information about the relevant antecedents, and motivating operations that should be included within the intervention context to maximize the likelihood of the acquisition and maintenance of new forms of communication.

Functional communication training (FCT) was initially developed as an intervention to reduce challenging behaviors, such as aggression and self-injury, by teaching a new response to obtain the maintaining reinforcer (Carr & Durand, 1985). FCT interventions include two components: functional analysis to identify the source of reinforcement maintaining the behavior, including positive reinforcement in the form of access to attention, food, or preferred activities/objects, and negative reinforcement in the form of escape from task demands or other non-preferred environments or events, and training and reinforcement of a novel communicative behavior.

Nearly 30 years of research supports the effectiveness of FCT as an intervention for severe behavior problems in a wide range of populations (Tiger, Hanley, & Bruzek, 2008). More recently, however, researchers have begun to recognize the potential of FCT as a strategy for increasing communication in the form of requesting for individuals with severe disabilities without severe behavior problems. For example, Keen, Sigafoos, and Woodyatt (2001) used FCT methods to replace prelinguistic behaviors with symbolic communicative acts with three children with autism in a classroom setting. They found that the replacement forms increased and prelinguistic behaviors decreased across most of the communicative functions for each of the participants. Similarly, Tait, Sigafoos, Woodyatt, O'Reilly, and Lanioni (2004) taught parents to use FCT to teach a range of graphic and gestural symbolic communicative behaviors with six young children with developmental and physical disabilities. All six children demonstrated increases in the replacement behaviors and corresponding decreases in the prelinguistic behaviors following intervention.

Overall, these studies support FCT as a potential intervention for increasing improving the interpretability of communicative acts among children with disabilities. In both studies, however, the participants showed a broader range of behaviors than is typically observed among individuals with RTT, possibly due to their more advanced motor skills compared to those demonstrated by individuals with RTT. Participants in the Keen study had no physical disabilities, and many of the replacement behaviors selected were manual signs. Although the participants in the Tait et al. study all had physical disabilities (spastic quadriplegia was an inclusion criteria for the study), most participants were able to point or produce approximations of signs. Given the degree of motor impairment associated with RTT, it is unclear whether similar procedures would be effective with this population.

Chapter 3: Methods

Participants

All participants were recruited through physician referral from a Rett syndrome clinic at a local children's specialty care hospital, or through a regional Rett syndrome parent advocacy group. To be included in the study, all participants had to have a clinical diagnosis of RTT, with or without supporting genetic testing, and meet the criteria for a diagnosis of classic RTT outlined by Neul et al. (2011). Additionally, only participants without any functional spoken language were included. Although participants of all ages and levels of physical functioning were included, participants with an onset of a seizure disorder within the past six months were excluded, due to the possibility of impaired learning associated with seizure onset. Finally, to ensure feasibility of the study, only participants living within a one hour's drive of the Twin Cities metropolitan area (or within 90 minutes of the Twin Cities if more than one participant lived in the area and could be scheduled on single day) were recruited into the study.

A total of seven consecutive referrals were recruited into the study (see Tables 1 and 2 for participant characteristics). Participants ranged in age from 4 to 47 years, with a mean age of 20 years. The four youngest participants (Ella, Eden, Jen, and Veronica) were receiving special education and speech-language services through the public school system at the time of the current study. Alyssa was receiving private music therapy services twice per month. Rose attended a day program for adults with disabilities, but no information regarding programming was available from her parents or the group home staff. Tammy spent her days cared for by her grandmother, and personal care assistants, along with her twin sister who also has a diagnosis of RTT. She was not receiving any educational services at the time of current study.

Of the seven participants, only one (Eden) was reported to communicate reliably using an augmentative communication device (Tobii C12 eye tracker). Eden's parents reported that she occasionally made comments that appeared to be appropriate to the ongoing activities, although she would frequently "refuse" to respond to questions or instructions using the device by whining or crying when placed in front of it. At the time of the study, Eden only used her device at meal times, and information on the accuracy and reliability of her use of the device was unavailable. Five of the participants (Ella, Eden, Jen, Veronica, and Rose) were reported to respond to yes/no questions at least some of the time with gestures or other motor behaviors (e.g., head or arm movements), but no clear patterns of responding were observed during the course of the study.

With the exception of Rose, all of the participants had some previous experience with augmentative switches for communication and/or activation of toys and appliances. No information on the independence of switch-pressing was available for any of the participants, and none of the participants were using switches for communication at the time of the study.

All analysis and intervention sessions were conducted in quiet locations within the participants' residence. With the exception of Rose's FCT sessions, at least one parent or caregiver was present for all sessions. The research team implemented all conditions. All study procedures were approved by the University of Minnesota Institutional Review Board.

Procedures

Frequency counts for all dependent variables (described below) were collected during all assessment and intervention sessions. Inter-observer agreement (IOA) was calculated for a minimum of 25% (range = 27-78%) of all assessment and intervention sessions for each participant by a second in-person observer when possible, or using retrospective analysis of videotaped sessions. Percent total agreement was determined by taking the smaller frequency count, dividing it by the larger count and multiplying by 100 (Primavera, Allison, & Alfonso, 1997). IOA results for each participant are presented in Table 3.

Operational Definitions

Individualized operational definitions were developed for each participant's potential communication acts (PCAs). Eye contact (Ella) was defined as the participant orienting her eyes towards the face of the researcher for a period of 1 s or more. Vocalizations (Eden; Alyssa) were defined as any vocal sound that was not identifiable as a laugh or automatic bodily function (e.g., burp, hiccough). Whining (Jen) was defined as any vocalization that was of louder and of a higher pitch than her typical vocalizations, and that lasted longer than 1 s. Hand gestures (Veronica) were defined a movement of her right hand toward her mouth from a distance of at least 3 inches. Self-injurious behavior (SIB; Tammy) was defined as hitting self (forceful contact between the participant's open palm or closed fist and abdomen) or scratching (contact between one or more of the participant's finger nails and abdomen that included movement of the fingernails across the surface in a scratching motion with sufficient force to result in indentation of the skin). Grabbing (Rose) was defined as a closing motion of the participant's fingers around the researcher's arm or hand. For all participants, switch pressing was defined as any body movement that results in sufficient contact with the switch to activate the mechanism.

Functional Assessment

A functional assessment was conducted for each participant, consisting of semi-structured interviews, unstructured observations, and analog functional analyses using conditions similar to those described in Iwata et al. (1982/1994). Interviews were conducted with the each participant's primary caregiver. The Inventory of Potential Communicative Acts (Sigafoos et al., 2006) was used to elicit a list of potential communicative acts (PCAs) observed by the caregivers in different settings. The interview consists of a series of questions regarding how the participant communicated in different settings, or for different purposes, including greeting; requesting activities, toys, or food; protesting; requesting information, etc. If the caregiver was unable to provide a response for an item, the he or she was provided an example (e.g., "Does your child ever look at you and smile when you enter a room?" for greeting), and the function was probed again. Once a list of potential behaviors was developed, caregivers were asked to identify any behaviors that were problematic for the participant or the family, such as aggressive, disruptive, or self-injurious behaviors. Finally, caregivers were asked general questions about their child's preferences in order to set up the functional analysis sessions. Specifically, they were asked about preferred activities or toys (for the the free play condition), non-preferred activities or tasks that the participant was regularly expected to perform (for the escape condition), as well as objects or activities that they had noticed that their child would request using a specific behavior.

Following the interview, the caregiver was instructed to carry out some of the participant's daily activities, such as meal times, interactive play, educational tasks, and physiotherapy or occupational therapy exercises. Any activities that were noted to be highly preferred or not preferred during the interview were arranged, if possible. Throughout the observations, the research team recorded all instances of potentially communicative acts produced by the participant (based on the behaviors reported by Sigafoos, Woodyatt, Tucker, Roberts-Pennell, & Pittendreigh, 2000, plus any behaviors specifically identified by caregivers as meaningful not previously studied), as well as the activities in which the behaviors occurred, and the caregivers' responses to the behavior (i.e., provide attention, provide tangible, allow escape from task, ignore).

If the caregiver had reported the occurrence of a problem behavior, and that behavior occurred at least once during the observations, that behavior was selected as the target behavior for the subsequent analysis and intervention sessions. If no problematic behaviors were reported or observed, the behavior occurring most frequently during the observations was selected.

Functional analysis conditions were individualized for each participant based on information from the caregiver interview. For each participant, a minimum of one session of each of three assessment conditions (free play, escape, and attention) was conducted. Items or activities reported as preferred (i.e., TV shows, music, toys, interactive games) were included during the free play sessions. Tasks or activities that the participant was regularly expected to perform, but were not highly preferred (e.g., physical therapy exercises, daily living tasks) were used for the escape conditions. Additional individualized conditions (e.g., tangible, alternative forms of attention) were developed, as appropriate, based on other forms of reinforcement reported by the caregivers to be common consequences for the target behavior. All sessions were 5 minutes in length, with a one-minute break between sessions. A brief functional analysis design was used (e.g., Northup et al., 1991), with at least two sessions of the highest and lowest conditions conducted to ensure reliability of the results. If no clear function was identified in the brief design, additional sessions were conducted to create a multi-element design. A maximum of 20 functional analysis sessions was conducted with each participant, with no more than 90 min of sessions conducted per day.

The functional analysis condition associated with the highest rate of the target behavior during the functional analysis sessions was selected as the functional reinforcer for use in the FCT intervention.

Functional Analysis (FA) Conditions.

Free play. The free play condition was designed as a control condition, during which the participant had frequent access to attention and unrestricted access to preferred toys or activities. For four of the participants, this condition involved watching a preferred TV show while the researcher made comments about the content of the show. For Rose, this condition involved continuous access to head and neck massage and verbal comments. For Tammy, it involved laying on the floor next to her twin sister listening to the radio and frequent social comments from the researchers. For Alyssa, free play involved looking at family photo albums while the researcher made comments. For all of the participants, there were no demands, and no programmed consequences for any behaviors. It was expected that a low rate of communicative behaviors would occur in this condition.

Escape. The escape condition was designed to assess the influence of negative reinforcement in the form of escape from demands or non-preferred activities. The specific tasks or activities used with each participant were individualized based on caregiver report, with one academic, therapy-related, or daily living task that was reported to be least preferred selected for each participant. Activities included folding towels, pointing to pictures in a book, and stretching exercises. For all activities, the participant was physically prompted to engage in the task. Contingent on the occurrence of the target behavior, the participant was told "OK, you can take a break" and given a 10-15 s break from the task, during which all task materials were moved away from the participant and no verbal interaction or eye contact was provided.

Attention. The attention condition was designed to assess the influence of positive reinforcement in the form of access to attention. The participant was given access to a toy or activity and 10 s of attention in the form of neutral social comments (e.g., "That's a cool toy") and physical attention (e.g., pat on the back) at the beginning of the session. After 10 s, the participant was told "It's time for you to play by yourself for a little while. I need to do some work over here". All adults moved at least 5 feet away from the participant and refrained from making eye contact or speaking to the participant. Contingent on the occurrence of the target behavior, the researcher would walk over to the participant and make a social comment (e.g., “Look at the cool toy you have”) and provide some physical contact (e.g., pat on the back).

In addition to the typical attention condition, an individualized attention condition was conducted to assess the influence of a specific type of physical attention (massage) on the behavior of one participant (Rose), based on observations and caregiver reports. In this condition, the participant was given unrestricted access to verbal attention from the researcher, as well as preferred activities. At the beginning of the session, the participant was given 10 s of physical attention in the form of head and shoulder massages. After this initial period, the research told the participant that massages were over, but continued interacting verbally with the participant at regular intervals. Contingent on the occurrence of the target behavior, the researchers provided 10-15 s of massage.

Tangible. If a caregiver reported that his/her daughter used the target behavior to request food, activities, or items, a tangible condition was included in the analysis to assess the influence of positive reinforcement in the form of access to preferred stimuli. During these sessions, the participant was given a brief period of access to the stimulus (e.g., 10 s of access to TV or preferred toys, or a bite of preferred food) at the beginning of the session, and then was told that they would have to wait for more and the stimulus was removed. Contingent on the occurrence of the target behavior, the participant was given 10-15 seconds of access to the stimulus (or a bite of food in the case of an edible). The researcher continued providing verbal attention at regular intervals throughout the session.

Functional Communication Training (FCT)

Switch activation training. An initial session was conducted with each participant to determine the best size and response type (e.g., press, squeeze, toggle) for the voice output switch, as well as its placement (e.g., near hands, feet, head). 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 the identified reinforcer (e.g., 10-15 seconds of TV or massage). 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 5 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 get the reinforcer. Independent switch presses resulted in praise and a brief period of access to the functional reinforcer (e.g., TV or massage). 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 minute on five consecutive opportunities.

FCT intervention. Fllowing switch-activation training, 5-min FCT sessions were initiated. Each participant experienced two conditions: reinforcement for potential communicative acts (PCA), during in which the functional reinforcer was presented contingent on each occurrence of the participant's PCA, and switch pressing was placed on extinction (e.g., no consequences), and reinforcement for switch activation, during which the functional reinforcer was presented contingent on each occurrence of switch activation, and the PCA was placed on extinction. The two conditions were implemented in single-subject A-B-A or A-B-A-B reversal designs, with a minimum of three conditions per phase. The frequency of both the target behavior and the alternative behavior was recorded throughout all FCT sessions. No more than 90 min of FCT sessions were conducted per day.

For each participant, the set-up for the FCT sessions was identical to the set-up of the Functional Analysis session associated with the highest rate of responding and therefore varied across participants. For all participants, a verbal instruction was provided describing the contingencies in place (e.g., "Now if you want to watch TV, you need to press the switch") at the beginning of the first session of each new visit, or the first session of a new phase. No additional signals of changes in the reinforcement contingencies were provided. During each session, each occurrence of the target behavior resulted in a brief period of access to the functional reinforcer (e.g., 10-15 seconds or massage, or access to toys) , and all instances of the alternative behavior were ignored.

A minimum of three and a maximum of eight sessions were conducted per phase. A phase change was implemented when one of three results was achieved: (a) there was an evident trend in the expected direction for at least one of the dependent variables (i.e., switch activation or PCA) across at least three consecutive sessions, (b) there was no overlap in the rates of at least one dependent variable between the final three sessions of the current phase, and the last three sessions of the previous phase, or (c) rates of the target behavior being reinforced were higher than the alternative behavior across a minimum of three sessions.

Data Analysis

Frequency counts for the PCAs and switch-pressing were converted to rate per minute and a line graph was created for each participant. The results were analyzed using visual inspection, using the guidelines established by Kratochwill et al., 2010. Visual inspection involved analysis of changes in level, trend, and variability between adjacent phases to determine whether an experimental effect was evident. The two dependent variables (PCA, switch pressing) were examined separately to assess the degree of change between conditions. Additionally, the rates of the two dependent variables were compared within each phase to assess the degree to which the response being reinforced was successfully “competing” with the alternative response.

Non-overlap of All Pairs (NAP; Parker & Vannest, 2009), a single-subject index of effect size, was calculated as a quantitative measure of the degree of change between conditions for each dependent variable using the original frequency counts. NAP was designed as an indicator of performance differences between conditions or phases, as indexed by data overlap. It is calculated by comparing each phase A data point with each phase B data point and then dividing the number of pairs showing no overlap by the total number of comparisons. Specifically, if the difference between the two phases was in the expected direction, the pair was assigned a score of 1. If the difference was in the opposite direction from what was expected, the pair was assigned a score of 0. If the two points are of equal value, the pair was assigned a score of 0.5. The total number of comparisons was equal to the total number of data points in phase A multiplied by the total number of points in phase B.

Similar to other single-subject effect sizes, NAP scores range from 0.0 to 1.0. According to Parker and Vannest (2009), an 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, whereas a score of 0.5 would indicate that there is an even chance that the value of each point from phase A would exceed the value of a point from phase B. A score less than .5 would indicate deterioration in performance, in that scores during the baseline or control condition exceeded performance in the experimental phase.

Chapter 4: Results

Identification of Behavioral Function

The caregiver interviews resulted in the identification of at least one potential communicative act (PCA) for each participant. For four participants, the initial behaviors targeted were identified by caregivers as being problematic because they were disruptive to others in the environment (Eden, Jen, and Rose), or self-injurious, causing bruising or other tissue damage (Tammy). For the remaining three participants, no problematic behaviors were reported or observed. Therefore, the behaviors that occurred most frequently during the initial observations were targeted for these participants (Ella, Veronica, and Alyssa).

The results of the experimental functional analyses conducted with each participant are presented in Figures 1 through **7.** For six of the seven participants, the target behavior was differentially sensitive to one form of reinforcement, indicating a possible social function for the behavior. Partial or total agreement between the functions reported by the caregivers and those identified in the experimental analysis was established for six of the seven cases. Table 4 reports the target behavior and identified function for each participant.

Ella. Ella's mother and Personal Care Assistant reported that she used eye contact to communicate in a variety of contexts, including to request attention, food, and preferred toys. During the functional analysis (Figure 1) no instances of eye contact occurred during either the escape or attention conditions. No eye contact occurred during the first free play session, and one instance occurred during the second. The highest rates of eye contact occurred in the tangible (toys) condition, during which a toy with lights and music was activated contingent on the occurrence of the target behavior. The results support access to toys as a maintaining consequence for Ella’s eye contact.

Eden. Eden’s mother reported that she would frequently whine when a non-preferred TV show came on, or when the TV was turned off. During the functional analysis (Figure 2), no vocalizations were observed during any of the free play sessions. Some vocalizations occurred during the first of each of the attention and escape sessions, but not in the final sessions, which may be evidence of Eden’s learning the contingencies associated with the conditions. The highest, consistent rates of vocalizations were observed during the tangible (TV) condition. The results support access to preferred TV shows as a maintaining consequence for Eden’s vocalization.

Jen. Jen’s parents reported that she would whine whenever the TV was turned of, or set to a non-preferred show. The hypothesis that whining was maintained by access to preferred TV shows was supported by the results of the functional analysis (Figure 3): Whining occurred only during the tangible (TV) sessions, not during the free play, attention, or escape sessions.

Veronica. Veronica’s mother and PCA reported that she whined and used hand gestures to request food items when they were visible. No whining occurred during the observations or in any of the functional analysis sessions (Figure 4). Hand gestures were observed only during the tangible (food) condition, and not in the free play, attention, or escape sessions. These results support the hypothesis that Veronica’s hand gestures were maintained by access to food.

Tammy**.** Tammy's grandmother and PCA reported that she engaged in self-injurious behavior (SIB) in the form of self-hitting several times per day. They reported that it appeared to be related to fatigue because it occurred more frequently on days following nights with poor sleep. They did not believe that the behaviors served a social function for either individual. During the functional analysis sessions (Figure 5), SIB occurred only during the attention sessions, and not in the free play, escape, or tangible (food) sessions, suggesting that the behavior was maintained by access to attention from caregivers.

Alyssa**.** Alyssa's mother reported that she did not reliably use any behaviors to communicate. During the observation sessions, Alyssa frequently held her breath for periods of 3-5 s. It was also observed that Alyssa would emit brief vocal sounds that frequently co-occurred with the breath holding. Alyssa's mother reported that both behaviors occurred frequently throughout the day, regardless of the ongoing activities and she did not believe that the behavior served any social functions. The results of the functional analysis of vocal sounds (Figure 6) indicated that the target behavior occurred in some conditions (e.g., tangible/food; escape) more than in others, although relatively high rates occurred across all sessions. Therefore, it remained unclear whether the vocalizations were operant behaviors maintained by multiple forms of reinforcement (e.g., escape from tasks, access to food), or were non-social orienting or physiological responses. Because no other potential communicative behaviors were reported or observed, vocalizations were retained as the target behavior for the FCT intervention, and the consequence associated with the FA condition with the highest rates (tangible/food) was used as the reinforcer.

Rose. Interviews were conducted with Rose's parents, and a staff person at her residence. Both reported that she frequently grabbed the arms or hands of people in the environment. They reported that they believed that this behavior served as a request for head and neck massages. This hypothesis was supported by the results of the functional analysis (Figure 7), during which high rates of grabbing were observed in the attention (massage) sessions, lower but non-zero rates occurred during the attention (verbal), and escape sessions, and no instances of the target behavior occurred during the free play sessions.

Functional Communication Training

All seven participants achieved the mastery criteria for the initial switch activation training within a single session, ranging in length from 15 to 90 min. Six of the participants (86%) were able to reliably activate the switch with their arms or hands. One participant (14%; Eden) did not reliably activate the switch with her arms or hands following 45 minutes of training. As a result, her positioning was altered so that she sat in a chair with the switch placed behind her right foot during intervention sessions. In this position, Eden was able to reliably move her foot backwards to activate the switch, and she successfully met the mastery criteria within 30 minutes.

Ella. Ella's FCT results are presented in Figure 8. Across the three phases, there was strong evidence of experimental control over Ella's rates of switch pressing. During the first phase, in which reinforcement was available for switch pressing, but not eye contact, Ella's rates of switch pressing were consistently high, ranging from 1.2 to 1.8 presses per minute. A large and immediate decrease was observed when the contingencies were reversed in phase two, (range = 0 to 0.4). A return to the initial contingencies replicated the results of the first phase, with rates ranging from 1.0 and 2.0 times per minute. The Nonoverlap of All Pair (NAP) index of .96 indicates little overlap in Ella's rates of switch pressing between the FCT conditions.

Evidence of experimental control over Ella's rates of eye contact was somewhat weaker. In the first phase, rates of eye contact were low (0.4 times per minute or less) in three of the four sessions, but rates in session 3 were higher (1.2). Rates in the second phases were somewhat higher and more stable (range = .08-1.2) than most of the sessions in phase 1. Finally, Ella's rate of eye contact showed a small, but immediate decrease when the contingencies were reversed again in the final phase, with rates ranging from 0.4 to 0.8 responses per minute. The associated NAP of .88 indicates that, although there was some overlap between the conditions, the overall patterns support the presence of an experimental effect on Ella's rates of eye contact throughout the study. Comparing rates of the two behaviors within each session, Ella produced higher rates of the behavior that resulted in reinforcement compared to rates of the alternative behavior in 100% of sessions.

Eden. The results of Ella's FCT study are presented in Figure 9. Eden's rates of switch pressing were relatively high and stable during the first phase (range = 0.2 – 0.8), during which reinforcement was available for switch pressing. A small and delayed decrease in Eden's rates of switch pressing was observed when the contingencies were reversed in the second phase (range = 0.2 – 0.6). A large and immediate increase was observed in the final phase (range = 0.8 to 1.0). Visual inspection of these results suggests a small but reliable experimental effect. The NAP index of .84 indicates some overlap between conditions.

Experimental control over Ella's rates of whining was much stronger overall, as there was very little overlap between the experimental conditions. During the first phase, in which the TV was provided contingent on activation of the switch, Eden's rates of whining were very low (range = 0 – 0.2). Rates were higher in the second phase (range = 0.2 – 0.8). A return to the initial conditions resulted in an immediate and persistent decrease (range = 0 to 0.2). Overall, visual inspection of these data suggests a strong effect of the contingencies on Ella's whining. The associated NAP of .96 indicates that there was very little overlap between conditions.

Comparison of Eden's rates of the two behaviors within each session indicates that she produced higher rates of the behavior that resulted in reinforcement in 11/13 (85%) of sessions. Closer examination indicates that, during 100% of sessions in which switch pressing resulted in access to the TV, Eden produced more switch presses than vocalizations. On the other hand, Eden produced more vocalizations than switch presses in only the final two sessions (50%) of the four-session condition in which vocalizations resulted in access to the TV.

Jen. In the first phase of Jen's FCT study (see Figure 10), reinforcement was available for whining, and switch pressing was ignored. A clear increasing trend in the rates of whining was observed across the sessions of the first phase (range = 1.0-1.6). An immediate decrease was observed when the contingencies were reversed in phase 2, although Jen's rates of whining remained at a moderate level (range = 0.6 to 1.0). Return to the initial conditions in phase 3 resulted in high stable rates of whining (range = 1.6 – 1.8), which was reversed again in phase 4 (range = 0.6 – 1.0). Overall, visual inspection indicates a strong relationship between the experimental conditions and Jen's rates of whining. The NAP associated with Jen's rates of whining was .96, indicating very little overlap between the experimental conditions.

Jen's rates of switch pressing were somewhat more variable across the experimental conditions. During the initial phase, when switch pressing was ignored, the rate per minute ranged from 0 to 1.0, but many of the switch activations during this phase occurred while Jen was rocking from side to side. This behavior resulted in Jen knocking the switch with her hip or arm, and these activations may have been accidental. Rates during the first two sessions of the second phase were similar to those in the first phase, but a clear increase occurred in the third phase, and this persisted until the end of the phase. Similarly, a decrease in the rate of switch pressing occurred in the third session of phase 3. When reinforcement for switch pressing was reintroduced in the final phase, however, a clear and immediate increase in switch pressing was observed (range = 1.2 – 1.6). Visual inspection of these results suggests that, by the end of the study, the experimental condition exhibited strong experimental control over Jen's rates of switch pressing. The NAP index of .74 indicates that there was a fair amount of overlap between conditions when all of the phases are considered, but the NAP of .85 for the last two phases indicates less overlap towards the end of the study.

Across the 18 sessions, Jen produced the behavior that resulted in reinforcement more frequently than the alternative behavior in 16 (89%) of the sessions. Specifically Jen produced more whines than switch presses in all of the sessions during which whining resulted in access to the TV, but she produced more switch presses than whines in eight of ten (80%) sessions in which switch pressing was required to access the TV.

Veronica. Veronica's FCT study was terminated before the criteria for completion were met. The results from the initial six sessions are presented in Figure 11. During phase one, reinforcement was available for switch pressing and Veronica demonstrated a clear increasing trend in the rate of switch pressing, and a clear decreasing trend in the rate of gestures across the three sessions. Both dependent variables showed relatively stable levels across the initial sessions of the second phase, but the levels do not differ substantially from those in the previous phase.

A comparison of the rates of switch pressing and gestures within sessions indicates that Veronica produced the response required to obtain access to food in only two of the six (33%) sessions conducted. Specifically, Veronica produced more switch presses in two of the three sessions during which switch pressing resulted in access to food, and in all three sessions during which hand gestures were reinforced.

During session 6, Veronica's mother asked that the gesture reinforcement sessions be terminated because Veronica was engaging in high rates of tooth grinding. Subsequently, a functional assessment was conducted to determine whether Veronica's tooth grinding served a social function. It was determined that the behavior occurred at high rates only when food was visible in the environment, but it did not appear to be sensitive to reinforcement contingencies. Further FCT procedures involving food were excluded with Veronica. Additional observations were conducted to determine whether an alternative communicative behavior serving a different function could be identified. Several potential behaviors that appeared to be maintained by access to food were excluded, but no other communicative behaviors occurred during the observations. As a result, Veronica's participation in the study was terminated.

***Tammy*.** The results of Tammy's FCT intervention are presented in Figure 12. Overall, evidence for experimental control over Tammy's rates of switch pressing was very strong. During the first phase, during which reinforcement was available for self-injurious behavior (SIB), Tammy engaged in consistently low rates of switch pressing (range = 0 – 0.4). The second phase resulted in an immediate increase in switch pressing (range = 1.0 – 4.0). When the reinforcement contingencies were reversed again, Tammy's rates of switch pressing were somewhat lower, but variable (range = 0.2-1.4), eventually showing a decreasing trend in sessions 13-15. An immediate and persistent change in level was observed when the final phase was implemented in session 16 (range = 2.0-3.2). The NAP associated with Tammy's rates of switch pressing was .92, indicating little overlap between the conditions.

Evidence for experimental control over Tammy's rates of SIB was weaker. In the initial phase, rates of SIB were variable (range = 0.6 – 3.0), when reinforcement for SIB was removed in the second phase, there was no immediate change, but a general decelerating trend occurred towards the end of the phase (range = 0 – 1.0). Similarly, no immediate change was observed when reinforcement was reimplemented in the third phase, but an accelerating trend was eventually observed (range = 0 – 1.6). Finally, rates of SIB showed an immediate decrease when reinforcement was withdrawn once again, although the high rate in the last sessions overlapped with many of the sessions in the "Attention for SIB" phases. The NAP associated with Tammy's rates of SIB was .64, indicated a substantial overlap between the conditions.

Comparison of Tammy's rates of the two dependent variables shows that she produced more instances of the behavior for which reinforcement was available in 14 of the 18 (78%) of sessions. Specifically, when reinforcement was available for switch pressing, Tammy produced more switch presses than instances of SIB in all but the first session (8 of 9; 89%). When reinforcement was available for SIB, however, Tammy produced more instances of SIB in only 6 of 9 sessions (67%).

Alyssa. The results of Alyssa's FCT intervention are presented in Figure 13. Clear differentiation in rates of switch pressing occurred across the three phases of the study, with relatively high rates (range = 1.6 – 3.2 responses per minute) occurring in the first phase when food was provided contingent on switch pressing. When reinforcement was provided for vocalizations in phase 2, no switch pressing occurred. Finally, when the initial contingency was reimplemented in phase 3, an increasing trend was observed (range = 0-0.4). Visual inspection supports a moderate to strong experimental effect on switch pressing. The NAP of .93 indicates very little overlap between the phases.

No clear changes in Alyssa's rate of vocalizations occurred during the FCT intervention. During the initial phase, rates ranged from 0.6 to 1.4 times per minute. No change was observed when reinforcement for vocalizations was introduced in phase 2 (range = 0.8-1.2). In phase 3, rates became more variable, ranging from 0.4 to 1.6 responses per minute. Overall, the effects of the FCT intervention on Alyssa's rate of vocalizing are unclear. The associated NAP was .50, indicating a great deal of overlap between the phases. Comparison of Alyssa's rates of the two dependent variables indicates that Alyssa produced more vocalizations than switch presses across 9 of the 10 sessions of the study.

***Rose*.** In the first phase of the FCT study (see Figure 4c), reinforcement was available for grabbing, and switch pressing was ignored. Rose exhibited high and stable rates of grabbing across the three sessions (range = 3.6-4.2). An immediate decrease was observed when the contingencies were reversed in phase 2 (range = 0 to 1.6). Return to the initial conditions in phase 3 resulted in an immediate increase, as well as an accelerating trend across the sessions (range = 3.2 – 5.4), which was reversed in phase 4 (range = 0.8 – 2.0). Overall, visual inspection of these indicates a strong relationship between the experimental conditions and Rose's rates of grabbing. The NAP index associated with grabbing was 1.0, indicating no overlap between the phases.

There was similarly strong evidence for experimental control of the FCT conditions over Rose's rates of switch pressing. During the initial phase, when switch pressing was ignored, Rose pressed the switch at very low rates (range = 0 – 0.2). An immediate increase occurred when the contingencies were reversed in the second phase (range = 0.8 = 3.0). In phase three, rates of switch pressing decreased to near 0 levels immediately (range = 0 – 0.4). When reinforcement for switch pressing was reintroduced in the final phase, Rose showed an immediate and persistent increase in switch pressing (range = 1.8 – 3.6). Because there was no overlap between the experimental phases, the NAP associated with switch pressing was 1.0.

Within the sessions, Rose produced more instances of the behavior that resulted in reinforcement than of the alternative behavior in 15 of the 18 sessions (83%). Within the sessions during which reinforcement was available for grabbing only, Rose produced more instances of grabbing than switch pressing in all 6 sessions (100%). On the other hand, when reinforcement was provided contingent on switch pressing, Rose produced more switch presses than grabs during 9 of the 12 sessions (75%). This occurred because rates of the two dependent variables were nearly identical in the first four sessions of the first phase in which switch pressing was reinforced.

Chapter 5: Discussion

Research Question 1

Will the participants increase the frequency of their PCAs in response to one or more types of positive and/or negative reinforcement contingencies (as compared to the control condition) within the context of experimental functional analysis (FA) sessions?

Overall, there is strong evidence that for six of the seven participants the PCAs identified by the participants' caregivers, or during the research team's observations were sensitive to reinforcement contingencies, as indicated by higher rates of PCAs identified in one or more of the reinforcement (positive or negative) conditions compared to the free play (control) sessions. In fact, for six of the seven participants, the target behavior occurred exclusively, or nearly exclusively in a single FA condition, suggesting that specific environmental variables (e.g., the presence of food, or the caregiver's attention being focused elsewhere) had developed strong stimulus control over the participant's behaviors. Because the free play sessions were designed to minimize the participants' motivation to act to access preferred items or events, the low rates that occurred during these sessions provide support for the hypothesis that most of the individuals were able to discriminate the environmental conditions and consequences in place, and produce the behaviors only when they would produce access to a preferred consequence.

The one exception to this pattern was Alyssa. In her case the PCA (vocalizing) occurred frequently (at least once per minute) across all of the functional analysis sessions. Visual inspection of the FA results show that there was differentiation between experimental conditions, with the highest rates reliably occurring in the tangible (food) condition, and the lowest rates occurring in the attention condition. This suggests that environmental stimuli influenced Alyssa's rates of vocalizations. It remains unclear, however, whether the observed differences were physiological or orienting responses due to changes in the antecedent conditions, such as the amount or type of social interaction, or due to changes in the reinforcement contingencies in place. Although it is possible that unintended motivating operations were in place during the free play sessions, resulting in the occurrence of communicative behaviors, the non-contingent availability of attention and preferred activities, as well as Alyssa's mother's reports that the target behaviors did not typically result in any consequences in Alyssa's typical environment, seem to support the hypothesis that the behaviors were not operant in nature. Unfortunately, no empirical methods currently exist for conclusively discriminating between operant and respondent behavioral processes in such cases.

Overall, the high degree of agreement between the experimental FA FCT results and the caregivers' reports of the functions of the PCAs provides support for the inference that many parents make about their daughters' behaviors. In only one case (Ella), did the participant's caregivers report possible functions that were not confirmed in the FA. Specifically, Ella's mother reported that she used eye contact to communicate about a variety of different things, including to request attention, food, and toys. Access to food was not tested in Ella's FA sessions, but no instances of eye contact occurred during the attention session. This may have been due to issues related to positioning, however, as Ella could not sit up unsupported, and was placed on her back on the couch during this condition, possibly making it difficult for Ella to see and make eye contact with the researchers. Regardless, there was at least partial agreement between Ella's mother's report of the function of her behavior and the FA results.

On the other hand, Tammy's caregivers reported that she did not reliably use any behaviors to communicate with those in her environment. Tammy's grandmother considered her self-injurious behavior (SIB) to be a direct result of her RTT diagnosis, and, reported that it was "just something she does". Neither Tammy's grandmother nor the personal care assistants who worked with her regularly considered the SIB to be a communicative behavior. The functional analysis results, however, indicated that Tammy's SIB was differentially sensitive to adult attention. This finding was complimented by observations of Tammy's daily routine, during which SIB occurred only when all of the adults turned their attention to Tammy's twin sister. In addition the caregivers consistently responded to Tammy's episodes of SIB by trying to calm her down using physical restraint and soothing voices.

Across participants, the results suggest that many individuals with RTT may be using the limited behaviors available to them to access preferred items and events in their environment. This provides support for the argument put forward by Burford and Trevarthen (1997) that caregivers who consistently respond to their daughter's behaviors as though they were intentionally communicative might facilitate the development of intentional communicative responses. Understanding the relationship between caregiver-child interactions and communicative competence among individuals with RTT may be an important step in the development of effective, long-term strategies for improving communication in this population.

Research question 2

Will the participants learn to produce a novel motor response (activating a voice-output switch) to obtain the functional reinforcer associated with the highest rate of responding during the FA sessions?

Given the extremely limited body of research demonstrating the capacity of individuals with RTT to reliably produce motor responses to impact their environments (e.g., Sullivan, Laverick, & Lewis, 1995; Watson, Umansky, Marcy, & Repacholi, 1996), the degree to which each participant would be capable of reliably activating the voice-output switch was unclear prior to the beginning of the study. Each of the participants, however, quickly learned to activate the switch, and all seven participants reached the switch training mastery criteria (5 consecutive independent responses with no more than one minute response latency) within a single session once an appropriate response mode was identified. Despite the manual motor impairments associated with RTT, all but one of the participants used one or both of her hands to activate the switch on the majority of occasions.

Research Question 3

Will the participants' rates of the two available responses (e.g., PCA and switch-activation) vary according to changes in the environmental contingencies (e.g., reinforcement vs. extinction)?

Across the seven participants, five showed moderate to strong evidence of experimental control over rates of both switch pressing and PCAs, in that overall rates of the target behavior were higher in sessions when reinforcement was available for that response compared to sessions in which reinforcement was available for the other response. NAP indices for these five participants ranged from .74 to 1.0 and .64 to 1.0 for switch presses and PCAs.

Similarly, when the rates of switch pressing and PCAs were compared within session for each participant, the same five participants showed higher rates of the target behavior that resulted in reinforcement compared to the alternative behavior in the majority of sessions (range = 78 – 100%).

Taken together, these results indicate that the behaviors of these five individuals were sensitive to the reinforcement contingencies in place, and that the two behaviors were effectively functioning as members of the same response class, or group of behaviors that differ in topography, but produce the same effect on the environment (Carr, 1988; Catania, 1998). This provides further support for the hypothesis that the PCAs identified by the caregivers functioned to access reinforcers for these participants in their daily lives, and that these behaviors were not merely physiological or orienting responses to events in the environment.

Of the participants who completed all phases of the study, the only exception to the general pattern of responding was Alyssa, who showed strong evidence of experimental control over switch pressing, but no clear changes in her rate of vocalizing across conditions. There are two possible explanations for this result. The first is that there was a longer reinforcement history associated with the vocalizing behavior, as it had presumably been in Alyssa's repertoire for many years, and this longer history of presumably intermittent reinforcement resulted in greater behavioral persistence than was observed for the novel behavior of switch pressing. Given that Alyssa's mother reported however, that none of her caregivers typically responded to the vocalizations in any consistent way, and no caregiver responses to vocalizations were observed at any point during the study. The second hypothesis is that Alyssa's vocalizing was due to changes in arousal, and as such the behavior was not a true conditioned operant. This is supported by the fact that vocalizations typically co-occurred with episodes of breath-holding, which appears to be related to autonomic dysfunction in RTT (Julu, Kerr, Hansen, Apartopoulos, & Jamal, 1997), In addition, Alyssa frequently vocalized during the reinforcement period during the FA and FCT sessions, (after the edible was placed on the table, and/or while she was chewing). Because the establishing operation of restricted access to edibles was not in place during this time, the rate of vocalizations during the reinforcement period should have been close to zero if the function of the behavior correctly identified. However, there is currently no definite method for ruling out either explanation.

Overall, Alyssa's results are somewhat surprising, because, although the research team was unable to identify any behaviors that Alyssa routinely used to influence her environment through interviews, observations, and experimental manipulations, clear changes in her rates of switch pressing across experimental conditions during the FCT study suggest that Alyssa is capable of producing operant behaviors in order to access reinforcers. This outcome further underscores the importance of caregiver responsiveness to potential communicative behaviors exhibited by individuals with RTT, as it is possible that Alyssa had exhibited behaviors in the past that could have become functional ways for her to effect changes in her environment. Her parents' perceptions that she was unable to produce communicative responses may have led them to avoid attributing intentionality to her behaviors, thereby reducing the likelihood that they would be shaped and maintained over time.

Veronica's results are in contrast to those of the other participants, as she did not show clear evidence of discrimination across the experimental conditions in either of the dependent variables. Once Veronica acquired the switch-pressing response, she continued to produce it to the exclusion of the previously observed hand gestures, despite changes to the reinforcement contingencies. Because her involvement was terminated after only six sessions, however, it is impossible to know whether she would have shown evidence of learning given more opportunities.

Limitations

Several limitations of the current study should be noted. First, the participants represent seven consecutive referrals, and likely do not represent the full range of individuals with RTT, although the participants ranged widely in age and levels of physical functioning. Additionally, individuals with variant forms of RTT were excluded from the study, although similar questions also apply to these populations.

As is the case in many research studies, the sample was likely biased by the fact that all of the participants were Caucasian, and most lived in households of relatively high socio-economic status, with highly educated caregivers. Because previous studies have demonstrated that socio-economic status and maternal education are highly correlated with a child's cognitive, language, and social outcomes (e.g., Deutch, 1965;

Teasdale, & Katz, 1968), it is possible that the individuals who participated in this study represented the best-case scenarios with regard to communicative competence as could be expected for this population, in the absence of intensive intervention.

Another limitation was the lack of follow-up and maintenance testing to determine whether any of the families continued to use the switches as a method of communication, or whether the switch-pressing response would have maintained over time in the participants' typical environments. As a result, no information about the long-term utility and feasibility of FCT procedures for improving communication in this population is available.

Finally, only one communicative behavior and function was selected for each participant. It is likely that each of the participants had several behaviors that served multiple functions, and that each of these could have been identified given enough time. Ultimately, any successful communication intervention would provide the participants with the means to produce a variety of responses to communicate a range of different messages.

Future Directions

A great deal of further research is needed to clarify the issues related to learning and communication among individuals with RTT. This study was a preliminary assessment of the feasibility of FCT strategies for teaching individuals with RTT to activate voice output switches, and further research is needed to determine the degree to which such procedures could be used to produce long-term changes in communication among individuals with RTT. Future studies should also examine the degree to which individuals with RTT can learn to produce multiple communicative responses to serve more than one function, as well as the degree to which individuals with RTT can discriminate the appropriate stimulus conditions for producing one response rather than another.

There were wide ranges in frequency and variety of communicative behaviors among the seven participants in the current study, and future research should examine possible reasons for this variability, including genetic differences such as mutation types and x-inactivation patterns; differences in motor skills and adaptive behavior; previous and concurrent medical, behavioral, and educational interventions the participants may have received, and variation in parent-child relationships, including parent responsiveness to potential communicative acts, and parent perceptions of child communicative competence.

Work revisiting the concept of intentional communication among individuals with RTT is needed to clarify the issue of whether communicative deficits in this population result primarily from cognitive deficits, motor deficits, or both. The pervasive belief among those the research community that individuals with RTT lack the developmental level necessary to demonstrate intentional communication dates back to a single, small-scale study from nearly two decades ago (Woodyatt & Ozanne, 1994). Interactions with parents active in the RTT community today indicate that many caregivers believe that their daughters are fully capable of engaging in complex communicative interactions through the use of novel technologies such as eye tracking communication systems. Therefore, additional work is needed to reconcile these two conflicting viewpoints.

Finally, as new pharmacological treatments directed at improving the symptoms of RTT become available, the degree to which behavioral interventions such as FCT can be used in conjunction with pharmacological interventions to produce the best possible outcomes for individuals with RTT needs to be examined.

Conclusions

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, 1993; Watson Umansky, Marcy, & Repacholi, 1996;). Evidence regarding intentional communicative behaviors, however, has been harder to obtain. Although several studies have demonstrated that potential communicative acts exhibited by individuals with RTT vary in frequency and intensity in different social contexts (e.g., Sigafoos, Woodyatt, Tucker, Roberts-Pennell, & Pittendreigh, 2000; Oliver, Murphy, Crayton, & Corbett, 1993), the research designs used in these studies have not ruled out the possibility that these behaviors may be orienting or physiological responses rather than intentional or unintentional operant behaviors.

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 majority of 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. This study provides preliminary evidence that at least some individuals with RTT use specific behaviors to affect their environments through their caregivers, and that they can quickly learn to switch their communicative signals in response to changes in environmental contingencies.

Table 1.

Summary of RTT syndrome diagnostic criteria.

RTT diagnostic criteria	Participants						
	Ella	Eden	Jen	Vienna	Alyssa	Tammy	Rose
Main criteria							
Period of regression followed by recovery/ stabilization	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Partial or complete loss of acquired purposeful hand skills	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Partial or complete loss of acquired spoken language	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Gait abnormalities	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Stereotypic hand movements	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Supportive criteria							
Breathing disturbances when awake	Yes	No	Yes	Yes	Yes	No	No
Bruxism when awake	Yes	Yes	Yes	Yes	No	No	No
Impaired sleep pattern	No	No	Yes	Yes	No	Yes	Yes
Abnormal muscle tone	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Peripheral vasomotor disturbances	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Scoliosis/kyphosis	No	No	Yes	Yes	Yes	Yes	Yes
Growth retardation	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Small, cold hands and feet	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Inappropriate laughing/screaming spells	No	Yes	Yes	No	Yes	Yes	Yes
Diminshed response to pain	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Intense eye communication	Yes	Yes	Yes	Yes	Yes	No*	Yes
Exclusion criteria							
Brain injury secondary to trauma, neurometabolic disease, or severe infection	No	No	No	No	No	No	No
Grossly abnormal psychomotor development <six months	No	No	No	No	No	No	No

* Tammy has a diagnosis of cortical blindness.

Table 2.

Summary of participant characteristics and physical functioning.

Participant	Age	Genetic mutation	Caregiver interviewed	Mobility	Highest hand function	Seizure disorder
Ella	4	C.30_31	Mother and nursing staff	Non-ambulatory	Eats finger foods	No
Eden	4	R168X	Mother	Walks unassisted	Eats finger foods	No
Jen	15	K114X	Mother and father	Walks with some assistance	Eats finger foods	Yes, not controlled
Vienna	17	Unsure	Mother	Non-ambulatory	Eats finger foods	Yes, controlled
Alyssa	26	Not tested	Mother	Walks unassisted	Eats finger foods	Yes, controlled
Tammy	27	Not tested	Grandmother	Non-ambulatory	Drinks from sippy cup	Yes, controlled
Rose	47	Not tested	Group home staff	Walks unassisted	Eats with spoon	No

Table 3.

Interobserver agreement results for functional analysis (FA) and functional communication training (FCT) sessions.

Participant	FA		FCT	
	Range	Mean	Range	Mean
Ella	100	100	83-100	89
Eden	82-86	85	100	100
Jen	83-100	94	87-100	98
Veronica	100	100	-	-
Tammy	83-100	94	80-100	96
Alyssa	85-90	87	82-94	88
Rose	84-100	92	92-100	96

Table 4

Summary of functional analysis results

Participants	PCA	Caregiver reported function	Experimentally identified function
Ella	Eye contact	Multiple	Tangible (toys)
Eden	Vocalizations	Tangible (TV)	Tangible (TV)
Jen	Whine	Tangible (TV)	Tangible (TV)
Veronica	Gesture	Tangible (food)	Tangible (food)
Tammy	Hit self	None	Attention (verbal)
Alyssa	Vocalizations	None	Unclear
Rose	Grab arm	Attention (massage)	Attention (massage)

Figure 1.

Rates of eye contact during functional analysis sessions with Ella.

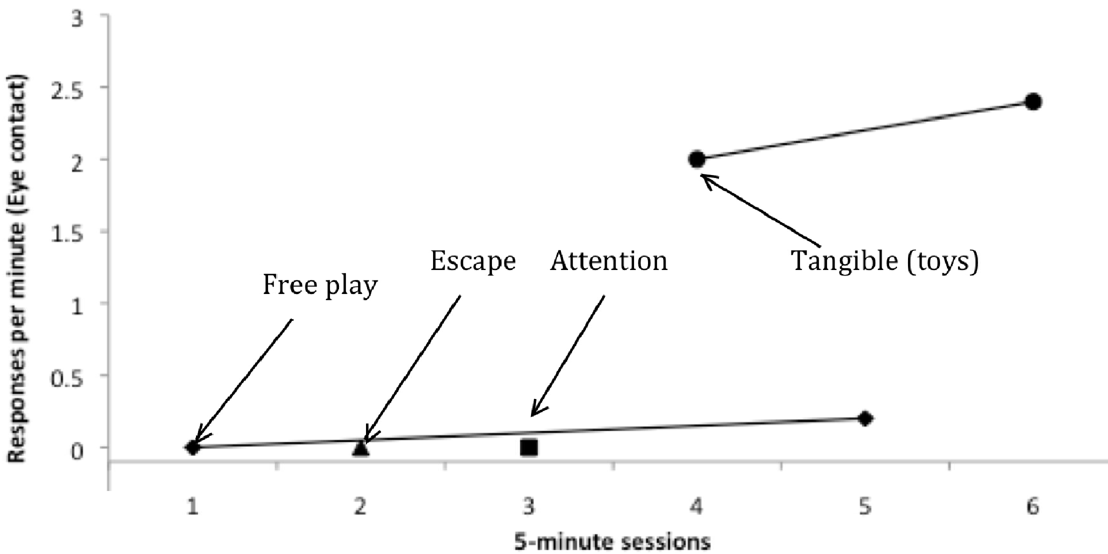


Figure 2.

Rates of whining during functional analysis sessions with Eden.

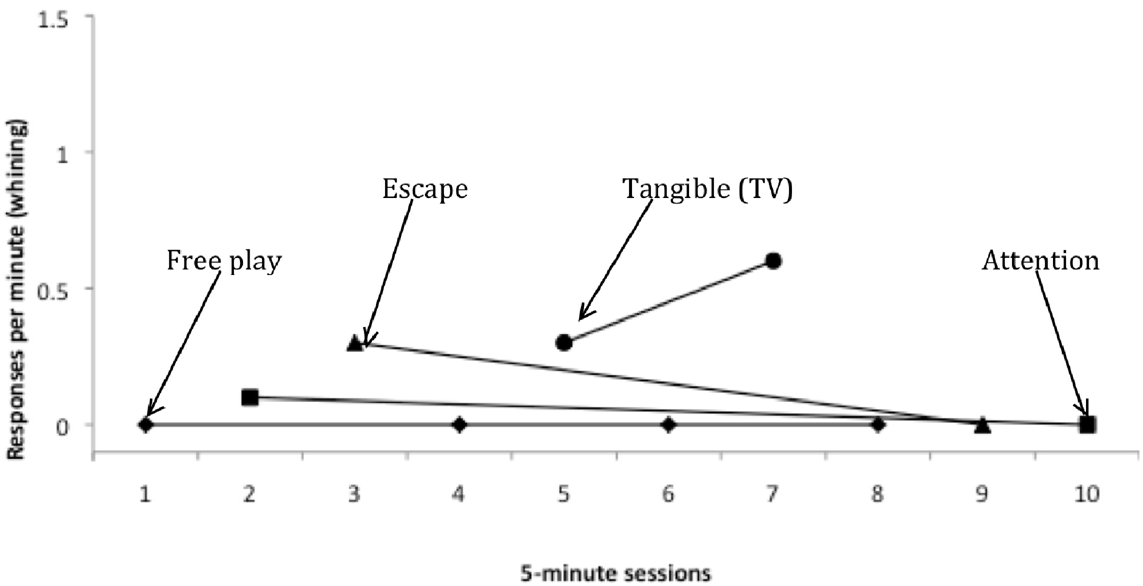


Figure 3.

Rates of whining during functional analysis sessions with Jen.

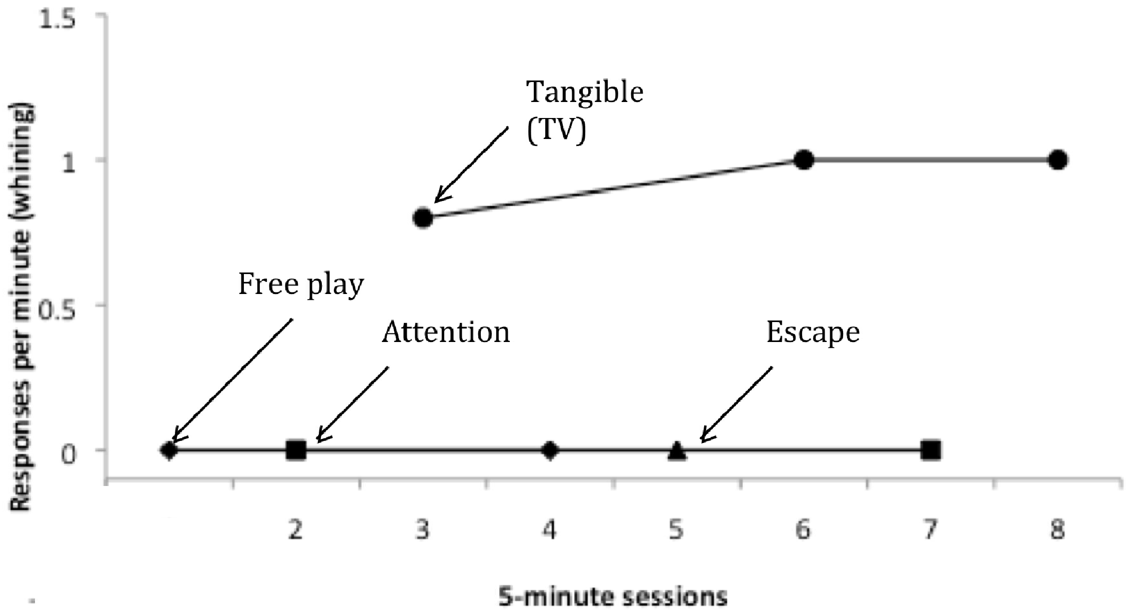


Figure 4.

Rates of hand gestures during functional analysis sessions with Veronica.

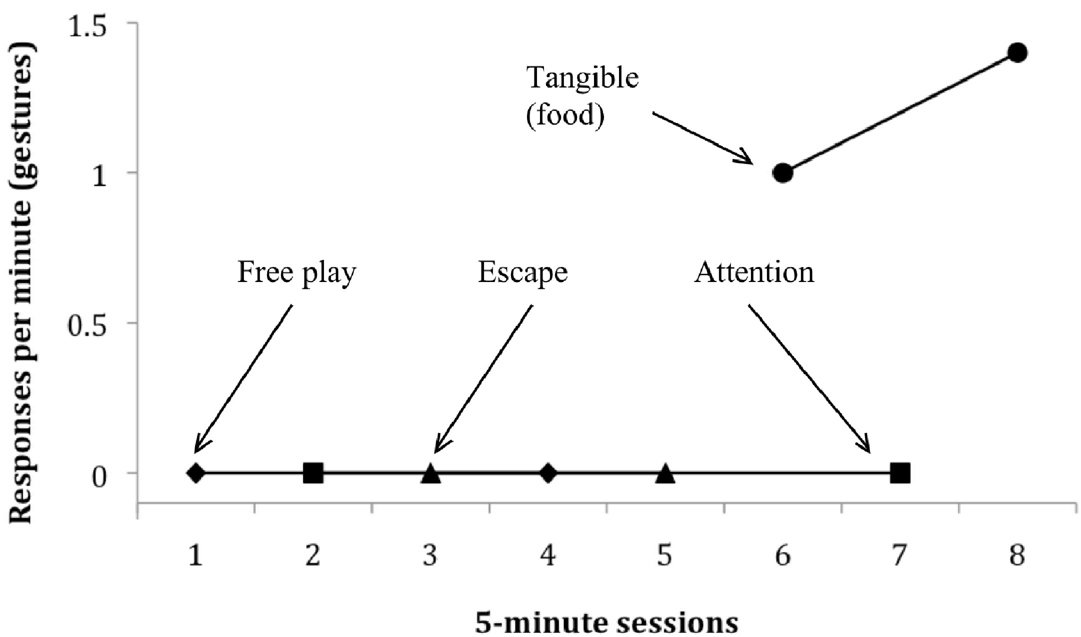


Figure 5.

Rates of self-injurious behavior (SIB) during functional analysis sessions with Tammy.

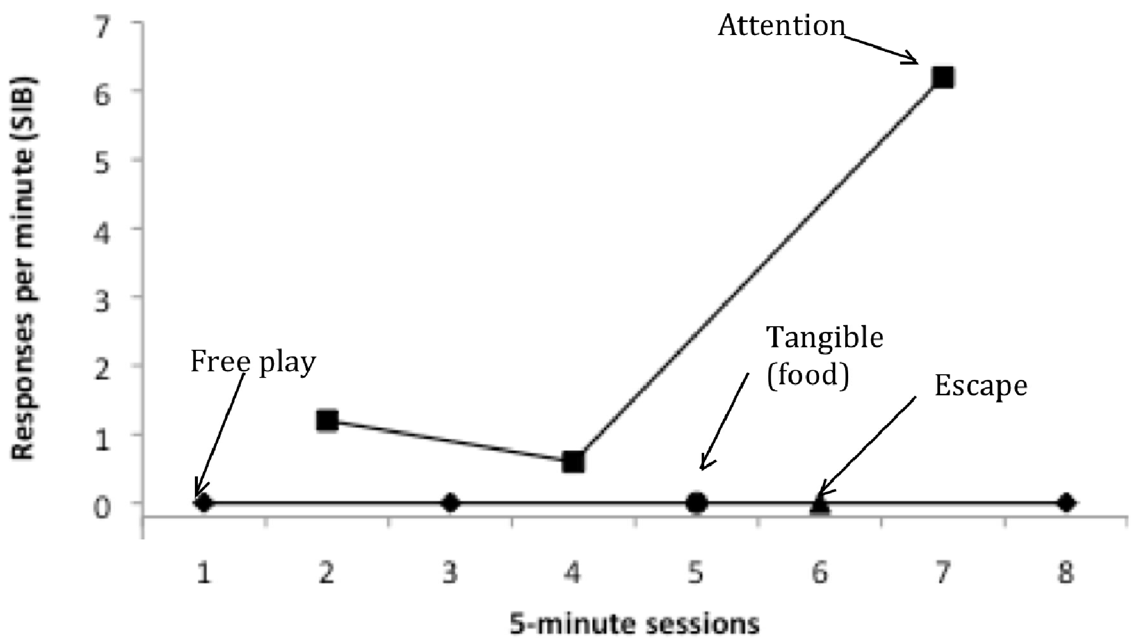


Figure 6.

Rates of vocalizations during functional analysis sessions with Alyssa.

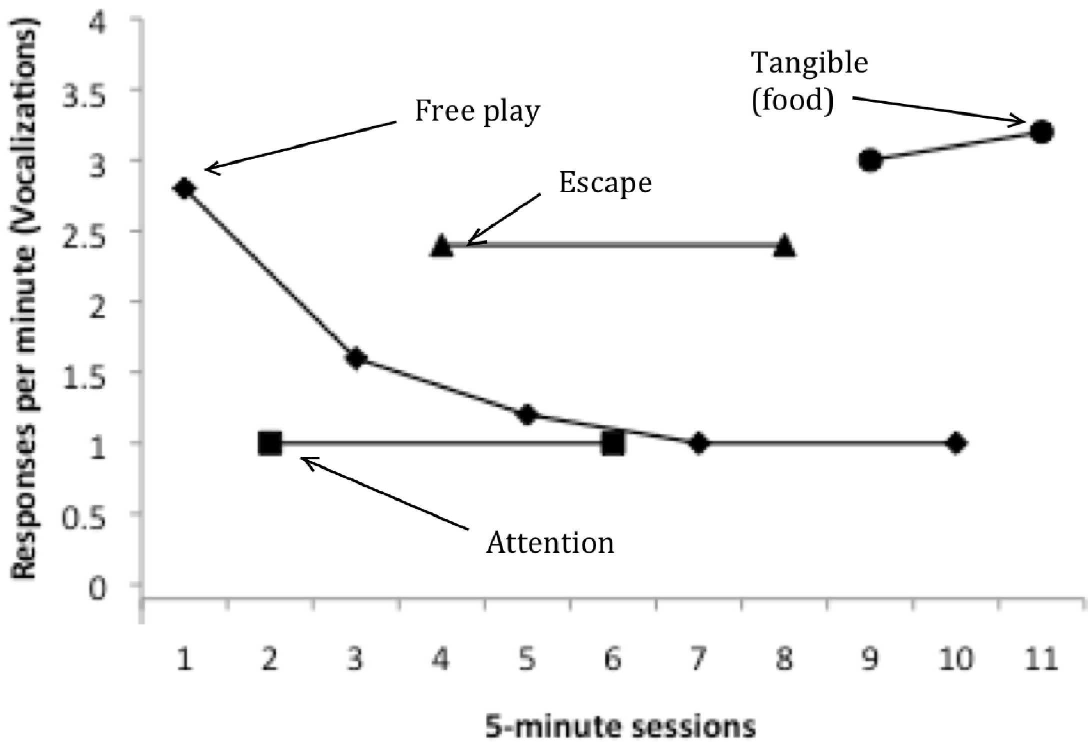


Figure 7.

Rates of grabbing during functional analysis sessions with Rose

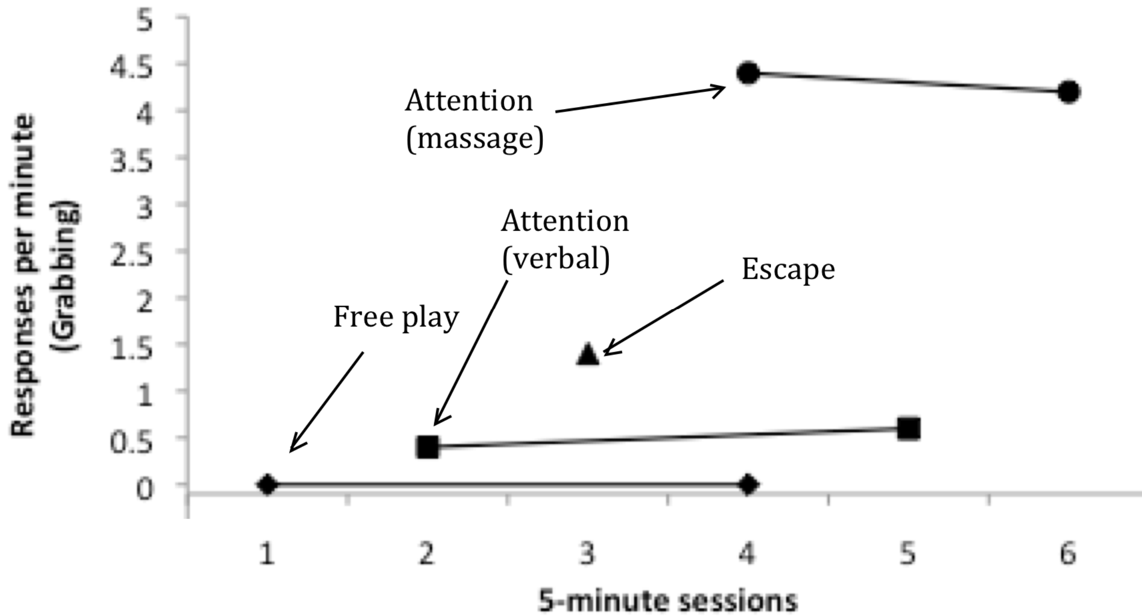


Figure 8.

Rates of eye contact and switch pressing during functional communication training sessions with Ella.

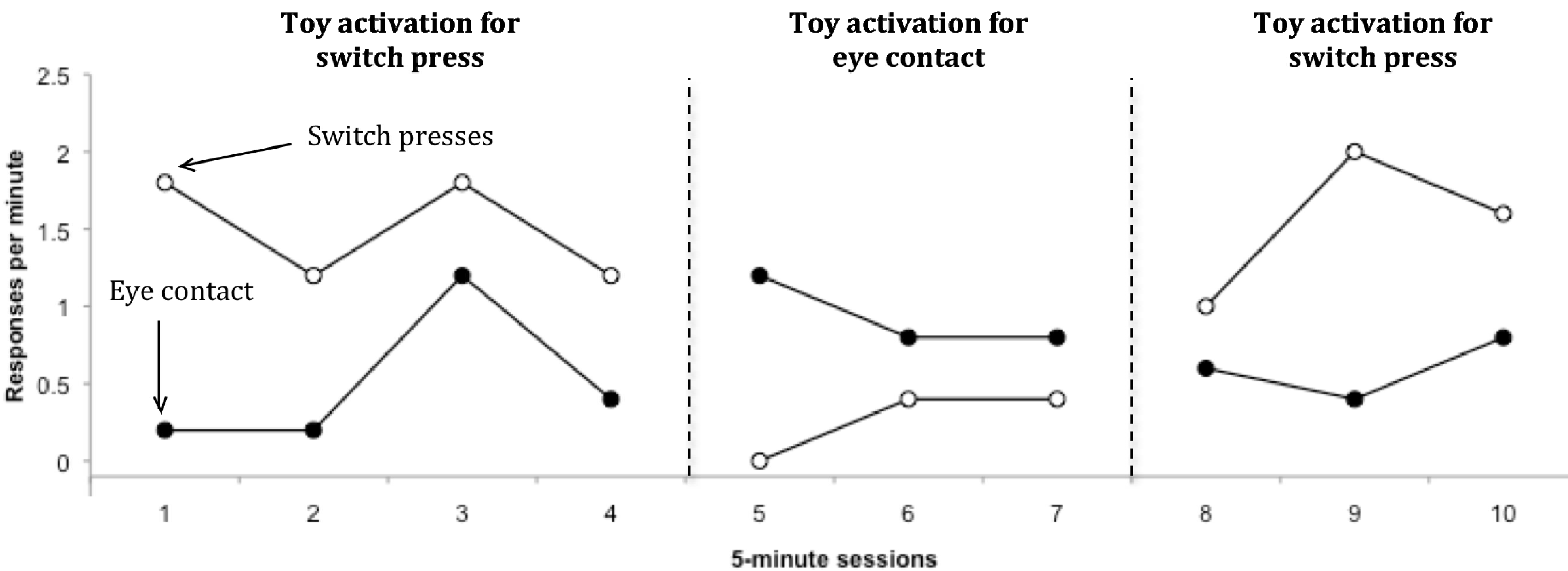


Figure 9.

Rates of vocalizations and switch pressing during functional communication training sessions with Eden.

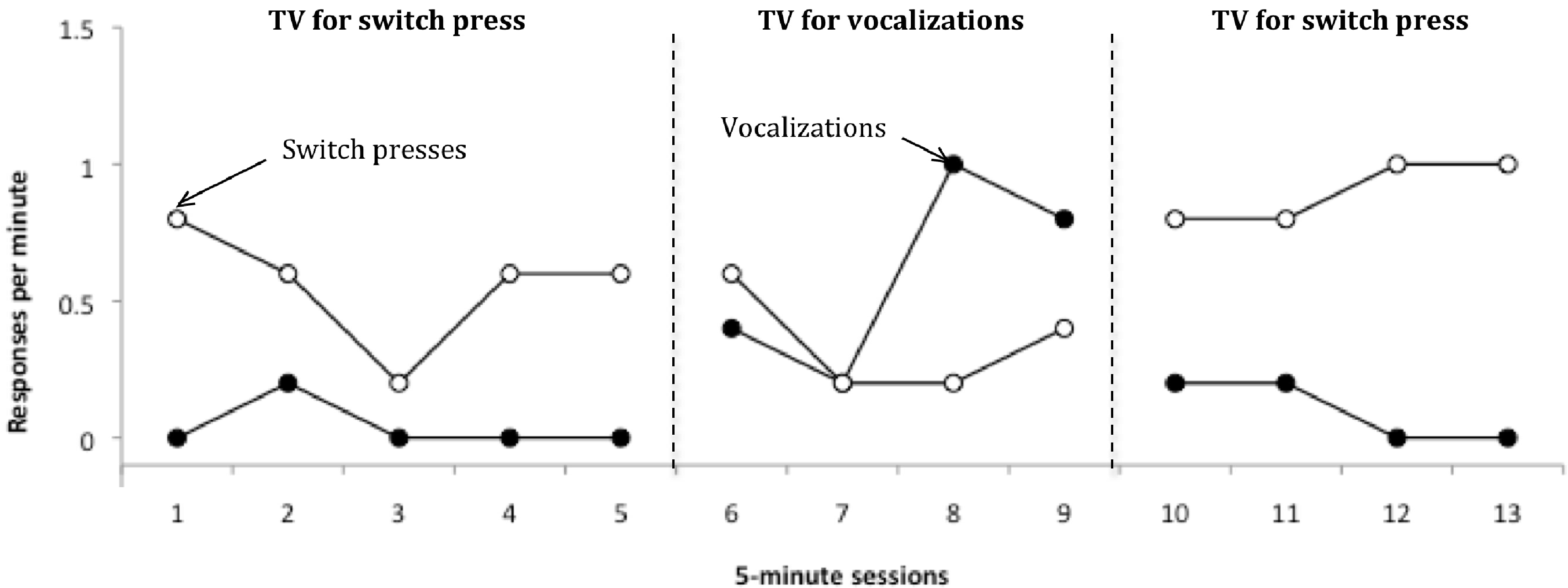


Figure 10.

Rates of whining and switch pressing during functional communication training sessions with Jen.

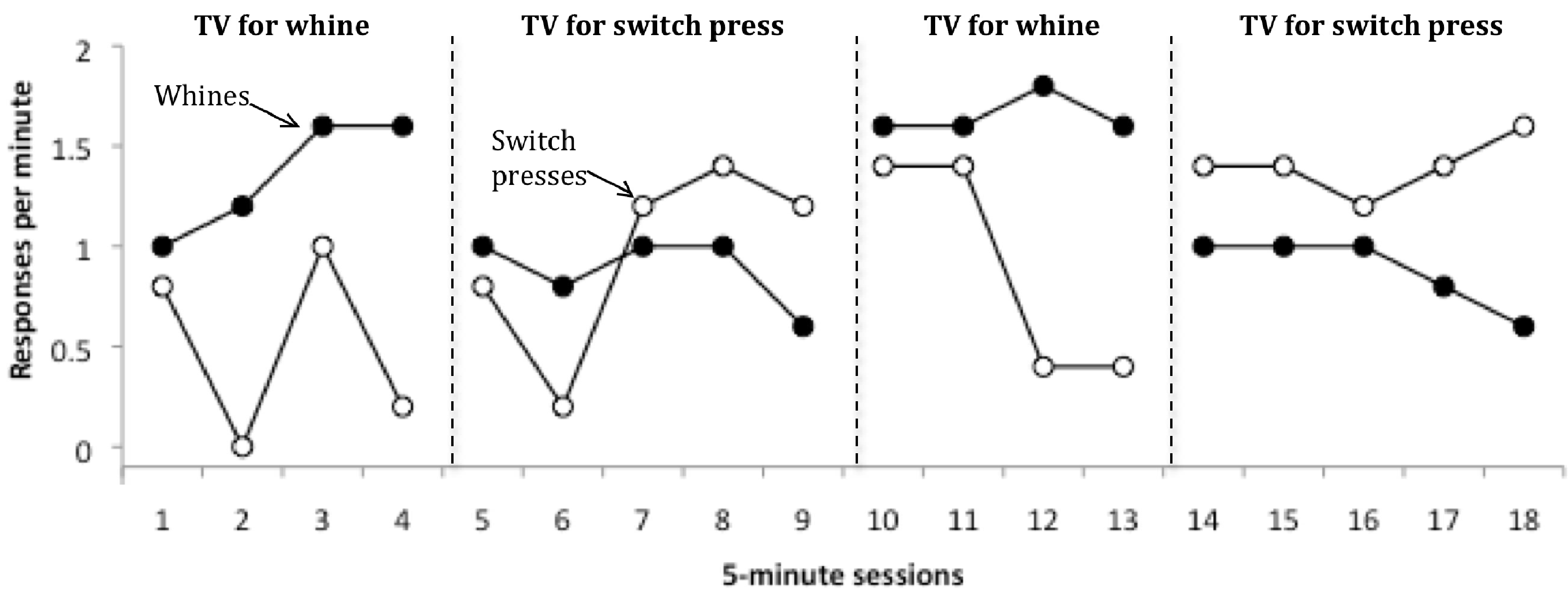


Figure 11.

Rates of hand gestures and switch presses during functional communication training sessions with Veronica.

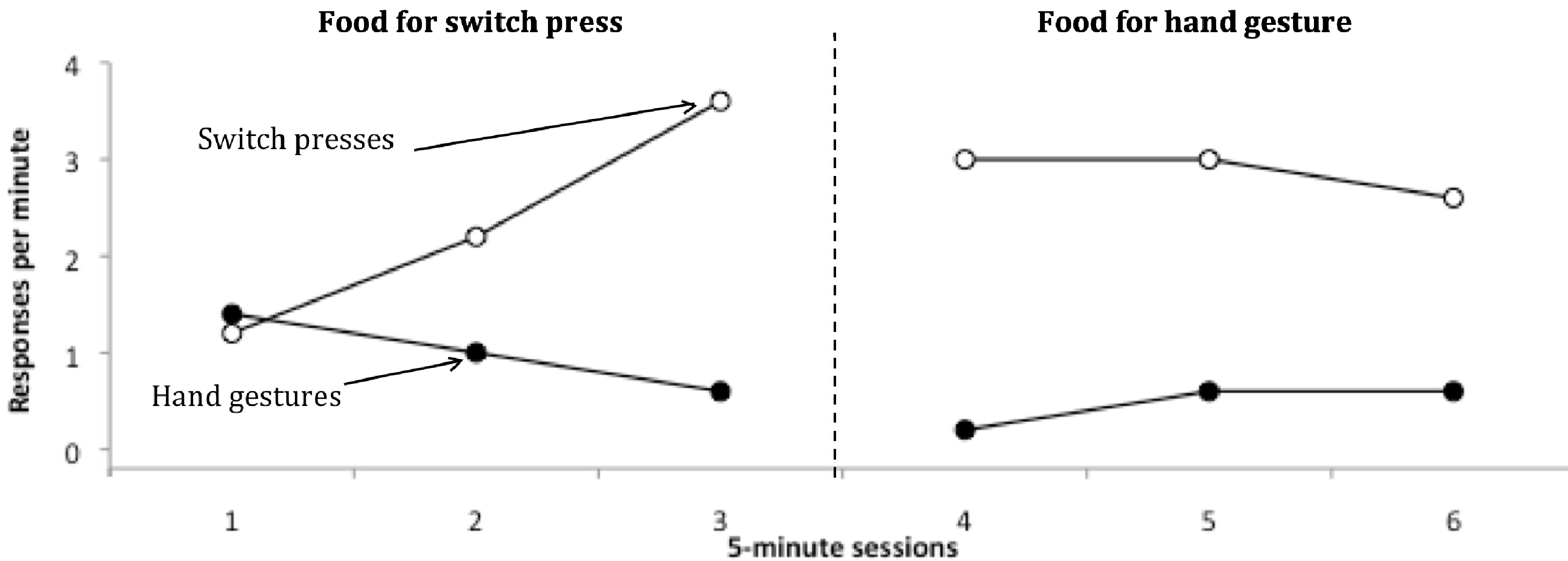


Figure 12.

Rates of self-injurious behavior (SIB) and switch pressing during functional communication training sessions with Tammy.

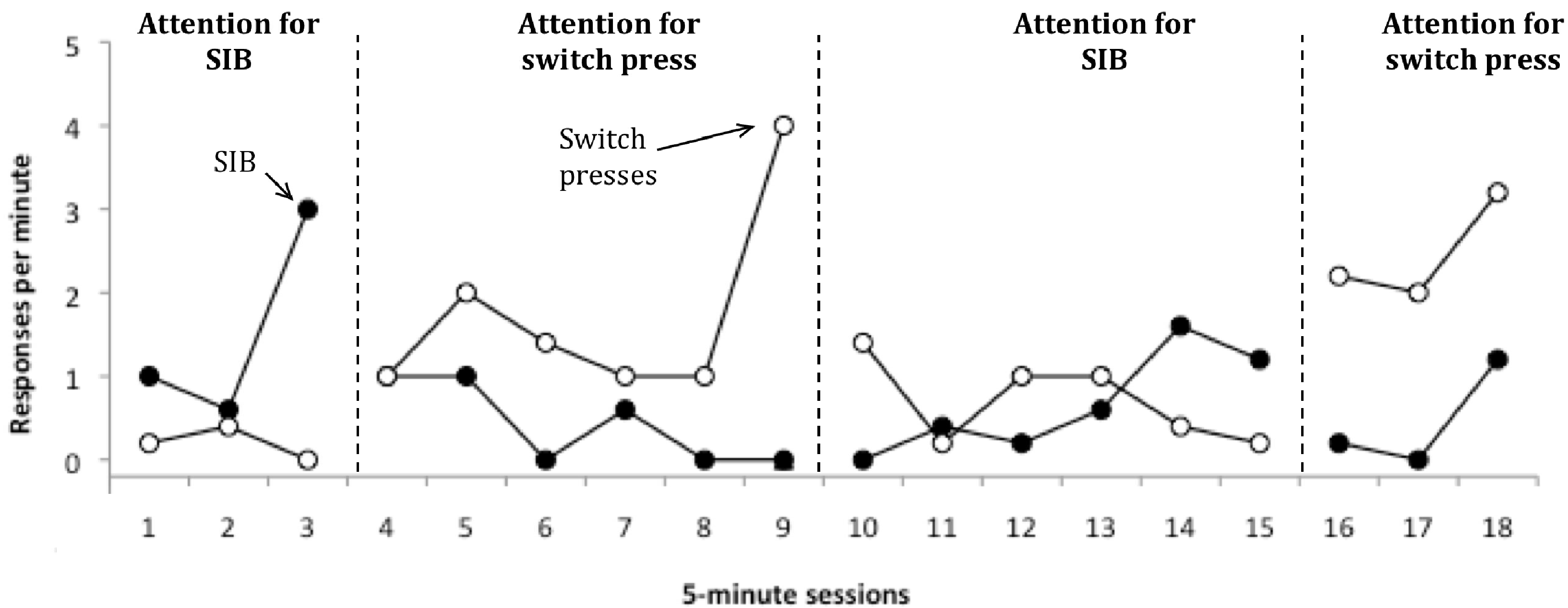


Figure 13.

Rates of vocalizations and switch presses during functional communication training sessions with Alyssa.

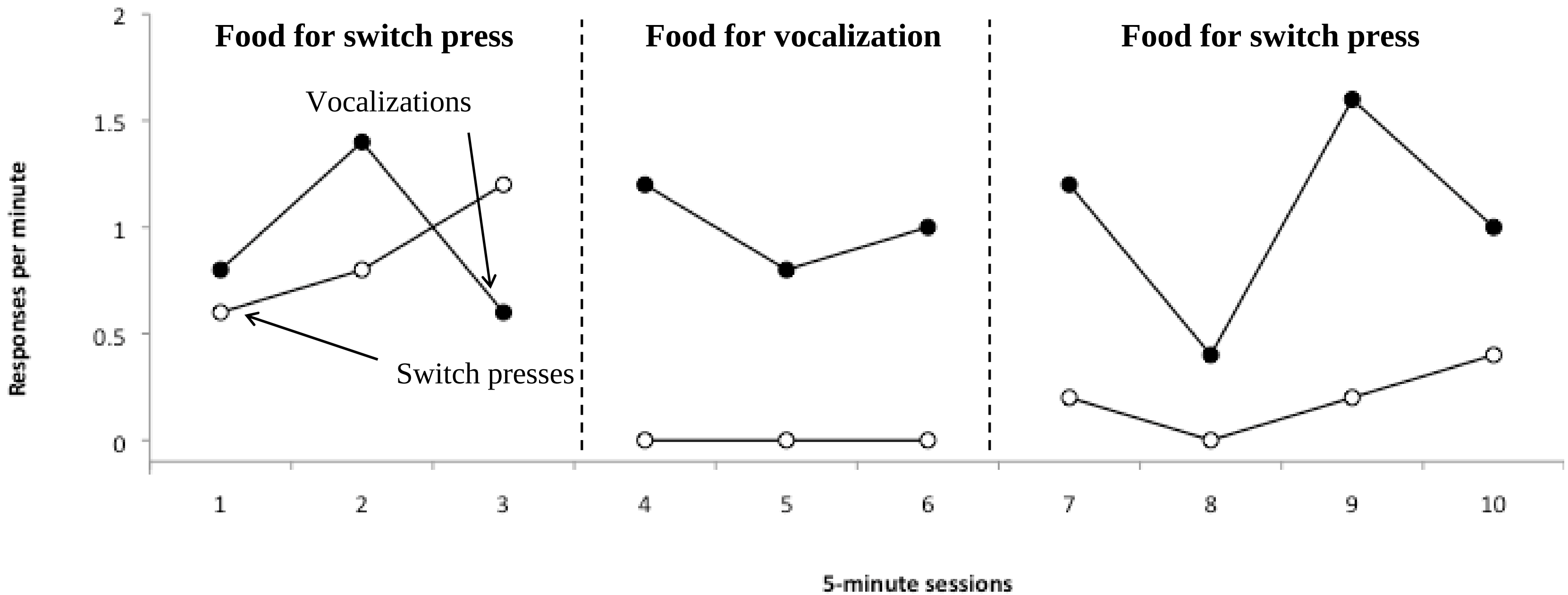
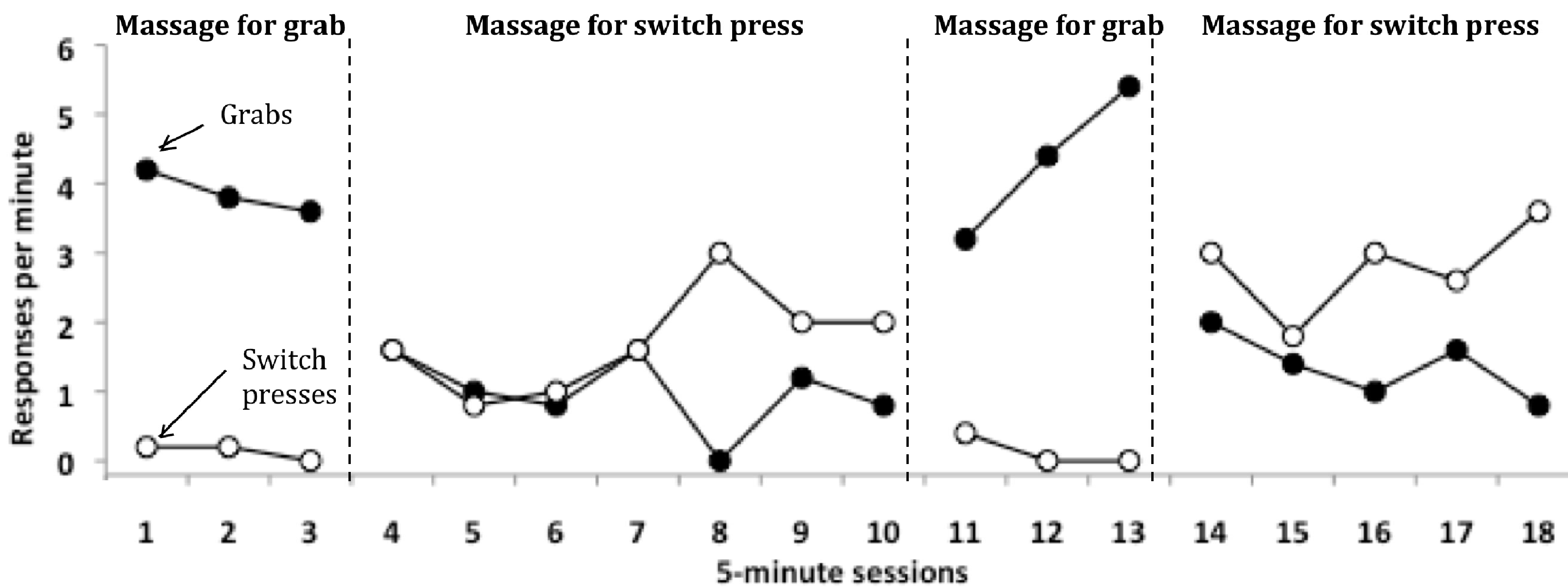


Figure 14.

Rates of grabs and switch presses during functional communication training sessions with Rose.



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