Research in Autism Spectrum Disorders 3 (2009) 304–318

Contents lists available at ScienceDirect

Research in Autism Spectrum Disorders

Journal homepage: http://ees.elsevier.com/RASD/default.asp

Review

Communication intervention in Rett syndrome: A systematic review

Jeff Sigafoos [a,*], Vanessa A. Green [a], Ralf Schlosser [b], Mark F. O'eilly [c], Giulio E. Lancioni [d], Mandy Rispoli [c], Russell Lang [c]

[a] Victoria University of Wellington, New Zealand
[b] Northeastern University, Boston and Childrens Hospital Boston at Waltham, MA, USA
[c] Meadows Center for Preventing Educational Risk, The University of Texas at Austin, Austin, TX, USA
[d] University of Bari, Bari, Italy

ARTICLE INFO

Article history:
Received 25 September 2008
Accepted 26 September 2008

Keywords:
Communication intervention
Rett syndrome
Systematic review

ABSTRACT

We reviewed communication intervention studies involving people with Rett syndrome. Systematic searches of five electronic databases, selected journals, and reference lists identified nine studies meeting the inclusion criteria. These studies were evaluated in terms of: (a) participant characteristics, (b) target skills, (c) procedures, (d) main findings, and (e) certainty of evidence. Across the nine studies, intervention was provided to a total of 31 participants aged 2:7–17:0 (years:months). Communication modes included speech, gestures, communication boards, and computer-based systems. Targeted communication functions included imitative speech, requesting, naming/commenting, and various receptive language skills (e.g., respond to requests, answer questions, receptively identify symbols). Intervention approaches included early intensive behavioral intervention, systematic instruction, and music therapy. Positive outcomes were reported for 26 (84%) of the 31 participants. However, these outcomes must be interpreted with caution because the certainty of evidence was inconclusive for all but one of the studies. Due to the limited number of studies and their methodological limitations, the evidence base supporting current approaches to communication intervention for individuals with Rett syndrome remains inconclusive.

* Corresponding author at: College of Education, Victoria University of Wellington, Karori Campus, P.O. Box 17-310, Wellington, New Zealand.
E-mail address: jeff.sigafoos@vuw.ac.nz (J. Sigafoos).

1750-9467/$ – see front matter © 2008 Elsevier Ltd. All rights reserved.
doi:10.1016/j.rasd.2008.09.006

Contents

1. Introduction

Rett syndrome is a progressive neurodevelopmental disorder caused by X-linked genetic mutations that occur almost exclusively in females (Amir et al., 1999). With an estimated prevalence of 1 per every 10,000 females, it is considered one of the most common causes of profound/multiple disability in girls (Ellaway & Christodoulou, 2001; Leonard, Bower, & English, 1997). The condition is characterized by progressive loss of functioning beginning at around 6–18 months of age. There is currently no cure for Rett syndrome, and hence there is a need for research that can assist treatment planning (Matson, Fodstad, & Boisjoli, 2008). In terms of treatment planning, the progressive deterioration and eventual loss of ability in most areas of functioning (e.g., cognitive, motor, communication, social) certainly creates unique challenges in relation to the design and delivery of effective education, therapy, and care.

The progressive decline in functioning associated with Rett syndrome appears to follow four fairly predictable stages (Dunn, 2001; Hagberg & Witt-Engerström, 1986). The first stage (early onset deceleration) occurs between 6 and 18 months. This stage is characterized by deceleration in head growth and noticeable reduction in the child's play, communication, and social interaction. In the next stage (rapid destructive; 1–3 years), there is often a complete or nearly complete loss of both speech and functional use of hands. During this stage the child is often highly irritable and can often be misdiagnosed as having an autism spectrum disorder due to the emergence of autistic-like symptoms (Matson et al., 2008; Smith, Klevstrand, & Lovaas, 1995). With the third, (pseudostationary) stage (2–10 years), the child may develop seizures, ataxia, and scoliosis. It is usually during this stage that the characteristic and almost constant washing-like hand mannerisms emerge. By about 10 years of age, in the final late motor deterioration stage, scoliosis worsens and mobility is often so severely limited that most children will require the use of a wheelchair. In the end, the child's behavioral repertoire and responsiveness to environmental stimulation are extremely limited. While there is variability in the degree and type of symptom presentation, both within and across these four stages (Matson et al., 2008), many, if not most, individuals with Rett syndrome will eventually become unable to walk, unable to talk, and unable to use their hands for any functional activity.

From an educational perspective, it might be possible to enhance functioning of individuals with Rett syndrome through well-designed educational interventions. Because of the profound and wide-ranging impairments associated with Rett syndrome, effective education would seem to require a careful

prioritization of treatment goals followed by skilled use of the best available interventions. In terms of prioritization, enhancement of communication skills would seem highly relevant because the condition is associated with early loss of speech and language and extremely limited residual communication ability (Sigafoos & Woodyatt, 1996). To compensate for this loss and limited communication ability, researchers have suggested that the acquisition of nonverbal or alternative modes of communication is a real possibility for many individuals with Rett syndrome (Burford & Trevarthen, 1997). An important issue is whether there are in fact any intervention studies that have demonstrated effective procedures for teaching alternative modes of communication to individuals with Rett syndrome.

To facilitate evidence-based practice in this important area of educational priority, we herein provide a systematic review of studies on communication intervention for individuals with Rett syndrome. The objective of this review is to describe the characteristics of these studies (e.g., participants, target behaviors, intervention procedures), evaluate intervention outcomes, and appraise the certainty of the evidence for the existing corpus of intervention studies. A review of this type is primarily intended to guide and inform evidence-based practice in the design of communication interventions for individuals with Rett syndrome. A secondary aim is to identify gaps in the existing database so as to stimulate future research efforts aimed at developing new and more effective communication interventions for this population.

2. Method

We conducted a systematic review of studies that were focused on evaluating the effects of communication intervention for individuals with Rett syndrome.

2.1. Inclusion and exclusion criteria

To be included in this review, the article had to be a research study that examined the effects of providing of communication intervention to at least one person with a diagnosis of Rett Syndrome. Communication intervention was defined as implementing one or more therapeutic/teaching procedures for the purpose of trying to increase or improve the person's communication skills or abilities. Examples could include: (a) being more responsive to the person's existing communication skills (e.g., responding to vocalizations, body movements, or eye gaze), (b) teaching new forms of communication (e.g., teaching the use of graphic symbols or a speech-generating device), (c) increasing specific communication functions (e.g., increasing requesting or initiations for interaction) and/or (d) increasing participation in communication interactions (e.g., increasing attention to a communicative partner). Studies that focused only on the description of communication skills or only on the assessment of communication skills were not included. This exclusion extended to studies that made use of the child's existing communication skills during an assessment. Two further studies (Koppenhaver, Erickson, & Skotko, 2001; Skotko, Koppenhaver, & Erickson, 2004) were excluded because they appeared identical, in terms of participants and procedures, to an already included study (Koppenhaver, Erickson, & Harris et al., 2001).

2.2. Search procedures

Systematic searches were conducted in five electronic databases: Cumulative Index of Nursing and Allied Health Literatures (CINAHL), Education Resources Information Center (ERIC), Medline, Linguistics and Language Behavior Abstracts (LLBA), and PsycINFO. Publication year was not restricted, but the search was limited to English-language journal articles.

On all five databases, the terms *Rett syndrome* (or *Rett's disorder*) and *Communication* were inserted into the *Keywords* field. Abstracts of the records returned from these electronic searches were reviewed to identify studies for inclusion in the review (see *Inclusion and Exclusion Criteria)*. The reference lists for the included studies were also reviewed to identify additional articles for possible inclusion. Hand searches, covering the period 1987–2008, were also completed for the journals that had published the included studies. Finally, using an author search, the five databases were searched again for additional related work by authors of the studies that met the inclusion criteria. Our initial search of the databases, journals, and reference lists occurred during the period August to October,

2007. The five electronic databases were searched again in August and September 2008 to check for recent additions to the literature. From this combination of search procedures, 32 articles were identified for possible inclusion in the systematic review.

2.3. Data extraction

Each identified study was first assessed to determine if it met the pre-determined inclusion criteria. Each identified study that met the pre-determined inclusion criteria was analyzed and summarized in terms of: (a) participants, (b) mode of communication (e.g., manual signs, communication board), (c) communication skill(s) taught to the participants, (d) intervention procedures, (e) results of the intervention, and (f) certainty of evidence. The certainty of evidence for each study was rated as either conclusive or inconclusive based on the definitions applied by Millar, Light and Schlosser (2006).

Appraising the certainty of evidence (for included studies only) followed a two-stage process. First, only studies that included a recognized experimental design (e.g., multiple-baseline, ABAB) could be considered as having the potential to provide conclusive evidence. Thus any study that lacked a recognized experimental design was automatically classified as inconclusive. This included narrative case studies, pre-post testing without a control group, and studies using A-B or intervention-only designs. Second, studies that made use of a recognized experimental design also had to meet four additional standards to be classified as providing conclusive evidence. First, the data had to provide a convincing demonstration of an intervention effect. This determination was based on visual inspection of data trends within a phase and across phases using criteria described by Kennedy (2005). For example, there had to be a clinically significant increase in correct requesting when intervention was introduced. Second, if relevant, there had to be adequate inter-observer agreement data (i.e., 20% of the sessions and 80% or better agreement). Third, the dependent and independent variables had to be operationally defined. Fourth, the procedures had to be described in sufficient detail to enable replication.

2.4. Inter-rater agreement

The first author made an initial determination as to whether each study from the initial pool of 32 met the inclusion criteria. A research assistant was trained to apply the inclusion/exclusion criteria by working through one randomly selected study with the first author. After this, the research assistant independently assessed each of remaining 31 studies against the inclusion/exclusion criteria. Agreement as to whether a study should be included or excluded was obtained on 26 of the 31 studies (84%). To resolve the discrepancies, a second research assistant independently assessed the five studies on which there was initial disagreement and, based on this, all five studies were excluded. The overall result was that 23 studies were excluded and 9 studies were included for analysis in this review.

The first author extracted information to develop an initial summary of the nine included studies. The accuracy of these summaries was independently checked by one of three co-authors (the second, sixth, and seventh authors) using a checklist that included the initial summary of the study and a number of questions regarding various details of the study (e.g., Is this an accurate description of the participants? Did the authors assess generalization? Is this an accurate summary of the certainty of evidence?). Co-authors were asked to read the study and the summary and then complete the checklist. In cases where the summary was not considered accurate, the co-authors were asked to edit the summary to improve its accuracy.

This approach was intended to ensure accuracy in the summary of studies, but it also provided a measure of inter-rater agreement on data extraction and analysis. There were 72 items on which there could be agreement or disagreement (i.e., 9 studies $\times$ 8 questions per study). Agreement was obtained on 67 items (93%). In the five instances where aspects of the summaries were considered inaccurate, changes were made to more fully describe participants, target behaviors, and procedural details.

3. Results

From 32 potential studies, 23 studies were excluded, leaving 9 studies for summary and analysis. Appendices A and B list included and excluded studies, respectively. Table 1 summarizes the

Table 1
Summary of communication intervention studies involving participants with Rett syndrome[a].

Study	Participants	Target Skills	Procedures	Main Findings	Certainty of Evidence
Number 1: Smith et al. (1995)	Three girls with Rett syndrome aged 37 (S1), 33 (S2), and 31 (S3) months at intake and 65, 57, and 58 months at follow-up. IQ scores were 48 (S1), 34 (S2), and 30 (S3) at intake. At intake, all three lacked play skills, had tantrums, self-stimulatory behavior, and were not toilet trained. S1 imitated words at intake; S2 and S3 were mute. Stage of Rett syndrome was not specified.	Speech mode communication was targeted. Treatment focused on increasing a range of skills (e.g., feeding, toileting, imitate sounds/actions, respond to requests, and name objects) and decreasing problem behaviors, such as tantrums and stereotypy.	Intake and follow-up data were derived from the Bayley Scales of Infant Development (Bayley, 1969), observations, and parent description. The manualized intervention (Lovaas et al., 1981) followed a basic (respond to adult request) to complex (conversation) sequence. Student therapists implemented discrete-trial training using operant procedures (e.g., shaping, chaining, discrimination training, reinforcement). Intervention occurred in a one-to-one format for 10–30 h/week over 8–24 months.	IQ was untestable for all three children at follow-up, suggesting regression of cognitive ability. S1 made progress in (a) affection to adults, (b) tantrums and self-stimulation, (c) toilet training, and (d) functional use of spoken single words. However, word articulation deteriorated over time. S2 showed fewer tantrums, but remained mute and showed no change in self-stimulation, play skills or toileting. S3 also remained mute at follow-up and showed no improvement in affection, toy play, tantrums, self-stimulation, or toileting. Follow-up gains were offset by deterioration in other areas.	The certainty of evidence for the lack of an intervention effect is inconclusive because the effects of intervention were evaluated in pre-post design and the study appeared to lack control conditions (e.g., baseline or a control group). In addition, it is unclear if the pre-post changes were statistically significant or if the observational data and parent descriptions were reliable. Treatment integrity was not assessed, although the therapists did receive "extensive training and supervision" (p. 318).
Number 2: Van Acker and Grant (1995)	Three girls with Rett syndrome aged 11-7 (S1), 6-8 (S2), and 5-2 (S3) years-months. Each had acquired single words or 2–3 word sentences, but had no speech when the study began. Current stage of Rett syndrome was not specified, although all 3 were said to have gone through Stage 2 (rapid deterioration).	Touching a dynamic symbol on a computer screen to request food/drink. Collateral effects of intervention on stereotypy were also measured.	During an initial baseline phase, the computer system was not operating, but drawings of preferred foods or drinks were placed on the screen (one drawing per trial). If the child touched the drawing, she received a small portion of the corresponding item. A second baseline assessed possible reinforcing effects of dynamic movement of symbols, but screen touching did not produce food/drink. During intervention, the computer displayed a symbol (e.g., a carton of milk) and emitted a synthesized speech prompt (e.g., "Would you like some milk?"). If the child touched the symbol, the symbol became animated, a relevant speech output was heard (e.g., "Yes, I would like some milk"), and child received a small portion of the requested item. Training occurred in the classroom with generalization probes conducted in the lunchroom and another classroom for all 3 students, and the student's homes for 2 of the 3 students.	S1 and S3 reached criterion on 7 symbols (60% correct across three sessions) within approximately 15–25 sessions. Each session included 20 opportunities to request each item. S2 reached criterion on only 1 of the 7 items (juice), but showed modest increases in correct requesting from baseline to intervention on an additional 5 items. S1 generalized correct use of the system to the lunchroom with peers and to a lesser extent at home. S2 showed generalization to the lunchroom with the one acquired symbol (juice). For S2 generalization was not assessed at home. S3 showed generalization at a level comparable to that achieved at the end of intervention to the lunchroom, another classroom, and the home setting. All 3 children showed evidence of discriminated requesting in that they were statistically more likely to request preferred versus non-preferred food/drink. Stereotypy decreased for S1 and S3, but not S2.	The certainty of evidence is conclusive because baseline and intervention phases were staggered across two sets of food/drink in a multiple-baseline design and acquisition to criterion was replicated in 2 of the 3 children. However, the experimental design was compromised in terms of number of replications because S3 showed increased requesting during baseline with the second set of items. In addition, while data on child requests were recorded automatically via the computer software, treatment integrity was not assessed.

Number 3: Sigafoos et al. (1996)	Four girls with Rett syndrome aged 17(S1), 12(S2), 7(S3) and 15(S4) years. S1, S2, and S4 had no speech when the study began, but S1 had acquired three single words prior to 18 months of age. S3 occasionally produced the single word *mum*. S1, S2, and S4 were in the final (late motor deterioration) stage, whereas S3 was in the third (pseudostationary) stage.	Touching a line drawing (*WANT*) affixed to a board to request food, drinks, and toys. S3 and S4 later received modified systems; either touching a flattened potato chip bag to request chips (S3) or pressing a switch to activate music (S4).	The number of correct requests during classroom sessions of 10 trials was compared across baseline and intervention. During baseline, preferred items (e.g., potato chips, apple juice, picture-book) were in view but out of reach and the communication board with the *WANT* symbol was in reach. The trainer initiated each trial by saying *Let me know if you want something* and then allowed the child 30 s to make a correct request by touching the WANT symbol. At the end of 30 s, preferred items were placed within reach and the child could chose one of the items. Correct requests were not reinforced in baseline. Intervention procedures were identical to baseline, except that correct requests were reinforced by allowing the child to choose one preferred item and, if the child did not make a correct request within 10 s, the therapist used a sequence of verbal, gesture, and physical prompts to recruit a correct request.	During baseline, the percentage of correct requests averaged 30, 20, 0, and 0% for S1, S2, S3, and S4, respectively. With intervention, correct requesting stabilized at 60-100% for S1 and increased to a mean of 47% for S2. S3 and S4, however, showed no correct requests during their first 5-6 intervention sessions. Given this lack of progress, the intervention was modified for S3 and S4 by conducing a second baseline to intervention sequence. For this second sequence, S3 was provided with a modified (potato chip) symbol and S4 requested music by touching a switch. During baseline with the modified system, correct requests occurred less than 10% of the trials. With the second intervention correct requests increased to 80 and 70% for S3 and S4, respectively, but remained highly variable for both children across the final intervention phase.	The certainty of evidence for the effects of the initial intervention is inconclusive because the percentage of correct requests during baseline did not stabilize for S1 and S2 and S3 and S4 failed to acquire the initial requesting response. The certainty of evidence is also inconclusive for the effects of the second intervention because although the multiple-probe design showed that changes occurred only when the intervention phase was implemented, this effect was replicated only once, from S3 to S4, and their two respective interventions were not comparable in terms of the dependent variable. Inter-observer agreement on correct requesting and treatment integrity were assessed on 17–23% of all sessions and in each case exceeded 80%.
Number 4: Evans and Meyer (1999)	One girl with Rett syndrome, aged 5 years, 11 months, with no speech. Stage of Rett syndrome was not specified.	Making an informal gesture (i.e., crossing her arms in front of her) to request a hug.	The authors describe a number of interventions conducted over a 3-year period, but this summary describes only the one intervention that focused on teaching the child to request a hug from a familiar adult. This intervention occurred in the classroom. Teaching opportunities were created whenever the child approached an adult. At this point, the teacher used "standard prompting, shaping, and social reinforcement procedures" (p. 196) to teach independent gesture use.	Observations of 8 min duration were conducted once per fortnight on random days and at different times of the day. Observations occurred within three activities (free-play, one-to-one training, and group instruction) during baseline and intervention phases. After 6 months of intervention, there was no evidence of acquisition of the arm-crossing requesting gesture.	The certainty evidence for the lack of an intervention effect is inconclusive because the design was pre-experimental (e.g., seemingly an A-B design) although it is not clear if baseline data were collected). No inter-observer agreement or treatment integrity data were presented. Outcome data for this intervention were presented only in anecdotal, narrative form.

Table 1 (*Continued*)

Study	Participants	Target Skills	Procedures	Main Findings	Certainty of Evidence
Number 5: Koppenhaver et al. (2001)	Six girls with Rett syndrome from 3.6 to 7 years of age. One child communicated via simple spoken requests; the others did not speak. Stage of Rett syndrome was not specified. Scores on the Bayley Scales of Infant Development ranged from 5 to 19 months. Scores on the Vineland Adaptive Behavior Scales ranged from 9 to 17 months.	Use of picture symbols and speech-generating devices to label pictures, make comments, and respond to wh-questions.	The study involved a baseline followed by three intervention phases. During baseline, mother and child were videotaped while reading familiar and unfamiliar storybooks. Videotaping continued during the 3 intervention phases. First, children received hand splints to investigate effects on the non-dominant hand. Second, children were given picture symbols and speech-generating devices with relevant vocabulary. Third, parents were trained to (a) attribute meaning to the child's communicative attempts, (b) ask questions and make comments to create communicative opportunities for the child, and (c) use time delay and verbal, model, and physical prompts to evoke correct use of the communication symbols and speech-generating device.	All six children made fewer than 0.5 communication responses per minute during baseline. This increased to a mean of approximately 1.4 per minute during the final intervention phase, but performance ranged from approximately 0.3–2.6 per minute, indicating considerable variability across children. Similar increases from baseline to the final intervention phase (and similarly high levels of variability across children) were obtained for familiar versus unfamiliar storybooks.	The certainty of evidence for an intervention effect is inconclusive because the sequential introduction of phases did not control for the additive effects of the three separate intervention components nor for familiarity, practice or maturation effects. Although the authors state that the study followed a multiple baseline across behaviors design, session-by-session data are not provided to evaluate trends within each phase. The high variability around the means suggests that there was a considerable amount of overlapping data across phases, reducing confidence of an intervention effect. In addition data were extracted from only a small sample of the videotapes made (i.e., 2 sessions per phase per child) and is therefore of unknown representativeness. The amount of intervention that parents provided was not controlled, and treatment integrity was not assessed.
Number 6: Yasuhara and Sugiyama (2001)	Three girls with Rett syndrome aged 5 (S1), 4 (S2), and 6 (S3) years. All 3 were described as having "no articulation." Stage of Rett syndrome was not specified.	Target behaviors included: (a) listen, (b) sing, (c) play instruments, (d) perform fine motor tasks, (e) use language, and (f) show social behavior.	Children received 30-min of music therapy once per week for a total of 40 (S1 and S2) or 12 sessions. Target behaviors were assessed from videotapes of each session and coded in terms of developmental level based on the Denver Developmental Screening Test. The music therapy program was individualized for each child based on parent interview, developmental age, and daily activity. Sessions included engaging the child in song and instrument play and providing choices of instruments.	Objective data were presented only for S1. Her ratings on target behaviors for S1 increased from 1–2 to 2.5–5 from Session 1 to Session 40. For S2 and S3 data were presented in the form of narrative summaries. S2 was said to have begun to ask for things by uttering sounds and learned to indicate her thoughts by pointing. S3 showed increased enjoyment of songs and responded by uttering sounds.	Certainty of evidence is inconclusive because the study did not include an experimental design or any control groups/conditions. The target behaviors were not objectively defined and the treatment was not described in sufficient detail to enable replication. The data for S1 are of unknown reliability and the results for S2 and S3 are reported in narrative form only. Treatment integrity was not assessed.

Number 7: Hetzroni et al. (2002)	Three girls with Rett syndrome aged 9–10 years of age. All three were described as having no speech. All 3 girls were in the third (pseudostationary) stage of Rett syndrome.	Select a named picture symbol or printed word by looking at and moving nose/head toward the correct symbol/word.	A computer screen projected 2 or 3 line drawings and/or printed words from a total of 24 (i.e., 4 sets of 6 symbols). For baseline, the girls were tested on their ability to select the correct drawing/word in response to its spoken word equivalent. During intervention, correct selections produced visual (smiley face) and auditory feedback ("*This is* ___."). Incorrect responses produced corrective verbal feedback (i.e., verbal naming of the correct symbol) and visual stimulation in the form of a sad face.	S1 reached criterion (80% across four consecutive sessions) on all 4 sets after 4–13 intervention sessions. At 6-month follow-up, her performance ranged from 50 to 100%. S2 only reached criterion on set 3 after 16 sessions. S3 received training on only 3 sets and reached a lower criterion (60% correct for 4 consecutive sessions) on two sets after 4 and 17 sessions. At 6-month follow-up, performance ranged from 50 to 100% correct.	Certainty of evidence for an intervention effect is inconclusive because, improving trends were evident during 5 of the 11 baselines. Inter-observer agreement was good, but treatment integrity data were not collected.
Number 8: Elefant and Lotan (2004)	One, 9 year-old girl with Rett syndrome. She had no speech, but could vocalize a few sounds. Stage of Rett syndrome was not specified.	Enhance nonverbal expression, vocalizations, and verbal ability and increase awareness of self and attention span.	A combination of music and physical therapy was provided in 2, 45-min sessions per week over several months. The program included 13 phases ranging from assessing the child's condition (Phase I) to jumping and rocking on a physical therapy ball (Phase VIII) to choosing songs from picture symbols (Phase XII). Each phase included a brief description (e.g., present the child with choices of song selection) and corresponding aims (e.g., improve communication skills).	The authors report that communication choice making abilities improved from an inability to choose objects to choosing from symbols. These new skills were said to have generalized from the classroom to the home.	Certainty of evidence for an intervention effect is inconclusive due to lack of an experimental design and objective data. The results are reported only in anecdotal narrative. Reliability of data and treatment integrity were not assessed. The dependent variables were not objectively defined and the therapeutic procedures were not described in sufficient detail to enable replication.
Number 9: Elefant and Wigram (2005)	Seven girls with Rett syndrome, 4–10 years of age. Level of speech was not reported. Six girls were in Stage 3 (pseudostationary) and 1 girl was in Stage 4 (late motor deterioration).	Requesting a song via eye gaze, nose pointing or touching picture or printed word symbols on a communication board.	Children received 20–30 min sessions of music therapy 3 mornings per week over 5 months. Follow-up data were collected 2, 6, and 12 weeks after intervention. Songs and corresponding symbols were arranged into 3 sets of 6 songs. During each session, children received opportunities to select a song from a set when presented with a choice of 2–4 symbols. When a symbol was selected, the symbols were rearranged and the child was required to confirm her original selection to validate her choice response.	The percentage of correct choice confirmations increased to 80–100% for all 7 children in 4–7 sessions for the first set of songs. Across sets, full data are presented for only one child. This child showed more rapid learning with each subsequent set. Data from the multiple baseline across sets design are also presented for only this one child. These data showed an increase in the number of songs selected and in the percentage of correct confirmation of choices from baseline to intervention in 1 of the 3 symbol sets.	Certainty of evidence for an intervention effect is inconclusive because summary data for the 7 children did not include pre-treatment levels of correct choice confirmation. While the study followed a multiple-baseline design across symbol sets, trend data are presented for only 1 of the 7 children. Trend data for this child demonstrate a convincing intervention effect for only 1 of the 3 symbol sets owing to the fact that improvements in target behaviors were evident during 2 of the 3 baselines.

"S" refers to "subject" and the following number refers to a particular subject.

[a] Studies are listed in alpha-chronological order by year of publication and then by the first author's surname.

participants, target skills, procedures, and main findings for each of the nine included studies. The final column in Table 1 explains the basis for the study's rating in terms of certainty of evidence.

3.1. Participants

Collectively, the nine studies provided intervention to a total of 31 participants. The sample size of individual studies ranged from 1 to 7 (mean = 3.4 participants per study). Two studies included only one participant [Studies 4 and 8]. Five studies had either three or four participants [Studies 1–3, 6–7]. And the two remaining studies [Studies 5 and 9] were comparatively large, with sample sizes of 6 and 7, respectively.

All 31 participants had a diagnosis of Rett syndrome, but the stage of the disorder was indicated for only 14 (45%) of the 31 participants. Most (n = 10) of these 14 children were in the third (pseudostationary) stage and the remaining 4 children were in the final (late motor deterioration) stage. Presence or absence of speech was reported for 24 (77%) of the 31 participants. The vast majority of these 24 children (n = 21, 88%) were described as having no speech. The level of speech for the other 3 children was described as (a) echoing words noncommunicatively, (b) occasionally producing a single word utterance (i.e., *mum*), or having the ability to speak. The nature and extent of the child's speech ability in this latter case was not further described.

Participant ages ranged from 2:7 to 17:0 (years:months). Precise ages for individual children were available for 22 (71%) of the 31 children, as two studies [Studies 5 and 9] provided only the age range of participants, not the ages of each individual child in the study. The mean age for these 22 children was 7:6 (years:months). Over half of these 22 participants (n = 14, 64%) were of primary school age (i.e., 5–12 years). The next largest age group (n = 6, 27%) was preschoolers aged 2:7–4:0 (years:months). The remaining two children were of high school age (15 and 17 years, respectively).

3.2. Settings

Setting descriptions were provided in six of the nine studies [i.e., all except Studies 1, 8, and 9], representing 20 (65%) of the 31 children. The interventions described in these six studies were most often undertaken in self-contained special education classrooms [Studies 2–4] or in classrooms that were not further specified [Study 7] and involved a total of 11 children. One study [Study 6, n = 3] was conducted in a hospital setting and one study [Study 5, n = 6] was conducted in the participants' homes.

3.3. Communication mode

Four different communication modes were coded across the nine studies: (a) speech or vocalization, (b) gesture, (c) graphic symbols and/or printed words, and (d) electronic or computer-based systems. Interventions to teach speech or communicative vocalizations were attempted in two studies [Studies 1 and 6] involving a total of 6 children. Various forms of communicative gestures (e.g., crossing arms, eye gaze, and pointing) were targeted in another two studies [Studies 4 and 9], involving a total of 8 children. Graphic symbols and a communication board were evaluated for the 4 children in Study 3, whereas Studies 8 and 9 also reported using symbols to enable the participating children to make choices. Speech-generating communication devices or computers that included speech output and graphic symbols or printed words were used with the remaining 12 children in Studies 2, 5, and 7.

3.4. Target skills

The communication skills targeted for intervention were coded into five pragmatic functions based on the classification system described by Sigafoos, Arthur-Kelly and Butterfield (2006): (a) imitative speech, (b) requesting access to preferred stimuli (e.g., food, drinks, toys, or songs), (c) requesting social interaction, (d) naming objects or commenting, and (e) receptive language (e.g.,

respond to requests, answer questions, receptively identify symbols). Another [General] category was used when the communication function was not clearly specified (e.g., use language, enhance nonverbal expression and verbal ability). Five studies [Studies 2–4, 7 and 9] targeted only a single communication function (e.g., requesting preferred stimuli, requesting social interaction, or receptive naming of symbols), whereas the remaining four studies [Studies 1, 5–6, and 8 targeted either multiple functions (e.g., imitative speech, naming/commenting) or general language development.

Three studies, involving 14 children, [Studies 2, 3, and 9] targeted requests for preferred food, drinks, toys, or music. One study with a single participant [Study 4] sought to teach a request for social interaction. Naming/commenting was targeted for intervention with 9 children in Studies 1 and 5. Receptive language skills; specifically, responding to requests, answering "wh"-questions, and receptively identifying symbols (e.g., *Find the symbol for mom.*) were targeted in Studies 1, 5 and 7. These three studies involved a total of 12 children. Only one study [Study 1], involving 3 children, appears to have targeted imitative speech. The two studies [Studies 6 and 8] that were coded as targeting general language development involved 4 children, but did not clearly specify the function(s) of the communication skills being taught.

3.5. Intervention procedures

Study 1 ($n = 3$ children) evaluated the effects of a comprehensive early intensive behavioral intervention program based on operant procedures and a manualized curriculum (Lovaas et al., 1981). In five studies [Studies 2–5 and 7], involving a total of 17 children, the researchers employed systematic instruction (Snell & Brown, 2006) that typically included some combination of the following operant/behavioral procedures: (a) presenting an opportunity or discriminative stimulus, (b) prompting communicative behavior if necessary, and (c) providing reinforcement/feedback on child responses. The remaining three studies [Studies 6, 8, and 9] evaluated the effects of music therapy or a combination of music therapy plus physical therapy. One of the goals of therapy in these studies was to increase communication responses. There was, however, limited detail on the actual procedures used during therapy sessions to achieve the goal of improved communication.

3.6. Study designs

Study designs were classified as experimental, pre-experimental, or unclear. The only experimental design employed in this set of studies was the multiple-baseline/multiple-probe design (Barlow, Nock, & Hersen, 2009; Kennedy, 2005). Three studies [Studies 2, 3, and 7], involving a total of 10 children used the multiple-baseline/multiple-probe design to evaluate the effects of the intervention on operationally defined target behaviors. Two additional studies [Studies 5 and 9] claimed to have employed a multiple-baseline or multiple-probe design, but it was not clear from the manner in which data were presented that this was in fact the case. For two studies, the design was classified as pre-experimental because the study involved either pre-post measures without a control group [Study 1] or a simple A-B design [Study 4]. The design for the remaining two studies [Studies 6 and 8] was not clear due to insufficient detail.

3.7. Follow-up and generalization

Only two studies [Studies 7 and 9] reported on participants' use of the acquired communication skills following intervention. The length of follow-up was either 3 months [Study 9] or 6 months [Study 7]. Only one study [Study 2] included data on generalization of intervention effects outside of the initial training context. In this study, training occurred in the classroom and generalization of requesting was assessed in the school lunchroom and home. Generalization probes in the lunchroom were implemented with all 3 children using other adults and peers as communicative partners. Generalization probes in the home were implemented for 2 of the 3 children using the child's mother as the communicative partner.

3.8. Reliability of data and treatment integrity

Most studies [Studies 1, 2, 4, 6, 8, and 9] failed to report on the reliability of data collection with respect to the dependent variables, such as by collecting inter-observer agreement (IOA). IOA would have been relevant to all of these studies except Study 2, where data were recorded automatically through electronic equipment. In contrast, Studies 3, 5, and 7 did collect IOA data and in all cases the resulting percentages of agreement were at or above the generally accepted standard of 80% (Barlow et al., 2009; Kennedy, 2005). Procedures to assess treatment integrity data were evident only in Study 3. In this study the researchers assessed the extent to which the interventionist implemented the procedures as written during one randomly selected trial per session. The results showed that the procedures were implemented with 97–100% accuracy.

3.9. Outcomes

Outcomes were classified as either showing progress or no progress based on the data presented by the authors. In many cases, data were presented in narrative form only and so judgments about improvement were necessarily more subjective and based on the author's narrative summary. In addition, some children showed improvement in certain areas, but losses in other areas. For example, in Study 1, one child (S1) was said to have made gains in moving from "echoing words noncommunicatively to speaking single words meaningfully", but "these gains were offset by losses in other areas." (Smith et al., 1995, p. 319). Because the gains were in the area of speech, we coded this an instance of improvement. Overall, at least some positive gains in the targeted communicative skills were reported for the majority of children (n = 26, 84%). The exceptions are Studies 1, where 2 of the 3 children did not make progress; Study 3, where 2 of the 4 children did not learn the initially targeted communication skill; and Study 4, where the participating child did not learn to request social interaction using an arm-folding gesture.

3.10. Certainty of evidence

The certainty of evidence for an intervention effect was rated as conclusive only for Study 2. For the other eight studies, the certainty of evidence for an intervention effect was judged to be inconclusive. These inconclusive ratings stemmed from reliance on pre-experimental designs [Studies 1 and 4] or lack of objective description of methods and failure to present sufficient data [Studies 6 and 8]. In other cases, [Studies 5 and 9], complete data from the multiple-baseline/multiple-probe designs were lacking making it impossible to visually inspect the trends for all of the children and across all of the phases of the two respective studies. In the two remaining studies [Study 3 and 7], visual inspection was possible, but this revealed considerable overlap/variability of data across the baseline and intervention phases and/or improving trends during baseline. These trends compromised the integrity of the multiple-baseline/multiple-probe design and thus rendered the results inconclusive.

4. Discussion

Our systematic search yielded nine studies on communication intervention for individuals with Rett syndrome. All nine of these studies were published between the years 1995 and 2005. Summaries of these studies revealed that, overall; the existing literature base is perhaps best described as limited. The limitations are evident in terms of the scope and quality of the existing corpus of studies.

In terms of scope, the current database must be considered limited because of the sheer paucity of studies (n = 9) and the relatively few number of participants (n = 31). These 31 participants were fairly homogeneous in terms of age ranges and ability levels. Indeed, the 31 children would seem to fit the pattern for classical Rett syndrome as described by Ellaway and Christodoulou (2001), but the classic picture does not necessarily represent the Rett syndrome population as a whole (Matson et al., 2008). Hagberg and Gillberg (1993), for example, described five Rett syndrome variants, ranging from individuals who retain speech to cases where the regression occurs later and the severity of impairment is milder. One might suspect that individuals with preserved speech and more mild

impairments would be more responsive to communication intervention than the 31 children who received treatment in these nine studies.

One might also expect that the effects of communication intervention would vary depending on the stage of Rett syndrome. For example, children in the rapid destructive phase might be less responsive to intervention than children who have stabilized in the final fourth stage of the disorder. Unfortunately, the existing database was too limited to determine whether there were any such stage-related effects. Our inability to examine stage-related effects stems from the fact that the stage of Rett syndrome was reported for less than half (45%) of the participants. In addition, none of these existing studies was specifically designed to investigate the relative effects of communication intervention for children in different stages of the disorder. Future research should investigate this question as it would seem plausible that there might be some important stage-by-treatment interactions. Recognition of any such interactions would perhaps enable clinicians to decide if, when, and how best to intervene with children who are at different stages in this progressive disorder.

In terms of methodological quality, perhaps the most important limitation is that nearly half of the studies appeared to lack a recognized experimental design. This general lack of experimental design, combined with the many additional methodological limitations that we noted (e.g., lack of procedural detail, limited generalization and follow-up, lack of reliability and treatment integrity data), meant that the certainty of evidence was inconclusive for all but one study. Thus the reports of positive outcomes for 26 (84%) of the 31 participants must be interpreted with extreme caution.

In terms of the main aim of this paper, we must regrettably conclude that the results of this review offer little that could be used to advance evidence-based practice in communication intervention for individuals with Rett syndrome. There is simply insufficient evidence to support any particular approach to intervention for this population. The best we can offer is to suggest that clinicians might want to begin by teaching a simple requesting response following the lead of Van Acker and Grant (1995). Given the nature of motor impairment in Rett syndrome it would seem critical to ensure that the targeted requesting response is within the child's physical capabilities. For individuals with the extremely limited motor abilities of classic Rett syndrome, this will most likely require the clinician to identify some very simple motor act, such as touching a switch, rather than attempting to teach speech or gestures. While this recommendation cannot be considered empirically validated for this population at the present time, there would seem to be little risk of harm from trying such an approach.

We have been more successful in achieving our second aim, which was to identify gaps in the literature. In general, our review identified pertinent gaps in terms of the range of procedures evaluated and the range of communication modes and functions targeted for intervention. More specifically, the studies conducted so far have investigated a rather restricted range of procedures and targeted only a few communication modes/functions. Furthermore, none of the studies appeared to have included pre-treatment assessments to inform the intervention. Bridging this latter gap could be critical to developing more effective interventions. Assessment data reported by Sigafoos, Woodyatt, Tucker, Roberts-Pennell and Pittendreigh (2000), for example, suggested that some children with Rett syndrome retain prelinguistic acts (e.g., facial expressions, body movements). In such cases, an effective intervention might involve teaching partners to shape such prelinguistic acts into intentional forms of communication (Sigafoos et al., 2006). Similarly, a pre-treatment assessment of motor skills may enable clinicians to identify motor acts that the person could use to control a speech-generating device via assistive technology (Lancioni et al., 2006). While research along these lines might someday yield new and more effective approaches, the evidence base supporting current approaches to communication intervention for individuals with Rett syndrome remains inconclusive.

Appendix A

A.1. Included studies

1. Smith, T., Klevstrand, M., & Lovaas, O. I. (1995). Behavioral treatment of Rett's disorder: Ineffectiveness in three cases. *American Journal on Mental Retardation, 100*, 317-322.

2. Van Acker, R., & Grant, S. H. (1995). An effective computer-based requesting system for persons with Rett syndrome. *Journal of Childhood Communication Disorders, 16*, 31-38.
3. Sigafoos, J., Laurie, S., & Pennell, D. (1996). Teaching children with Rett syndrome to request preferred objects using aided communication: Two preliminary studies. *Augmentative and Alternative Communication, 12*, 88-96.
4. Evans, I. M., & Meyer, L. H. (1999). Modifying adult interactional style as positive behavioral intervention for a child with Rett syndrome. *Journal of Intellectual and Developmental Disability, 24*, 191-205.
5. Koppenhaver, D. A., Erickson, K. A., Harris, B., McLennan, J., Skotko, B. G., & Newton, R. A. (2001). Storybook-based communication intervention for girls with Rett syndrome and their mothers. *Disability and Rehabilitation, 23*, 149-159.
6. Yasuhara, A., & Sugiyama, Y. (2001). Music therapy for children with Rett syndrome. *Brain and Development, 23*(Suppl. 1), S82-S84.
7. Hetzroni, O., Rubin, C., & Konkol, O. (2002). The use of assistive technology for symbol identification by children with Rett syndrome. *Journal of Intellectual & Developmental Disability, 27*, 57-71.
8. Elefant, C., & Lotan, M. (2004). Rett syndrome: Dual intervention — music and physical therapy. *Nordic Journal of Music Therapy, 13*, 172-182.
9. Elefant, C., & Wigram, T. (2005). Learning ability in children with Rett syndrome. *Brain and Development, 27*(Suppl. 1), S97-S101.

Appendix B

B.1. Excluded studies [and reason for exclusion]

1. Baptista, P. M., Mercadante, M. T., Macedo, E. C., & Schwartzman, J. S. (2006). Cognitive performance in Rett syndrome: a pilot study using eyetracking technology. *Journal of Intellectual Disability Research, 50*, 662-666. [Assessment study].
2. Brady, N. C., & Halle, J. W. (1997). Functional analysis of communicative behaviors. *Focus on Autism and Other Developmental Disorders, 12*, 95-104. [Assessment study].
3. Burford, B., & Trevarthen, C. (1997). Evoking communication in Rett syndrome: comparisons with conversations and games in mother-infant interaction. *European Child and Adolescent Psychiatry, 6*, 26-30. [Review].
4. Dahlgren Sandberg, A., Ehlers, S., Hagberg, B., & Gillberg, C. (2000). The Rett syndrome complex: Communication functions in relation to developmental level and autistic features. *Autism, 4*, 249-267. [Assessment study].
5. Fyfe, S., Downs, J., McIlroy, O., Burford, B., Lister, J., Reilly, S., Laurvick, C. L., Philippe, C., Msall, M., Kaufmann, W. E., Ellaway, C., & Leonard, H. (2007). Development of a video-based evaluation tool in Rett syndrome. *Journal of Autism and Developmental Disorders, 37*, 1636-1646. [Assessment study].
6. Garber, N., & Veydt, N. (1990). Rett syndrome: A longitudinal developmental case report. *Journal of Communication Disorders, 23*, 61-73. [Assessment study].
7. Garber, N., & Veydt, N. (1992). Speech and language development in Rett syndrome: Literature review and case study. *Australian Journal of Human Communication Disorders, 20*, 51-62. [Review with descriptive case study].
8. Gillberg, C. (1997). Communication in Rett syndrome complex. *European Child and Adolescent Psychiatry, 6*, 21-22. [Descriptive study].
9. Hetzroni, O. E., & Rubin, C. (2006). Identifying patterns of communicative behaviors in girls with Rett syndrome. *Augmentative and Alternative Communication, 22*, 48-61. [Assessment study].
10. Katsiyannis, A., Ellenburg, J. S., Action, O. M., & Torrey, G. (2001). Addressing the needs of students with Rett syndrome. *Exceptional Children, 33*, 74-78. [Review].
11. Koppenhaver, D. A., Erickson, K. A., & Skotko, B. G. (2001). Supporting communication of girls with Rett syndrome and their mothers in story book reading. *International Journal of Disability, Development and Education, 48*, 395-410. [Duplication of Study #5].

12. Lava, J., Slotte, A., Jochym-Nygren, M., Van Doorn, J., & Witt Engerstrom, I. (2006). Communication and eating proficiency in 125 females with Rett syndrome: The Swedish Rett Center Survey. *Disability and Rehabilitation, 28*, 1267-1279. [Descriptive study].
13. Raglio, A., Traficante, D., & Oasi, O. (2006). A coding scheme for the evaluation of the relationship in music therapy sessions. *Psychological Reports, 99*, 85-90. [Assessment study].
14. Ryan, D., McGregor, F., Akermanis, M., Southwell, K., Ramke, M., & Woodyatt, G. (2004). Facilitating communication in children with multiple disabilities: three cases of girls with Rett syndrome. *Disability and Rehabilitation, 26*, 1268-1277. [Assessment study].
15. Skotko, B. G., Koppenhaver, D. A., & Erickson, K. A. (2004). Parent reading behaviors and communication outcomes in girls with Rett syndrome. *Exceptional Children, 70*, 145-166. [Duplication of Study #5].
16. Uchino, J., Suzuki, M., Hoshino, K., Nomura, Y., & Segawa, M. (2001). Development of language in Rett syndrome. *Brain and Development, 23* (Suppl. 1), S233-S235. [Assessment study].
17. von Tetzchner, S. (1997). Communication skills among females with Rett syndrome. *European Child and Adolescent Psychiatry, 6*, 33-37. [Descriptive study].
18. Wigram, T., & Lawrence, M. (2005). Music therapy as a tool for assessing hand use and communicativeness in children with Rett syndrome. *Brain and Development, 27*(Suppl. 1), S95-S96. [Assessment study].
19. Woodyatt, G., & Ozanne, A. (1992a). A longitudinal study of cognitive skills and communication behaviours in children with Rett syndrome. *Journal of Intellectual Disability Research, 37*, 419-435. [Assessment study].
20. Woodyatt, G., & Ozanne, A. (1992b). Communication abilities in Rett syndrome. *Journal of Autism and Developmental Disorders, 22*, 155-173. [Assessment study].
21. Woodyatt, G., & Ozanne, A. (1992c). Communication abilities in a case of Rett syndrome. *Journal of Intellectual Disability Research, 36*, 83-92. [Descriptive study].
22. Woodyatt, G., & Ozanne, A. (1994). Intentionality and communication in four children with Rett syndrome. *Australia and New Zealand Journal of Developmental Disabilities, 19*, 173-183. [Assessment study].
23. Woodyatt, G., & Ozanne, A. (1997). Rett syndrome (RS) and profound intellectual disability: cognitive and communication similarities and differences. *European Child and Adolescent Psychiatry, 6*, 31-32. [Assessment study].

References

Amir, R. E., Van den Veyver, I. B., Wan, M., Tran, C. Q., Francke, U., & Zoghbi, H. Y. (1999). Rett syndrome is caused by mutations in X-linked MECP2, encoding methyl-CpG-binding protein 2. *Nature Genetics, 23*, 185–188.

Barlow, D. H., Nock, M. K., & Hersen, M. (2009). *Single case experimental designs: Strategies for studying behavior change*. Boston, MA: Pearson Education Inc.

Burford, B., & Trevarthen, C. (1997). Evoking communication in Rett syndrome: Comparisons with conversations and games in mother-infant interaction. *European Child and Adolescent Psychiatry, 6*(Suppl. 1), S26–S30.

Dunn, H. G. (2001). Importance of Rett syndrome in child neurology. *Brain and Development, 23*(Suppl. 1), S38–S43.

Ellaway, C., & Christodoulou, J. (2001). Rett syndrome: Clinical characteristics and recent genetic advances. *Disability and Rehabilitation, 23*, 98–106.

Hagberg, B., & Gillberg, C. (1993). Rett variants – rettoid phenotypes. In B. Hagberg, M. Anvert, & J. Wahlstrom (Eds.), *Rett syndrome: Clinical and biological aspects* (pp. 40–60). London: MacKeith Press.

Hagberg, B., & Witt-Engerström, I. (1986). Rett syndrome: A suggested staging system for describing impairment profile with increasing age toward adolescence. *American Journal of Medical Genetics, 24*, 47–59.

Kennedy, C. H. (2005). *Single-case designs for educational research*. Boston: Allyn & Bacon.

Koppenhaver, D. A., Erickson, K. A., Harris, B., McLennan, J., Skotko, B. G., & Newton, R. A. (2001b). Storybook-based communication intervention for girls with Rett syndrome and their mothers. *Disability and Rehabilitation, 23*, 149–159.

Koppenhaver, D. A., Erickson, K. A., & Skotko, B. G. (2001a). Supporting communication of girls with Rett syndrome and their mothers in storybook reading. *International Journal of Disability, Development and Education, 48*, 395–410.

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., Sigafoos, J., Oliva, D., & Baccani, S. (2006). Teaching 'Yes' and 'No' responses to children with multiple disabilities through a program including microswitches linked to a vocal output device. *Perceptual and Motor Skills, 102*, 51–61.

Leonard, H., Bower, C., & English, D. (1997). The prevalence and incidence of Rett syndrome in Australia. *European Journal of Child and Adolescent Psychiatry, 6*, S8–S10.

Lovaas, O. I., Ackerman, A. B., Alexander, D., Firestone, P., Perkins, J., & Young, D. (1981). *Teaching developmentally disabled children. The ME book*. Austin, TX: Pro-Ed.

Matson, J. L., Fodstad, J. C., & Boisjoli, J. A. (2008). Nosology and diagnosis of Rett syndrome. *Research in Autism Spectrum Disorders, 2*, 601–611.

Millar, D. C., Light, J. C., & Schlosser, R. W. (2006). The impact of augmentative and alternative communication intervention on the speech production of individuals with developmental disabilities: A research review. *Journal of Speech, Language, and Hearing Research, 49*, 248–264.

Sigafoos, J., Arthur-Kelly, M., & Butterfield, N. (2006). *Enhancing everyday communication for children with disabilities*. Baltimore: Paul H Brookes Publishing Co.

Sigafoos, J., & Woodyatt, G. (1996). Educational implications of Rett syndrome. *European Journal on Mental Disability, 3*, 19-28. Reprinted in *Learning Disability Bulletin, 106*, Sept 1997.

Sigafoos, J., Woodyatt, G., Tucker, M., Roberts-Pennell, D., & Pittendreigh, N. (2000). Assessment of potential communicative acts in three individuals with Rett syndrome. *Journal of Developmental and Physical Disabilities, 12*, 203–216.

Skotko, B. G., Koppenhaver, D. A., & Erickson, K. A. (2004). Parent reading behaviors and communication outcomes in girls with Rett syndrome. *Exceptional Children, 70*, 145–166.

Smith, T., Klevstrand, M., & Lovaas, O. I. (1995). Behavioral treatment of Rett's Disorder: Ineffectiveness in three cases. *American Journal on Mental Retardation, 100*, 317–322.

Snell, M. E., & Brown, F. (2006). *Instruction of students with severe disabilities* (6th ed.). Upper Saddle River, NJ: Pearson.

Van Acker, R., & Grant, S. H. (1995). An effective computer-based requesting system for persons with Rett syndrome. *Journal of Childhood Communication Disorders, 16*, 31–38.