

Communication Intervention to Teach Requesting Through Aided AAC for Two Learners With Rett Syndrome

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Abstract Evidence on effective communication interventions for persons with Rett syndrome is needed to drive the standard of care with this population. This study examined the effectiveness of an intervention package to teach multiple, aided communication requests for two persons with Rett syndrome (ages 27 and 7) through within participant, adapted multiple baseline designs across items/activities. Participants were taught graphic mode requests on speech generating devices, with access methods based on motor ability; one participant responded by pressing a touch screen, and one participant responded by fixed eye-gaze. Results are discussed in relation to the effectiveness of the intervention packages on increasing the accuracy of independent request selection responses emitted and the number of sessions required to reach an *a priori* performance criterion for both participants. Difficulties during initial prompting and during prompt fading with the eye-gaze response are considered. The findings suggest implications related to emerging evidence on the intervention methods to teach requesting skills to this population, and future research directions for communication intervention options for persons with severe communication impairment and limited motor repertoires.

Keywords Rett syndrome · Communication intervention · Requesting · AAC · Eye-gaze

Ellaway and Christodoulou (2001) reported that Rett syndrome (RS), a neurodevelopmental disability linked to the X chromosome that often leads to profound and multiple disability, impacts approximately 1 in 10,000 newborn females. Typically, a diagnosis of RS is made in early childhood, as children with RS display regression/deterioration in a number of developmental areas during four commonly recognized

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stages that involve head growth, motor, communication, and cognitive functioning. Regression continues into later childhood with the majority of persons with RS eventually having significant communication, motor, and possible cognitive impairment (Hagberg and Witt-Engerstrom 1986; Rett Syndrome Diagnostic Criteria Working Group 1988; Hagberg 2002). Many girls lose the ability to walk, talk, and use their hands functionally (Sigafoos et al. 2009, p. 305). Given that the disabilities in persons with RS are severe and multiple, it is not surprising that parents of children with RS prioritize communication among their top areas of concern (Urbanowicz, Leonard, Girdler, Ciccone, and Downs 2014).

Much of the literature describing communication skills in RS is based on both naturalistic and structured observations (see Sigafoos et al. 2011, for a review). This focus is likely the result of the difficulty that persons with RS have participating in the standardized assessment protocols commonly employed in assessing communicative comprehension and production (Demeter 2000). Sigafoos and colleagues (2011) systematically examined the “potential communicative acts” of 41 individuals with RS in a scoping review of eight studies that used behavioral observations to determine whether idiosyncratic responses produced by individuals with RS served a communication function. Half of the investigations in their sample utilized standardized assessments and structured interviews in addition to the direct observations as their assessment protocol, although the validity and reliability of standardized assessment protocols have not yet been well established for persons with RS. Sigafoos and colleagues (2011) concluded that based on the available evidence (Baptista, Mercadante, Macedo, and Schwartzman 2006; Brady and Halle 1997; Sandberg, Ehlers, Hagberg, and Gillberg 2000; Hetzroni & Rubin, 2006; Ryan et al. 2004; Sigafoos et al. 2000; Woodyatt and Ozanne 1997), individuals with RS emit a range of behaviors (e.g., shifting eye-gaze, vocalizations, stereotyped hand movements, facial expressions) that may be intended for a communicative partner. Alternatively, rather than being intended for a communicative partner these emissions may not be intentional. For example, emissions by persons with RS may be emitted as, (a) a response to changes in environmental consequences, (b) a response to changes in internal physiological states, or (c) as a communicative act intended for the benefit of a listener.

Researchers have provided a variety of empirically supported criteria for differentiating intentional communication from non-intentional behavior emissions (e.g. see Wetherby and Prizant 1989; Reichle and Brady 2012). For example, consider a child looking at a bowl of food that is out of reach. To determine whether the fixed gaze is intentional communication different features of the child’s behavior can be examined, including the degree to which the child; (a) shifts his/her gaze between the food and a caregiver, (b) persists at the eye contact until they are given a bite of food, and (c) uses different forms of behavior to request the food in addition to the fixed gaze (Wetherby and Prutting 1984; Reichle and Brady 2012). Parent reports have described a number of behaviors used by individuals with RS to express intentional communication acts that have included, eye-gaze, eye-pointing, facial expressions, vocalizations, and challenging behavior (Stephensen & Dowrick, 2000; Brady and Halle 1997; Urbanowicz et al. 2014). Urbanowicz and colleagues (2014), interviewed 16 families who reported that their child with RS engaged in multiple modalities of idiosyncratic behaviors to express a variety of communicative functions that included; (a) expressing discomfort and happiness, (b) making choices, (c) requesting items, (d) requesting activities, and (e)

obtaining/maintaining attention. Several studies have examined the communication function of the behaviors emitted by children with RS using parent interview and structured observation methodologies (Brady and Halle 1997; Woodyatt and Ozanne 1992; 1993; 1994; 1997; Baptista et al. 2006; Sigafos et al. 2000; Julien, Parker-McGowan, Byiers, and Reichle 2014) to determine whether well defined idiosyncratic behaviors occurred more or less in specific environmental contexts (e.g., how often a person made eye contact when there was food present and when there was not).

Sigafos and colleagues (2000) examined the variability of several forms of idiosyncratic behavioral emissions across different environmental contexts among three participants with RS to assess whether the emissions served as communicative acts. These investigators observed that the frequency of idiosyncratic behavior emission forms varied during high and low social interaction conditions for all of the participants. The frequency of idiosyncratic behavioral emissions also varied as a function of several communicative contexts (e.g. care-providers offered greetings, conversations, requests, protests, and 'more' bites of food). Due to the high levels of variability, investigators were unable to determine many patterns to the idiosyncratic behavioral emissions across environmental conditions, thus the study did not provide evidence that these responses were communicative acts. However, the authors reported that multiple care-providers interpreted the idiosyncratic behavioral emissions of the participant they worked with as communicative at a high level of agreement, suggesting the potential of idiosyncratic behavioral emissions to be recognized and responded to as potential communicative acts.

Empirical examinations of receptive communication (comprehension) among persons with RS have been very limited. Some parents have reported that their children with RS can understand words or short sentences (von Tetzchner 1997) and may follow simple communication requests (Demeter 2000). However, in the bulk of these reports (with respect to both expressive and receptive communication skills) there has been no empirical validation. Baptista and colleagues (2006) examined communicative intentionality among seven participants with RS during three simple receptive tasks by tracking the amount of eye-gaze directed at correct pictures during a spoken task ("look at the picture"), an identity-matching task ("look at the one that is the same"), and a categorization task ("look at the one that is similar"). They found that only one of the participants responded to all of the tasks correctly by fixing her eye-gaze on the correct items, one participant did not respond correctly to any of the tasks, and the remaining participants were only able to respond to the spoken instructional task.

As a result of the limited knowledge about communication skills in persons with RS, it is not surprising that there is a strikingly small amount of experimental evidence addressing effective intervention strategies to teach and support communication skills in this population. In a systematic review of the literature, Sigafos and colleagues (2009) identified nine studies that implemented intervention protocols to teach communication skills to persons with RS. Eighty-four percent of the combined participants' data extracted from this review experienced improvement in communication. However, only one of these studies (Van Acker and Grant 1995) also met the researchers' criteria for demonstrating certainty of evidence (when a study had a controlled experimental design, demonstrated effectiveness of the intervention, and demonstrated sufficient methodological detail and rigor to permit replication). From this review, the identified gaps in the RS communication literature were an important need for quality information

describing replicable intervention procedures along with descriptions of nonverbal communication response forms, and communicational functions in this population.

Byiers, Dimian, and Symons (2014) implemented experimental functional analyses to demonstrate the social function associated with specific idiosyncratic behavior among three individuals with RS. Results indicated that the participants engaged in an idiosyncratic behavior more during individualized conditions designed to enable access to different social reinforcers including head massage, attention, and TV. Subsequently, the researchers implemented functional communication training to shape the participants' idiosyncratic behavior into a requesting response, which all participants acquired, through voice-output switch activation to gain access to the social reinforcer identified from the functional analysis. The study demonstrated that for the participants, it was likely that the idiosyncratic behavior had (1) been serving communicative purposes and (2) were easily shaped into a more conventional communication form (switch activation) during functional communication training.

Unfortunately, relying on idiosyncratic behaviors as a sole communicative means poses potential limitations. One challenge is that many idiosyncratic behaviors emitted as intentional communicative acts may also represent stereotypes of behavior commonly associated with RS (e.g., hand-ringing, persistent eye-gaze, and hyperventilation). Consequently, caregivers may correctly or incorrectly interpret stereotyped behavior as intentionally communicative. Additionally, stereotypic behavior may interfere with performance during discrimination tasks requiring sustained attention specific motoric responses. This, in turn, could make more difficult the task of shaping these behaviors teaching a more conventional communicative form while teaching the discriminative use of symbols (Fabio, Giannatiempo, Antonietti, and Budden 2009). Finally, relying on idiosyncratic behaviors as a primary communicative means is highly context-dependent, interpretation of meaning rely heavily on the caregiver's familiarity with the individual and the presence of coinciding visual or spoken stimuli. Given the inherent limiting factors of relying on idiosyncratic behaviors to communicate, teaching more conventional communication forms to individuals with RS requires investigation.

Among early communicative functions acquired, requesting, which falls within the definition of a mand (Skinner 1957), is an important communication function that builds opportunities for environmental access, choice making, and provides a base on which to build increasingly complex communication skills (Cannella, O'Reilly, and Lancioni 2005). Sigafos and Couzens (1995) taught a participant with RS to produce a generalized request using low technology eye-gaze. Byiers and colleagues (2014) successfully taught 3 participants to request a functional reinforcer by touching a microswitch with corresponding voice output. Both of these studies taught simple discriminations in that only one request was targeted for each participant and thus, did not require the participant to discriminate between multiple response options.

Stasolla, De Pace, Di Leone, and Albano (2014) compared teaching a low technology graphic mode modification of the Picture Exchange Communication System (PECS, Bondy and Frost 1994) request with teaching a high technology SGD request with three participants. Their successful implementation involved elements of conditional discrimination in which the participant had to discriminate between a distracter (S-, an incorrect and neutral item) and a target request (S+, the preferred item). This study holds promise for the likelihood of teaching individuals with RS conditionally discriminated, or more complex, communication. However, a measure of accuracy was

not reported, making it difficult to ascertain the degree to which the participants were accurately discriminating between the S^+ and S^- options.

A possible solution to providing more conventional communication forms while compensating for the impaired motor function among many individuals with RS is the use of Alternative and Augmentative Communication strategies (AAC). These nonverbal communication forms include unaided responses (e.g., signs, gestures, vocalizations) and technology-aided responses encompassing both low technology (e.g., pictures, 2D symbols, 3D objects) and high technology (e.g., speech-generating devices (SGD), computer-based interfaces) (Beukelman and Mirenda 2013). Unaided communication forms are often too difficult for many individuals with RS due to the associated severe motor deterioration and the high likelihood of little to no persevered hand function coinciding with the higher motor effort required to form gestures or signs. An important aspect of intervention planning with aided communication systems involves choosing a feasible response form for which the individual has sufficient motor control to produce (Beukelman and Mirenda 2013). Therefore, persons with RS may benefit most from a number of aided AAC options.

There have been few experimental studies investigating communication interventions using aided AAC strategies among individuals with RS. Communication forms that have been examined experimentally range from low technology, including: (a) eye-gaze with a choice board (Sigafoos & Couzens, 1991), (b) picture exchange communication systems (PECS, Stasolla et al. 2014), and (c) micro switches (Byiers et al. 2014) to high technology, including: (a) computer touch-screen (Van Acker and Grant 1995) and (b) voice output device (Stasolla et al. 2014). Several investigators have utilized eye-gaze as a communication means with persons having RS. Sigafoos and Couzens (1995) examined low technology eye-gaze, in which the participant signaled to her listener by gazing at 3D items or representations of items on a board. A low technology eye-gaze response may be limiting in that it is context dependent (the caregiver must interpret the gaze of the individual). Therefore, for this communicative strategy it is important that symbols be relatively far apart to ensure an accurate interpretation by a listener.

One somewhat recent advance with respect to aided AAC options involves the use high technology eye-gaze interface with speech generating devices (SGDs) (Higginbotham, Shane, Russell, and Caves 2007). An SGD utilizing eye-gaze interface measures duration that an individual fixes his or her gaze on a symbol (referred to as “dwell”). Once the preset dwell criterion is met, the device produces speech output (Fager, Bardach, Russell, and Higginbotham 2012). Eye-gaze technology has been a successful AAC communication option for adults with amyotrophic lateral sclerosis (ALS) and traumatic brain injury (TBI), making it a promising possibility for individuals with developmental and multiple disabilities who have limited motor control (Ball et al. 2010). However, the SGD eye-gaze literature for persons with RS and related disabilities is sparse in refereed journals (Beukelman et al 2007). To date, there has not been an experimental communication intervention study examining the teaching of SGD with eye-gaze in RS, thus providing limited empirical support for the use of this method.

Overall, intervention research is needed to support and drive evidence-based communication intervention for persons with RS. In particular, due to the confounding effects of motor deterioration on communication response effort, aided AAC

interventions need to continue to be investigated for this population. The purpose of this study was to extend the current literature in RS and communication intervention by examining the effects of an intervention package that included response and stimulus prompting and prompt fading methods on accuracy across multiple, discriminated requests for two participants with RS. The second purpose of this study was to examine the intervention package for requesting with two forms of aided AAC communication responses on SGDs that with one participant involved the utilization of a fine motor response (pressing) and with a second participant, who had impaired hand function, involved the utilization of an ocular response (eye-gaze).

Method

Participants and Setting

Two females, Annie and Rory, participated in this IRB approved study following parental request for communication intervention after participation in a previous study involving functional analysis of challenging behavior. Both participants met the following inclusion criteria: (a) a diagnosis of typical or atypical RS, (b) did not use gestures, vocalizations, low or high technology AAC communication strategies consistently, (c) displayed idiosyncratic responses that allowed researchers to assess preference for items and/or activities (i.e., looked at and/or reached for items, protested removal of items/activities).

Annie Annie, a 27-year-old woman, had a clinical diagnosis of atypical RS due to a late onset of regressive symptoms; she had not been tested for a genetic phenotype of RS. She lived in her family home and attended a day-program for adults with intellectual and developmental disorders. Annie experienced tonic clonic seizures several times per day that were poorly controlled by medication. She was ambulatory, although she required physical assistance with standing, sitting, and navigating stairs or uneven areas. She had partially preserved hand function that included the ability to press, grasp, hold, pick up, and set down objects.

Parental interview and interventionist observations conducted in her home yielded information that; (a) Annie did not produce any spoken words, (b) her vocalizations included laughing and occasional moaning sounds, (c) she did not use any conventional communication method (i.e. natural gestures, vocalizations, or graphic symbols), (d) Annie's idiosyncratic "potential requests" involved reaching for items and slapping her upper thigh, which were interpreted by her caregivers as Annie wanting access to items or activities. A functional analysis was conducted in a previous study with Annie (Byiers et al. 2014) and the results supported the hypothesis that these responses were reinforced by access to tangible items.

Rory Rory was 7-year-old girl who had a clinical diagnosis of typical RS, and she tested with a genetic mutation of MECP2 (R168X). She resided with her family and attended first grade in a self-contained public school special education program with access to selected activities in the general education with the support of a paraprofessional. She had limited ambulatory movement (if she was assisted to stand, she could

walk with close supervision) and she required supportive assistance with sitting positions (left without support she occasionally slouched downwards and required repositioning). She spent the majority of her school day in a wheelchair, and seated in a chair, on a couch, or in a wheelchair at home.

Parental interview and interventionist observations in her home yielded information that Rory; (a) did not produce any spoken words, (b) produced vocalizations that included laughing, crying, and occasional sounds similar to 'ma,' 'mm,' and 'no', (c) did not use any conventional method of communication, and (d) did not communicate her wants and needs through the graphic mode.

Rory's idiosyncratic requesting responses at the beginning of this investigation involved fixed eye-gaze—or eye pointing—by looking intently at items or between the items and her caregivers to communicate requests in her home, school, and community. For example, when Rory was being fed, she would often fix her eyes on a bowl of food or on her parent. Rory also engaged in tantruming behavior that consisted of crying, screaming, whining, or flopping her body over when items, such as the TV, were removed or unavailable.

Prior to and during the study, Rory had access to a Tobii™ SGD with eye-gaze at school and home. Her device was set up with a variety of different comments, requests, literacy, and phrase-building options across multiple pages, with button arrays ranging from approximately 10-30 buttons on each page. Although she had this device for approximately two years, her use of it was highly inconsistent, perhaps due to the difficulty of the discrimination with between the responses and with navigation. Her parents reported they often needed to interpret what her communication responses on the SGD meant. When she did attempt to use the device, her response latency was extremely long and the accuracy of her communication selection was unclear. Additionally, her parents reported that they frequently helped Rory navigate to a new page.

Rory's requesting with the device was tested prior to the onset of the study with a consequent-based functional analysis, which indicated she did not reliably request with eye-gaze on the SGD during programmed consequences of contingent attention from her parent or access to the TV or food; she did not ask for any of these activities or items and was as likely to engage in tantrum behavior during the tested conditions. Her parents reported that they wanted her to have the ability to request through the graphic mode SGD more reliably to avoid her frustration and challenging behavior.

Setting Sessions were conducted in each participant's home with a parent in close proximity who assisted with procedures and physical positioning throughout all of the sessions. Participants were positioned close to the preferred items/activities, in ways that were similar to how they usually spent their leisure time.

Annie was positioned seated on the carpeted floor, with her parent and the interventionist also seated on the floor. This position was selected because she often sat on the floor during leisure time and crawled on her knees to access items in different locations in the living room area. Annie required assistance to stand from this position and if she attempted to stand, her parent assisted her.

Rory was positioned on the couch or in her wheelchair in the family living room area near her mother. At times, she slouched to one side and had to be propped up by her

mother to return to a seated position. Beginning at intervention session 44, Rory was positioned in a new wheel chair in the same area of the family room.

During baseline and intervention sessions, one of the authors served as interventionist, parent coach, video recorder, and data collector. Parents participated in the intervention sessions as well. For example, Annie's mother sang to her during the 'Mom' sessions and looked through a book with her during the 'book' sessions. Rory's mother offered the snacks in the 'food' sessions, and played with Rory by talking to her and manipulating toys with her during the 'doll' sessions.

Materials

Both participants used a SGD with synthesized speech output upon response selection. The three requests taught were each located on a separate page of the participant's SGDs; the layout of each page included a target symbol (representing the request) and a distracter symbol, (representing a neutral item). The purpose of the distracter symbol, a putatively item of little or no interest to the learner (e.g., towel), was to teach and measure discrimination of the target request. The target symbols were represented as photographs for Annie and line drawings for Rory; the distracter symbols were line drawings for both participants (see Fig. 1 for an example). Navigation between pages was not required during the implementation of this investigation; the interventionist opened the relevant request page prior to the session.

Annie's Materials A Vmax™ by Dynavox™ with a touch sensitive screen served as the SGD used by Annie. It was programmed with three pages, each page containing four symbols; (1) the target request—a photograph of the item or her mother—was placed in a differing location on each page, (beads, book, and Mom), (2) a distracter symbol that was never targeted as a request and was scored as an incorrect selection was placed in a differing location on each page, (towel, tissue, and pen, respectively), and (3) each page had two additional distracters that consisted of white (blank) buttons that did not have an symbol nor produce voice output when pressed. These were locations for additional symbols to be placed once the initial symbols were mastered.

Rory's Materials Rory used a Tobii™ SGD that contained a C12 computer with a 12in screen, and a C-series™ eye-gaze bar that used a safe infrared-laser to measure the amount of dwell time (500 ms) that Rory fixed her eye-gaze on an symbol to activate synthesized speech output. For the purposes of this investigation, Rory's targeted pages

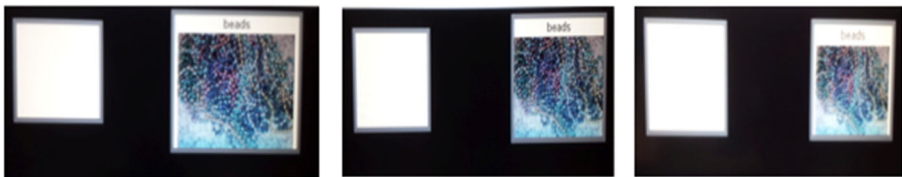


Fig. 1 Stimulus fading procedure demonstrated on one of the requests on the SGD for Annie, fading from larger symbol (*stimulus prompt*), to partially faded symbol (*partial stimulus fade*), to same size symbol (*full stimulus fade*). Not included in this figure is the distracter symbol, which was located in lower left-hand corner and second non-reactive, *white button*, which was located in the lower right-hand corner

were set as 2-button displays, with one target request in varying positions per page, (TV, food, doll) and one distracter item in varying positions per page, (pen, tape, paper clip, respectively). Rory's pages were limited to the two symbols and were not set with any non-reactive white buttons.

Response Definitions, Data Collection, and Inter-Observer Agreement

For both learners the dependent variable was independent accurate selection of the target request (pressing the symbol for Annie and fixed eye-gaze selection of the symbol for Rory). Accuracy was defined as opportunities with correct requests and was measured as the percentage of opportunities with correct target requests divided by the total opportunities in a session. For Annie, accuracy was measured as independent and correct responses divided by total opportunities within a session. For Rory, accuracy was measured as correct responses divided by total opportunities within a session. During fading phases, stimulus (Annie and Rory) or safe laser-prompted responses (Rory) were no longer counted as accurate responses in a trial. Errors for both participants were defined as selection of the distracter symbol or non-responding with the press (Annie) or eye-gaze (Rory) response within the trial.

Inter-Observer Agreement All sessions were videotaped and dependent measures were coded from videos by one of the authors. Inter-observer agreement data (IOA) were collected on 22 % of randomly selected sessions, distributed across baseline and intervention phases. IOA was calculated as the total number of opportunities agreed upon by the two observers divided by the number of opportunities observed and multiplied by 100. IOA averaged 94 % for Anne (range, 88-100 %) and 92 % for Rory (range, 86-100%).

Procedural Integrity Twenty percent of videotaped baseline and intervention sessions were randomly selected for procedural integrity recording by two different observers. A 13-item checklist of the task analyzed steps for the procedure was used to record procedural integrity data (percentage of occurrence of the items on the checklist) with items representing procedures for: general session protocol, prompt-hierarchy procedures, prompt-fading procedures, and error correction procedures. Procedural integrity averaged 96 % for Annie (range 85 to 100 %). Procedural integrity during baseline and throughout intervention averaged 93 % for Rory (range 77 to 100 %).

Experimental Design

Two within participant, adapted concurrent multiple baseline designs (Gast and Ledford 2014) across requests were used to experimentally analyze the effects of the request-intervention packages with response prompting and stimulus prompting on the accuracy of three separate requests per participant. The baseline procedures consisted of the preferred item located within view but out of the participant's reach and access to the SGD. Subsequent to baseline, request-intervention was introduced for both participants in a staggered fashion across preferred items/activities. Performance criterion for each participant was defined as 80 % or better accuracy for three consecutive sessions.

Additionally, adjacent phases within requests were compared for efficacy of prompting techniques for Rory.

Data Analysis

Within each intervention phase and across items in the multiple baseline design, visual analysis was used to examine the effects of the intervention package on the requesting response accuracy, the progress towards performance criterion, and the effectiveness of the fading procedures.

Pre-Intervention Assessment

Prior to the onset of the study, information was obtained on the participants' preference for items and activities through parent interviews, structured observation of the participants during leisure time activities, and formal preference assessment (Annie) and informal preference assessment (Rory). Additionally, prior to the onset of the study, Rory's use of the SGD eye-gaze and tantrum behavior was compared to determine the relative frequency of each response during programmed environmental consequences.

Pre-Intervention Preference Assessment With Annie Prior to the onset of the study, a multiple stimulus without replacement preference assessment (DeLeon and Iwata 1996) was conducted by placing multiple items including books and beads in random order and instructing Annie to "Take what you want." Her selections were recorded across five sessions and the items 'beads' and 'book' were among the first and second choices she made across all opportunities.

Pre-Intervention Preference Assessment With Rory Rory's preference was assessed by informal pre-intervention observation of items/activities that Rory typically engaged in per parent report. Consistent with parent report, during these observations Rory was observed crying, tantruming, and fixing her gaze on these items when they were in sight. Her idiosyncratic behaviors were interpreted as requests for these items or activities. Per the pre-assessment observation, 'TV', 'food,' and 'doll' were selected as target requests.

Rory Pre-Intervention Functional Analysis A consequent-based, extended functional analysis was conducted using a multi-element design to experimentally test and determine the influence of social consequences on tantruming and SGD use. The following conditions were arranged: (a) tangible (TV being turned on for 30 sec), (b) attention (parent giving contingent attention for 30 sec), and (c) free play (TV on and parent giving frequent attention, no programmed consequence for tantruming or SGD use). During the FA, Rory's SGD was positioned next to her and she was given immediate access to the programmed consequence contingent on eye-gaze activation of the device (regardless of accuracy) or engagement in tantrum behavior. The results of the FA indicated a high degree of overlap between conditions, as well as overlap between percentage of intervals with eye-gaze and tantrum behavior. There was no discernable pattern to her responding with either of these responses, suggesting that Rory did not reliably use her SGD to communicate in these situations. The results

further suggested that Rory was at least as likely to defer to tantrum behavior during these programmed consequences (the functional analysis data are available from the first author upon request).

General Procedures

An overview of the session phases and procedures for both participants is available in Table 1. Treatment intensity parameters (Warren, Fey, and Yoder 2007) of the intervention were as follows: (a) sessions consisted of approximately 8 opportunities (range 5-10), (b) sessions lasted between 5 and 15min, (c) there were between three and six sessions conducted per day, (d) with 1-2 days of sessions per week, and (e) occurred over approximately 6 months for Annie and 8 months for Rory.

At the beginning of a session, the interventionist provided access to a targeted item or activity for 30 sec (e.g., she was given the beads, or the TV was turned on). The interventionist then removed access to the item, but kept it close within sight and said

Table 1 Summary of the progression of baseline and intervention procedures for Annie and Rory

Participant and request	Order of procedures	Description
Annie, 'beads'	Baseline	General baseline procedures
	Phase 1: R&SP	Response and stimulus prompting
	Phase 2: F. Fade	Full stimulus fading
	Phase 3: SP	Return to stimulus prompting
	Phase 4: P. Fade	Partial stimulus fading
	Phase 5: F. Fade	Full stimulus fading
Annie, 'book'	Baseline	General baseline procedures
	Phase 1: R&SP	Response and stimulus prompting
	Phase 2: P. Fade	Partial stimulus fading
	Phase 3: F. Fade	Full stimulus fading
Annie, 'Mom'	Baseline	General baseline procedures
	Phase 1: R&SP	Response and stimulus prompting
	Phase 2: P. Fade	Partial stimulus fading
	Phase 3: F. Fade	Full stimulus fading
Rory, 'TV'	Baseline	General baseline procedures
	Phase 1: RP	Response prompting
	Phase 2: RP+ Size	Response prompting/increase size
	Phase 3: RP+ Dwell	Response prompting/decrease dwell
	Phase 4: LP	Safe laser point prompt
	Phase 5: RP	Response prompting
	Phase 6: LP	Safe laser point prompt
Phase 7: LP Fade	Safe laser point prompt fade	
Rory, 'food'	Baseline	General baseline procedures
	Phase 1: RP	Response prompting
	Phase 2: LP	Safe laser point prompt
	Phase 3: RP	Response prompt
	Phase 4: LP	Safe laser point prompt
	Phase 5: LP Fade	Safe laser point prompt fade
Rory, 'doll'	Baseline	General baseline procedures
	Phase 1: LP	Safe laser point prompt

“Tell me what you want” or a similar variation of this direction. She was given approximately one minute to respond. If no response occurred or the distracter symbol (a putatively neutral item (e.g., towel), to teach and measure discrimination of the target request) was selected, the trial was counted as an error and an error correction procedure was implemented. If the distracter symbol had been selected, the participant was given the distracter item for 30 sec in addition to the error correction procedure. The error correction procedure consisted of resetting the trial and briefly placing a piece of paper over the SGD for 30 sec, then repeating the interventionist direction and using a more intrusive step in the prompt hierarchy. Prompting occurred until the correct request was produced, however, the entire trial was coded as an error. When the participant engaged in the correct response she was given approximately 30 sec of access to the item or activity.

If the participants engaged in any pre-existing idiosyncratic request responses, these responses were placed on extinction from the tangible; the participant was repositioned if necessary, and redirected by the interventionists or the parent stating ‘You need to tell me what you want.’ At times during sessions, Annie engaged in self-injury (slapping her upper high) or attempted to grab or throw the items. When this occurred, the behavior was ignored; she was blocked from grabbing items, and repositioned, if necessary. Occasionally during sessions, Rory engaged in eye pointing (fixing her gaze on the item instead of the SGD) or tantruming behavior (crying, screaming, whining, or flopping her body over). When this occurred, she was redirected by her parent or the interventionist who stated, ‘You need to tell me what you want,’ and repositioned, if necessary.

Baseline

Baseline Sessions With Annie Baseline sessions followed the general procedural protocol except without response prompting or the error correction procedure.

Baseline Sessions With Rory Baseline sessions followed the general procedure protocol except that no prompting or error correction was used (Rory’s parent and interventionist made several comments to increase Rory’s attending such as ‘Perk up,’ or ‘You need to tell us on the Tobii™’ which were not recorded as prompts throughout the study). These comments were used throughout the study if Rory seemed to be falling asleep or fixing her eye-gaze away from the intervention area and device.

Intervention

Response Prompting With Annie Phase 1, following baseline, consisted of response and stimulus prompting (R&SP), during which interventionists used and quickly faded a most-to-least response prompting hierarchy (Gee, Graham, Goetz, Oshima, and Yoshioka 1991). In this study, sessions began with more intrusive prompting and across subsequent opportunities (within a session) response prompts were faded until independent responses occurred. The response prompting hierarchy began with physical guidance of Annie’s hand to the correct symbol during the first opportunities of new requests; partial physical (lightly guiding her hand to the

correct symbol) and then gestural prompting (the interventionist gesturing at the correct symbol) were used in subsequent opportunities. When Annie responded independently with the least intrusive level of prompt, no response prompting was used for all subsequent opportunities for that request unless errors occurred. Opportunities requiring response prompting were not counted as independent responses during any phase of the study. During all opportunities of the R&SP (Phase 1), a stimulus prompt was used that involved the target request symbol being twice the size of the distracter symbol. Stimulus prompting was faded in later intervention phases.

Stimulus Fading of Symbol Size With Annie Systematic fading of stimulus prompts was initiated in Phases 2-5 (beads), Phases 2-3 (book), and Phases 2-3 (Mom). Stimulus fading was initiated after Annie met performance criterion in the R&SP (Phase 1) and maintained accuracy without the use of response prompting. In Phase 2, during the initial stimulus fading effort with the first request (beads), a subtler fade was implemented. This involved first a partial stimulus fade in which the target symbol was decreased in size to 1X larger than the distracter and second a full stimulus fade in which the target symbol was decreased to the same size as the distracter symbol. The second and third requests, 'book' and 'Mom,' were moved directly to partial and then full stimulus fading when accuracy reached criterion in Phase 1, R&SP (see Fig. 1).

Response Prompting With Rory Phase 1, following baseline, consisted of a most-to-least response prompting (RP) hierarchy. For Rory, the prompts ranged from a most intrusive (the interventionist modeling by pressing the symbol to make voice output with a continued point at symbol), to a less intrusive prompt (the interventionist briefly pointing at the symbol), to a least intrusive prompt (the interventionist gesturing towards the SGD). The target request symbol was 2X larger than the distracter symbol and the dwell time that Rory was required to fix her gaze to activate synthesized voice output was set to 500ms. Stimulus prompts were not recorded as a prompt for Rory; the increased size of the target requests in comparison to the distracter symbols was used throughout all study phases and was not faded.

Procedural Changes During Initial Request Intervention With Rory Due to difficulty in achieving accuracy or reaching criterion with RP in Phase 1, additional procedural changes were implemented. In Phase 2, the target symbol was increased from 2X to 4X larger than the distracter symbol. In Phase 3, the dwell time was decreased from 500ms to 420ms. After these modifications were introduced, they were not considered a prompt for Rory; the increased size and decreased dwell time were used throughout the remaining phases of the study and were not faded.

Safe Laser Pointer Prompt With Rory Due to low levels of accuracy in Phases 1-3 with the RP hierarchy and the procedural changes, a safe laser pointer (LP) was used as a prompt by fixing it on the target request symbol until Rory looked at the symbol for the full dwell time required for the device to activate speech. The effect of the laser prompt was compared to the effects of the RP hierarchy for Rory in Phases 4-7 of the TV request.

Stimulus Fading of Safe Laser Pointer Prompt Rory The laser prompt was faded using a time delay procedure (Wolery and Gast 1984) across two requests in Phase 7 (TV) and Phase 5 (Food). The time delay procedure consisted of the interventionist beginning the trial and waiting 30 sec for Rory to engage in the eye-gaze response without the laser prompt. If she did not establish any eye-gaze, then the experimenter used the laser and the response was coded as prompted.

Results

Annie Figure 2 displays Annie’s accuracy of requests across baseline, R&SP intervention, and stimulus fading phases. The three panels depict accuracy during request intervention staggered across the three requests. The top panel

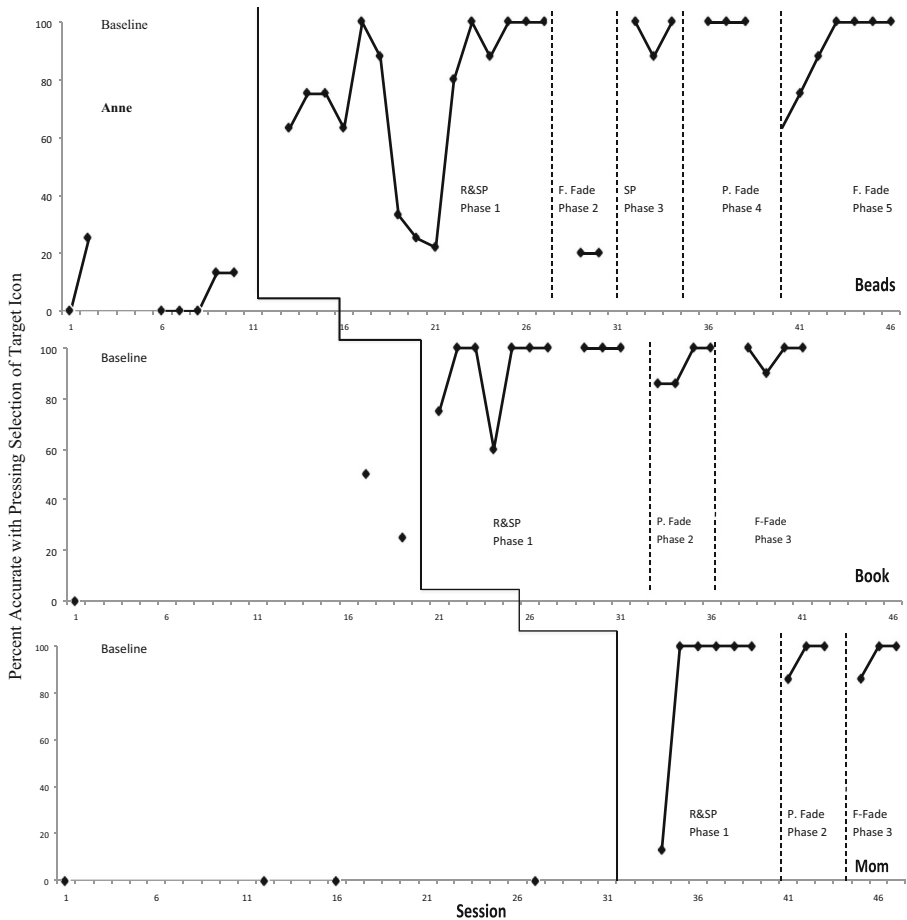


Fig. 2 The percent accurate of Annie’s pressing of the target request, ‘beads’ (top panel), ‘book’ (middle panel), and ‘Mom’ (bottom panel) across baseline and intervention sessions. Phases are notated in the following way on the figure: response and stimulus prompting=R&SP; stimulus prompting=SP; partial stimulus fading=P. Fade; full stimulus fading=F. Fade

depicts ‘beads’, the middle panel depicts ‘book’, and the bottom panel depicts ‘Mom.’ An overview of the phases included in Annie’s intervention is provided in Table 1.

Response and Stimulus Prompting (R&SP) With Annie Upon the staggered introduction of R&SP in Phase 1, there was immediate improvement in accuracy level and trend for all three requests, and there was continued reduction in variability (particularly for ‘book’ and ‘Mom’ requests).

The first request for ‘beads,’ remained below criterion range during baseline probes (range, 0 to 25 %), and upon introduction of R&SP, there was an immediate and overall increase in level and trend, despite some deteriorating accuracy and variability in sessions 18-21. Beginning in session 22, accuracy improved until Annie reached criterion during session 25 (after a total of 13 intervention sessions).

The second request for ‘food’ remained below criterion range during baseline probes (range, 0 to 50 %), and upon staggered introduction of R&SP there was an immediate increase in level (from 20 to 70 %) with an increasing and stable trend in accuracy (range, 60 to 100 %); criterion was reached at intervention session 29 (after a total of 7 intervention sessions).

The third request for ‘Mom’ remained at zero during baseline probes, and after the introduction of the R&SP there was a slight increase in level upon introduction of the first intervention session (from 0 to 10 %); there was a large increase in level upon the remaining 100 % and remained at that level with a stable trend (range, 10 to 100%). Criterion was reached at intervention session 38 (after a total of 4 intervention sessions).

Stimulus Fading With Annie After Annie reached a criterion level of accuracy with ‘beads’ in Phase 1, full stimulus fading in Phase 2 resulted in an immediate deterioration in accuracy (from 100 to 20 % during sessions 30 and 31). Accuracy recovered to criterion levels (range, 88 to 100 %) during a return to stimulus prompting procedures in Phase 3. Beginning in Phase 4, partial stimulus fading resulted in a high and stable level of accuracy (range, 100% to 100 %). Annie reached criterion during session 38 (after a total of 3 sessions). Beginning in Phase 5, full stimulus fading resulted in a slight decrease in accuracy level followed by an increase in trend and level (range, 63 to 100 %). Criterion was established by session 43 (after a total of 6 sessions).

After Annie reached criterion for ‘book’ in Phase 1, partial stimulus fading was introduced in Phase 2. This resulted in a high and stable accuracy with criterion being established at session 33 (after a total of 3 sessions). Beginning in Phase 3, full stimulus fading resulted in an immediate decrement in performance level (from 100 to 63 %), followed by improved accuracy with criterion being established at session 40 (after a total of 5 sessions).

When Annie reached criterion levels of accuracy with ‘Mom’ in Phase 1, partial stimulus fading was introduced in Phase 2, which resulted in increased accuracy with criterion being reached after three sessions. Beginning in Phase 3, full stimulus fading resulted highly accurate and stable responding with criterion being established after 3 sessions.

Rory Figure 3 displays Rory’s requesting accuracy in eye-gaze requesting during baseline, response prompting (RP), procedural changes, safe laser point prompting (LP) and fading phases. The three panels depict accuracy data during baseline and intervention staggered across the three requests; the top panel depicts ‘TV’, the middle panel depicts ‘food’, and the bottom panel depicts ‘doll.’ An overview of the phases included in Rory’s intervention is provided in Table 1.

Response Prompting With Rory After the introduction of the RP for the first request ‘TV,’ accuracy did not improve from baseline performance (range, 0 to 10 %). Similarly, when ‘food,’ was introduced during intervention using a RP there was no improvement in performance. Consequently, the RP was not implemented when the third request ‘doll’ was introduced.

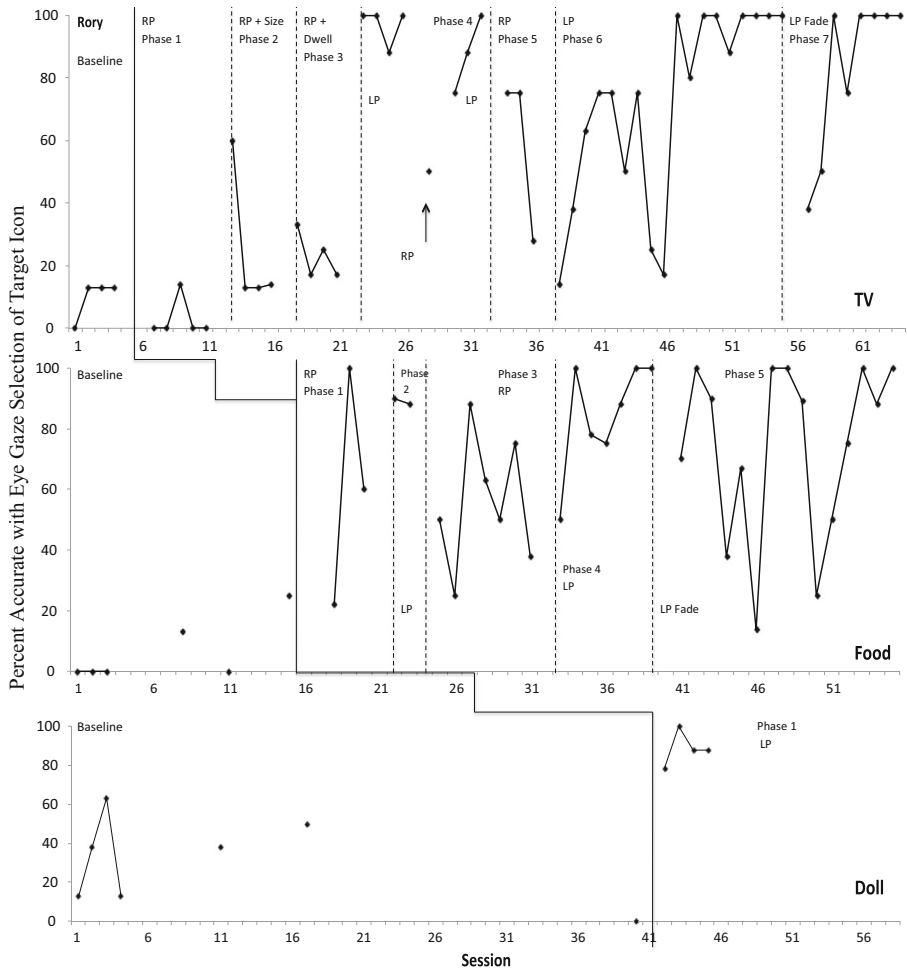


Fig. 3 The percent accurate of Rory’s eye-gaze selection of the target request, ‘TV’ (top panel), ‘food’ (middle panel), and ‘doll’ (bottom panel) across baseline and intervention sessions. Phases are notated in the following way on the figure: response prompting=RP; response prompting with increase symbol size=RP+size; response prompting with decreased dwell time=RP+dwell; safe laser pointer prompt=LP; fading of safe laser pointer prompt=LP fade

Procedural Changes During RP With Rory Upon introduction of increased size stimulus prompt in Phase 2 and decreased dwell time from 500ms to 420ms in Phase 3, accuracy did not improve enough to approach the criterion range (range, 10 to 60 % in Phase 2 and 10 to 38 % in Phase 3).

Safe Laser Pointer Prompting With Rory When Phase 4 was introduced for the symbol, ‘TV’, accuracy immediately improved with a level change (from 20 to 100 %) and remained stable (range, 80 to 100 %) with criterion being established by session 26 (after 3 sessions). Interventionists briefly removed the LP at session 28, which resulted in an immediate decrease in accuracy (from 100 to 50 %). The LP was reintroduced at session 29, resulting in immediate increase in level and trend (range, 70 to 100 %) and accuracy returned to criterion range after 3 sessions. The LP was briefly removed again in Phase 5 performance again deteriorated (range, 70 to 24 %). After the LP was reintroduced in Phase 6, accuracy continued to decrease with an immediate level change to 10 % with high degree of variability within sessions 38-46 (range, 17 to 75 %). During session 47, accuracy began to improve. Between sessions 47-55 accuracy increased substantially and accuracy stabilized (range, 80 to 100 %). By session 49, performance had returned to a criterion level of accuracy. However, the researchers continued to conduct intervention for 6 more sessions to ensure the stability of the criterion performance given the preceding excessive variability during the phase.

For the second request, ‘food,’ upon introduction of the LP in Phase 2, there was an immediate increase in level (from 60 to 90 %) and stability improved (range, 90 to 86 %). The LP was removed in Phase 3 resulting in an immediate decrease in level (from 86 to 50 %), and increase in variability (range, 20 to 85 % across 7 sessions). The LP was reinstated in Phase 4, resulting in immediate increase in trend and level (from 38 to 50 %), and improved stability (range, 50 to 100 %); criterion was reached at session 39 (after a total of 7 sessions).

For the third request, ‘doll,’ upon introduction of the LP following baseline, accuracy immediately increased in level (from 0 to 78 %) and trend (range, 70 to 100 %); criterion was reached after a total of 4 sessions.

Stimulus Fading of Safe Laser Point Prompting Rory The LP was faded for two requests, ‘TV’ and ‘food.’ Upon LP fading in Phase 7 for ‘TV,’ accuracy immediately decreased in level (from 100% to 38%); however an increasing and stable trend followed (range, 38 to 100%), Criterion was reached at session 63 (after a total of 7 sessions).

Upon LP fading in phase 5 for ‘food,’ accuracy immediately decreased in level (from 100 to 70 %) and trend, and a high amount of variability persisted in sessions 41–50 (range, 14 to 100 %). At session 51, accuracy began to improve and an increasing trend and reduced variability followed; criterion was reached at session 55 (after a total of 14 sessions).

Discussion

This study examined the effectiveness of an intervention package to teach requesting for two participants with Rett syndrome using a graphic mode, aided communication

system. For Annie, a relatively rapid acquisition was observed during response and stimulus prompting across all three requests taught. Establishing a more conventional communication means for this learner was important because, at the age of 27, she had never previously learned to use conventional communicative symbols and she experienced severe motor impairments. Yet, in the current investigation, she met mastery criterion within 25 response and stimulus prompting sessions for the first request, and 3 sessions each for the other two requests. She was able to maintain this high level of accuracy throughout the majority of the sessions. Throughout response-prompt fading, Annie was quickly able to use the stimulus prompt (of the larger size of the target item) as the main prompt for the response. Upon stimulus size fading, it appeared that when attempting to fade too quickly (Phase 2 for 'book'), accuracy decayed. Systematic fading through smaller changes to the target stimulus, and the use of previously mastered sizes as prompts appeared to facilitate fading of the sizes of the symbol while maintaining accurate responding. Annie's relatively rapid acquisition of these skills demonstrates the potential to teach requesting skills to an individual with RS, who had very limited exposure and intervention with any means of formal communication. It is also important to note that Annie met criterion accuracy faster with each successive request; suggesting that if she continued to receive intervention, it may not take as many opportunities (dose) to teach additional requests and further expand her communication repertoire.

For Rory, the initial response-prompting hierarchy was unsuccessful for increasing accuracy, and subsequent changes to symbol size and dwell times were also unsuccessful. Upon introduction of the safe laser pointer prompt, Rory's accuracy quickly increased and remained relatively high when the laser prompt was being utilized. The efficacy of the laser prompt and the response prompt hierarchy was compared in a withdrawal, and it was demonstrated that when the laser prompt was removed, accuracy decreased and a high degree of variability was observed.

Overall, Rory was able to increase accuracy and reach criterion levels of performance with all three requests using an eye-gaze access method on the SGD. This is an important finding, as there has not been a previous experimental demonstration of a learner with RS acquiring multiple requests using eye-gaze on the SGD. However, there were also limitations to the intervention package effectiveness for Rory's requests, first, there was a significant amount of overall variability throughout the study, second, the laser prompt was not faded for the last request, and third, the stimulus prompts were not faded for any of the requests. The difficulties encountered with teaching the requests on the eye-gaze device to Rory highlight the need to investigate the effectiveness of potential intervention strategies to: (a) ensure that intervention time is not wasted with ineffective prompts, (b) limit the frustration of the individual as he or she learns the response, and (c) differentiate learners who are capable of learning eye-gaze responses with the careful application and fading of prompting procedures.

The temporary use of prompting allows for shaping a communicative response, while simultaneously allowing for the communicative context to come under stimulus control (the response happens following a contextual cue in the environment or a discriminative stimulus). However, for the appropriate context to come under stimulus control, the prompting must be faded so that the prompt itself does not begin to serve as the discriminated stimulus (SD) for the communicative response to occur. The safe laser prompt was effective at teaching the eye-gaze response to Rory, however it was

also a difficult prompt to fade. One reason for this difficulty may be that the laser served as a non-criterion related prompt, meaning that it did not manipulate or isolate Rory's ability to visually discriminate the salient differences between the S+ (the target symbol) and the S- (the distracter symbol) (Etzel and LeBlanc 1979; Dietz and Malone 1984). As fading occurs, non-criterion related prompts make the required transfer of stimulus control more difficult because the individual may not have been attending to the most important features of the target stimulus. In this case, Rory may have been getting the response correct during safe laser-prompted trials because she was only attending to the saliency of the light and not the symbols. Non-criterion related prompting warrants further investigation, as it may help to teach the initial response, (i.e., Rory fixing her gaze in one spot for the set dwell time). Whereas, criterion-related prompting might help with the transfer of stimulus control as fading occurs, (i.e., Rory discriminating the salient features of the symbols).

There are several other important factors to consider when interpreting the results of the current study. First, in regards to treatment intensity parameters (Warren, Fey, and Yoder 2007), there were a high number of opportunities per session, with multiple sessions occurring per day (dose); however, the number of sessions per week was low, with sessions occurring only 1-2 times per week (dose frequency), and a long duration of the intervention was required for the participants to reach criterion levels of performance. Though an optimal level of treatment intensity for communication intervention with learners with RS has not been investigated, it is possible that the mismatch of treatment intensity (a high dose occurring over a low dose frequency spread across a long duration) may have contributed to both the number of sessions required to meet mastery criterion for the participants and the high degree of variability in Rory's accuracy. There may have also been factors related to maintenance when sessions were spaced apart by 7 days. The participants, particularly Rory, likely used other means of idiosyncratic requesting behavior during non-session times. For example, Rory often fixed her eyes on a bowl of food itself or looked between the caregiver and the food. During intervention sessions, idiosyncratic requests were redirected and did not result in delivery of the reinforcers; however, idiosyncratic requests often competed with attempts to prompt her to look at the SGD. Toward the end of intervention sessions, she engaged in idiosyncratic requesting less often and was more easily prompted to look at the SGD. Ideally, communication interventions should be provided in the school or by related service providers and, therefore, should be delivered on a denser schedule, which may help to alleviate these problems.

Another important factor to consider with interventions targeting requests is the potential impact of properties of the reinforcing item on the requesting behavior. With a relatively high number of opportunities within a session (short inter-trial interval), satiation may have acted as an abolishing operation to requesting after the participant received the reinforcer several times. Therefore, the reinforcement value and satiation effects of the targeted requests may have contributed to variability in responding, or otherwise influenced the results of the current study.

The majority of the communication intervention literature for RTT has been focused on teaching generalized requests, through low technology AAC (Sigafoos and Couzens 1995) and microswitch technology AAC (Byiers et al 2014). Emerging evidence for explicit requests has been investigated both with low and high technology AAC (Stasolla et al. 2014); however, without reported accuracy data in this study, it is

difficult to determine the level of the participants' ability to discriminate between distracter symbols or target requests above chance. The results of the current study were consistent with previous literature demonstrating that participants with Rett syndrome can acquire some basic communication skills. Our results were also consistent with Stasolla and colleagues (2014) that demonstrated the participants required a high number of intervention sessions to acquire the communication skills. Although promising, previous literature, because of the small number of participants, requires additional studies demonstrating the experimentally controlled acquisition of new vocabulary items among girls and women with RS. Additional replications are needed to determine effective prompting strategies and dosage effects for teaching initial communication skills to individuals with RS. Future research must also examine how to teach more complex communication skills so that individuals with RS are not limited to only generalized and context-dependent communication repertoires.

This study aimed to address the gap in the experimental intervention literature designed to teach requests to girls and women with RS. The results can serve as a starting point for potential prompting and fading techniques to initially teach multiple requests, that require precursory discrimination, to individuals with RS or other related disorders. Both participants were able to achieve performance within criterion range of accuracy with the three requests, and to accurately engage in the requests at a level of 80 percent or higher while ignoring the distracter symbols. Both participants also displayed a relatively high level of consistent responding during the opportunities (as it was counted as an error if they did not respond within the trial).

Another contribution of this study is the demonstration of a participant with RS acquiring multiple requests through eye-gaze on a SGD. Eye-gaze holds promise as a high technology solution to teaching more complex communication skills to individuals who have severely impaired motor and cognitive function than can be achieved by relying on low technology AAC systems. However, there are also many gaps in the current literature pertaining to teaching eye-gaze using SGD for girls and women with RS. The majority of the existing literature on SGD eye-gaze and communication is focused on adults with ALS who have intact cognition and likely a previous history of fluent communication to build on, (Fager, Bardach, Russell, and Higginbotham 2012) which limits the generalization of those findings when applied to girls and women with RS who may not have intact cognition and who may not have a history of complex or fluent communication. Moreover, eye-gaze is a response type that could inadvertently lead to misinterpretation of communication intent when the individual has not been taught how to complete the response (i.e., fix his or her gaze), nor how to discriminate the communication options on the device (attend to the salient differences in the symbols). If a SGD with eye-gaze is programmed with many communication messages and a person is not taught to use the device but is just looking at it and this activates speech output, the individual may not be intending to communicate the messages it contains. Caregivers may attribute communicational intent to this type of behavior. An example of this problem lies in the Facilitated Communication literature, which documents the accidental interpretation of non-intentional responses as communication of people with severe disabilities (see Mostert 2001 for a review). This misattribution of intent may inhibit seeking or implementing other interventions to teach eye-gaze communication skills more explicitly to individuals with RS. It is important to

examine under what conditions acquisition and accuracy of initial communication skills using eye-gaze responding can be enhanced.

Potential areas for future research on communication intervention for girls and women with RS using high technology AAC, such as SGDs, should investigate prompting methods, increased treatment intensity including a denser intervention schedule (dose frequency), and discrimination among multiple communication responses. As AAC technology continues to grow, another important aspect of future research must investigate the conditions under which eye-gaze can be effectively and efficiently taught to individuals with severe and multiple disabilities.

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Compliance with Ethical Standards “All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.” “Informed consent was obtained from parents or guardians for all individual participants included in the study.”

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