The acquisition of stimulus equivalence in individuals with fragile X syndrome

S. S. Hall, G. M. DeBernardis & A. L. Reiss

Department of Psychiatry & Behavioural Sciences, Stanford University, Stanford, CA, USA

Abstract

Background Few studies have employed stimulus equivalence procedures to teach individuals with intellectual disabilities (IDs) new skills. To date, no studies of stimulus equivalence have been conducted in individuals with fragile X syndrome (FXS), the most common known cause of inherited ID.

Method Five adolescents with FXS were taught basic math and geography skills by using a computerized stimulus equivalence training programme administered over 2 days in 2-h sessions.

Results Four of the five participants learned the math relations, with one participant demonstrating stimulus equivalence at post-test. Three of the five participants learned the geography relations, with all three of these participants demonstrating stimulus equivalence at post-test.

Conclusions These data indicate that computerized stimulus equivalence procedures, conducted in time-limited sessions, may help individuals with FXS learn new skills. Hypotheses concerning the failure of some participants to learn the training relations and to demonstrate stimulus equivalence at post-test are discussed.

Keywords fragile X syndrome, stimulus equivalence

Introduction

Since its inception in the early 1970s, stimulus equivalence procedures have been successfully employed to teach a variety of new skills to individuals with intellectual disabilities (IDs) (Sidman 1994). Reading skills (Mackay 1985), math skills (Maydak et al. 1995), money skills (Trace et al. 1977; Cuvo et al. 1978) and geography skills (LeBlanc et al. 2003) have all been taught to individuals with a variety of diagnoses including Down syndrome (Sidman & Cresson 1973) and autism (LeBlanc et al. 2003), and individuals with non-specific IDs (Devany et al. 1986). The basic procedure involves presenting three sets of related stimuli (commonly labelled ‘A’, ‘B’ and ‘C’, respectively) in successive blocks of matching-to-sample training trials. Most commonly, individuals are first taught to match the B stimuli to the A stimuli (A = B training). Next, individuals are taught to match the C stimuli to the B stimuli (B = C training). Studies have shown that after this training, individuals are subsequently able to match a number of additional relations (e.g. C = A, A = C, B = A, C = B) successfully without explicit training. Given this positive outcome, the stimulus equivalence paradigm offers educators a useful method to teach individuals...
a variety of new skills quickly and efficiently. Indeed, the creation of stimulus equivalence classes has been considered a rudimentary form of concept formation (Sidman 1994).

In their seminal paper on stimulus equivalence, Sidman & Cresson (1973) presented three sets of related stimuli to two young boys with Down syndrome. The stimuli were dictated words (A), pictures (B) and printed words (C). The participants were first trained to match the pictures to the dictated words (A = B training). Next, the participants were trained to match the printed words to the dictated words (A = C training). After training only 40 relations between the stimuli (20 A = B relations and 20 A = C relations), not only were the participants subsequently able to produce the correct oral responses to the printed word (C = A, reading aloud), but they were also able to choose pictures corresponding to the printed word (C = B, reading comprehension). In fact, in subsequent tests, 80 new instances of correspondence had emerged indirectly from the original training procedure. Sidman (1994) has suggested that the ability of participants to perform these additional relations is analogous to the mathematical properties of symmetry (if A = B, then B = A) and transitivity (if A = B and B = C, then A = C). Indeed, once participants can pass the C = A test (a relation involving both symmetry and transitivity), they can be said to have formed an equivalence class.

Numerous studies of stimulus equivalence have been conducted since then. Maydak et al. (1995), for example, trained two adults with IDs, aged 30 and 49 years, to match the numerals 1–5 to dots making up the quantities and to their corresponding numbers. All stimuli were presented on a touch-sensitive computer screen, with pairs or trios of comparison stimuli appearing below the sample stimulus. After a series of training and testing blocks, both participants were able to identify the correct numbers when they were given drawings of the stimuli in an oral test.

Math skills were also trained to seven students aged 11–13 years who had difficulty on fraction and decimal tasks (Lynch & Cuvo 1995). All stimuli were presented on a computer screen and arranged in six sets (A, B, C, D, X, Y), with 12 stimuli in the A, B and C sets and eight stimuli in the D, X and Y sets. Set B consisted of pictorial representations, which were shown as shaded portions of grids divided into 100 subunits. Sets C and Y contained printed numerals as decimals (e.g. 0.20), while sets A, D and X consisted of printed fractions (e.g. 1/5). Post-test scores demonstrated that all participants learned the relations between the stimuli and demonstrated symmetry and transitivity.

In a more recent study of stimulus equivalence, LeBlanc et al. (2003) trained basic geography skills to two children diagnosed with autism, aged 6 and 13 years. Here, the stimuli consisted of state shapes, state names and capitals from three regions of the United States. Children received training in time-limited sessions conducted over several days with no more than two, 20- to 30-min sessions conducted per day. Both children mastered the trained relations and demonstrated stimulus equivalence after receiving 360 and 550 training trials, respectively.

Despite a large amount of literature attesting to the effectiveness of stimulus equivalence in teaching new skills to children with IDs, to our knowledge, stimulus equivalence has never been employed to teach new skills to children with fragile X syndrome (FXS), the most common inherited form of ID. FXS affects approximately 1 in 4000 males and 1 in 8000 females in the general population (Crawford et al. 1999), and is caused by a mutation to the FMR1 gene on the long arm of the X chromosome at Xq27.3 (Verkerk et al. 1991). In unaffected individuals, the gene contains a sequence of CGG nucleotides that repeats approximately 5–45 times. If the sequence expands to approximately 200 repeats or more (the ‘full mutation’), methylation of the promoter region of the gene occurs and production of the ‘Fragile X Mental Retardation Protein’ (FMRP), the protein product of the FMR1 gene, ceases. FMRP is thought to participate actively in the translational machinery that converts messenger RNA into protein (Brown et al. 2001). Low levels of FMRP therefore increases the risk for the physical, cognitive and behavioural manifestations of the disorder (Taylor et al. 1994). Females, who have the mutation on only one of their two X chromosomes, consequently have higher levels of FMRP than males with FXS and are therefore less affected by the disorder.

The characteristic profile of intellectual abilities associated with the full mutation includes weaknesses in the following domains: visual memory and perception, manipulation of visual–spatial relationships among objects, visual–motor coordination, processing of sequential information, processing of
Stimulus equivalence in fragile X

S. S. Hall et al.

© 2006 The Authors. Journal Compilation © 2006 Blackwell Publishing Ltd

Methods

Participants

The participants were five individuals (one female, four males) aged between 12 and 19 years and diagnosed with FXS. Table 1 shows the participants’ characteristics. Full scale IQ (FSIQ) as measured by the WISC-III or WAIS-III tests ranged from 40 to 65, verbal IQ ranged from 46 to 69, and performance IQ ranged from 46 to 68. The participants were recruited during the biannual National Fragile X Foundation Conference by asking parents attending the conference if they would like their child to learn some new skills using a computer-based teaching programme.

Procedure

The participants were required to sit down in front of a laptop computer and were then shown how to use the computer mouse. Instructions were delivered orally as follows: ‘Today I am going to work with you to learn some math and geography. First, you will see a word or picture appear on the computer screen with a question below. I will read the question out loud to you. You will need to click on the word or picture with the computer mouse. Three boxes will then appear below, one of which will be the correct answer to the question. Your task will be to click on the box that contains the correct answer. Another question will then appear and so on. You will have as much time as you need to select each answer. Unfortunately, at this stage, I cannot tell you whether you have got each question right or wrong. Please try to do the best you can.’ On the first two trials, participants were prompted if they appeared not to understand the procedure.

Two conditions were employed: geography and math. In the geography condition, the stimuli consisted of state names (A), state pictures (B) and state capitals (C), similar to the stimuli employed by LeBlanc et al. (2003). In the math condition, the stimuli consisted of fractions (A), pie charts (B) and decimals (C) (see Fig. 1). Each set consisted of three stimuli. The participants were trained in both conditions, with condition order randomized across participants with three phases to each condition: pre-test, training and post-test. Sessions were conducted over 2 days in two sessions lasting approximately 2 h each.

Table 1  Participant demographics

<table>
<thead>
<tr>
<th>Participant</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Full scale IQ</th>
<th>Verbal IQ</th>
<th>Performance IQ</th>
</tr>
</thead>
<tbody>
<tr>
<td>P1</td>
<td>M</td>
<td>13</td>
<td>55</td>
<td>64</td>
<td>54</td>
</tr>
<tr>
<td>P2</td>
<td>M</td>
<td>14</td>
<td>65</td>
<td>69</td>
<td>68</td>
</tr>
<tr>
<td>P3</td>
<td>F</td>
<td>12</td>
<td>50</td>
<td>56</td>
<td>53</td>
</tr>
<tr>
<td>P4</td>
<td>M</td>
<td>15</td>
<td>40</td>
<td>46</td>
<td>46</td>
</tr>
<tr>
<td>P5</td>
<td>M</td>
<td>19</td>
<td>40</td>
<td>46</td>
<td>46</td>
</tr>
</tbody>
</table>
Pre-test phase

The pre-test phase consisted of C = A (equivalence), A = C (transitivity), B = A (symmetry) and C = B (symmetry) tests, respectively, each containing 12 trials. At the beginning of each trial, a sample stimulus was displayed at the top of the computer screen with accompanying text positioned directly underneath the stimulus (see Table 2). Once the participant clicked on the sample stimulus using the computer mouse, three comparison stimuli were then displayed directly below the sample stimulus. A click on one of the comparison stimuli led to a 3-s inter-trial interval (ITI) during which the screen remained blank. The next trial was then displayed. The order of presentation of the sample stimuli and comparison stimuli was randomized across trials. Once all 12 trials had been completed, the percentage of correct trials obtained by the participant was displayed briefly in the middle of the screen. All participants obtained scores of 66.6% or less on each of the pre-tests.

Training phase

In the training phase, each participant received A = B, B = C and A = B & B = C (mixed) training, respectively. In this phase, if an incorrect comparison stimulus was selected on a trial, the comparison stimuli disappeared and another trial was instituted with the same sample stimulus being presented on the screen. The participant was instructed to ‘try again’ by clicking the sample stimulus in order to display the comparison stimulus.

Figure 1 Stimuli employed for the geography (upper panel) and math (lower panel) skills.

Table 2 Instructions displayed on the computer screen and the sequence of training and testing phases employed

<table>
<thead>
<tr>
<th>Phase</th>
<th>Relation</th>
<th>Geography condition</th>
<th>Math condition</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-test</td>
<td>C = A</td>
<td>‘__is the capital for which state?’</td>
<td>‘__is equal to which fraction?’</td>
</tr>
<tr>
<td></td>
<td>A = C</td>
<td>‘__is the state for which capital?’</td>
<td>‘__is equal to which decimal?’</td>
</tr>
<tr>
<td></td>
<td>B = A</td>
<td>‘What is the name of the red state?’</td>
<td>‘Which fraction is equal to the red piece?’</td>
</tr>
<tr>
<td></td>
<td>C = B</td>
<td>‘__is the capital of which red state?’</td>
<td>‘__is the decimal of which red piece?’</td>
</tr>
<tr>
<td>Training</td>
<td>A = B</td>
<td>‘__is the name of which red state?’</td>
<td>‘__is the fraction of which red piece!’</td>
</tr>
<tr>
<td></td>
<td>B = C</td>
<td>‘What is the capital of the red state?’</td>
<td>‘Which decimal is equal to the red piece?’</td>
</tr>
<tr>
<td></td>
<td>A = B &amp;</td>
<td>Alternating questions</td>
<td>Alternating questions</td>
</tr>
<tr>
<td></td>
<td>B = C (mixed)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Post-test</td>
<td>C = A</td>
<td>‘__is the capital for which state?’</td>
<td>‘__is equal to which fraction?’</td>
</tr>
<tr>
<td></td>
<td>A = C</td>
<td>‘__is the state for which capital?’</td>
<td>‘__is equal to which decimal?’</td>
</tr>
<tr>
<td></td>
<td>B = A</td>
<td>‘What is the name of the red state?’</td>
<td>‘Which fraction is equal to the red piece?’</td>
</tr>
<tr>
<td></td>
<td>C = B</td>
<td>‘__is the capital of which red state?’</td>
<td>‘__is the decimal of which red piece?’</td>
</tr>
</tbody>
</table>
stimuli. When the participant selected the correct comparison stimulus, a computer-generated tone sounded, and the participant was given encouraging feedback from experimenter (e.g. ‘Well done. Good job’). A 3-s ITI was then instituted and the next trial began. The stimuli were presented in blocks of 12 preprogrammed trials and the order of presentation of the sample and comparison stimuli was randomized across trials within each block. The participants continued to learn a relation (i.e. $A = B$) until they had obtained a score of 92% correct or higher in a block of trials. If the participant was unable to reach this criterion over 20 blocks of trials, training was stopped and the post-test phase was not administered for that participant. The number of training trials required for participants to reach criterion for each training relation was recorded directly by the computer.

**Post-test phase**

The post-test consisted of $C = A$ (equivalence), $A = C$ (transitivity), $B = A$ (symmetry) and $C = B$ (symmetry) tests, respectively. This phase was identical to the pre-test phase, i.e. no feedback was presented to the participant concerning correct or incorrect responses.

**Results**

Table 3 shows the number of trials that each participant received in the training phases of each relation for each condition. Four of the five participants successfully learned the trained math relations, requiring between 64 and 847 trials to complete the training. The remaining participant (P3) failed to reach criterion responding after receiving 833 $B = C$ training trials in this condition. Three of the five participants successfully learned the trained geography relations, requiring between 80 and 523 trials to complete the training. Of the remaining two participants, one participant (P4) failed to reach criterion responding in this condition after receiving 695 $A = B$ training trials, while the other participant (P5) failed to reach criterion responding after receiving 449 $B = C$ training trials.

Figure 2 shows the children’s performance in the geography and math conditions on the pre- and post-tests.

In the geography condition, all three participants who had successfully learned the trained geography relations improved their performance on the post-tests, obtaining 100% correct responding on some of these tests. In the math condition, the scores of one of the four participants who had successfully learned the trained math relations improved on the post-test. The remaining three participants (P2, P4 and P5) improved their performance on the $B = A$ relation only.

Participant 2 required very few training trials to learn the trained relations in both the geography and math conditions (requiring a total of 80 and 64 trials, respectively), and this participant also had the highest FSIQ (i.e. 65). Interestingly, this participant performed poorly in the post-test in the math condition by consistently substituting the $\frac{1}{2}$ and $\frac{1}{4}$ stimuli. The three participants who did not complete the training in either the math or geography condition (P3, P4 and P5) had the lowest FSIQ scores (50, 40 and 40).

<table>
<thead>
<tr>
<th>Table 3</th>
<th>Number of training trials required in each training relation for each participant (P)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Geography condition</td>
</tr>
<tr>
<td></td>
<td>$A = B$</td>
</tr>
<tr>
<td>P1</td>
<td>336</td>
</tr>
<tr>
<td>P2</td>
<td>26</td>
</tr>
<tr>
<td>P3</td>
<td>75</td>
</tr>
<tr>
<td>P4</td>
<td>695*</td>
</tr>
<tr>
<td>P5</td>
<td>351</td>
</tr>
</tbody>
</table>

* Did not reach criterion.
Discussion

Children with FXS have been shown to have weaknesses in math and spatial skills. In this study, we specifically targeted those skills for intervention by training five adolescents with FXS on basic geography and math skills, using a stimulus equivalence training procedure. Four of the five participants successfully learned the math training relations, but only one showed improvement on subsequent symmetry and transitivity tests. Three of the five participants successfully learned the geography training relations, and all three of these participants showed improvement on subsequent symmetry and transitivity tests.
Two of the participants in the geography condition and one of the participants in the math condition failed to learn the training relations after receiving large numbers of training trials on specific relations. These data support previous studies showing that children with FXS may have specific difficulties learning math and visual–spatial skills.

This study is unique in two respects. First, to our knowledge, this is the first investigation to implement a stimulus equivalence procedure on a group of children with FXS. By studying a relatively homogenous group of individuals with known weaknesses in acquiring new skills, we wanted to determine whether a stimulus equivalence training procedure could aid the acquisition of skills in this population. Second, we trained participants across two skill areas to allow us to compare skill acquisition in each condition. Interestingly, only two of the five participants in this study were able to learn the trained relations in both skill areas. These participants also had the highest IQ scores. Of the remaining three participants, one participant demonstrated stimulus equivalence on the geography stimuli, but was unable to learn the trained math relations that would permit the testing of new relations emerging through equivalence. The other two participants were able to learn the trained math relations, but these participants were unable to learn the trained geography relations.

Several factors may have accounted for the varied performance in the two conditions. First, our participants received the training sessions over a limited time period, i.e. 2-h training sessions conducted over 2 days. In previous studies, participants received varied amounts of training. In the Sidman & Cresson (1973) study, sessions were conducted one to three times per week for 1–7 months. In the LeBlanc et al. (2003) study, participants received up to two 20- to 30-min sessions per day, with two to five sessions being administered per week. In the Lynch & Cuvo (1995) study, sessions were 20 min long with approximately 16 sessions being conducted 2–5 days per week over a period of 5 weeks. Finally, in the Maydak et al. (1995) study, sessions lasted 10–15 min and were conducted once or twice a day, three to five times per week. Future research studies should determine the optimal number of sessions and/or session lengths that participants may require to learn these skills successfully. Clearly, however, if some individuals require an extremely large number of training trials and/or sessions to learn these skills, and yet fail to show stimulus equivalence at post-test, other training strategies may need to be investigated.

A second caveat of the present study concerns the fact that feedback was not provided in the post-tests. LeBlanc et al. (2003) evaluated the effects of three testing conditions: providing continuous feedback during pre-test, training and post-test phases, providing feedback during training phases only, and providing continuous feedback during training with interspersed feedback during post-test phases. Through direct comparison of the three testing procedures, LeBlanc et al. found no substantial differences in performance. Given this finding, we did not implement feedback in either the pre-test or post-test phases. It is possible, however, that poor performance in the post-test phase in some participants may have occurred because of extinction from the absence of feedback. This explanation does not seem likely in most cases, however (perhaps with the exception of P1), because the graphs in Fig. 2 do not generally indicate a gradual reduction in the accuracy of responding across the sequence of trial blocks. In fact, the data from P2, P4 and P5 seem to indicate some tentative emergence of symmetric B = A from trained A = B relations in the math condition, and this was three blocks into the post-test. It is interesting that all three participants (P2, P4 and P5) seemed to demonstrate symmetry with B = A relations in the math condition but not with C = B relations because it suggests that their performance was not due to an inability to derive symmetry per se. In view of the apparent visual–spatial difficulties of individuals with FXS, it could be speculated that these participants may have found it difficult to discriminate between the shapes when they were presented simultaneously as comparison stimuli. However, this does not appear to be the case for P2 because he accomplished the same task with the geographic shapes. It should be noted that for this participant, poor performance during the post-test in the math condition was due to a perseveration on error responses (substituting 1/2; and 1/3). It should also be pointed out that the geography test involves verbal labelling as well as visual–spatial skills. Given that verbal-based skills may be spared in females with FXS, this may have been why the only female in our sample (P3) successfully completed the geography relations but could not complete the math
relations, despite receiving a large number of trials in that condition.

A final caveat of the study is that the feedback and instructions we supplied during the training sessions were limited to a computer-generated tone and a brief comment delivered by the experimenter. Other studies have employed a variety of reinforcers to maintain the participants’ responding. For example, in the Devany et al. (1986) study, the experimenter asked the child to ‘touch the one that goes with this one’ as the experimenter pointed to the sample. Reinforcement included praise, singing, blowing soap bubbles and food rewards. Physical and visual prompting was also employed after the participant had performed two incorrect responses in a row. It is possible that implementing these types of reinforcers and/or prompts may have improved skill acquisition.

The data from the present experiment replicate and extend the findings of LeBlanc et al. (2003) and Lynch & Cuvo (1995) by showing that children with FXS can also learn geography and math relations using a stimulus equivalence procedure. In the study conducted by LeBlanc et al., similar stimuli were employed to teach geography relations to two children diagnosed with autism. In that study, both participants successfully learned the trained relations after receiving 360 and 550 training trials, respectively, and both participants demonstrated stimulus equivalence at post-test. In the geography condition in our study, three of the five children demonstrated stimulus equivalence at post-test, requiring between 89 and 523 trials to learn the trained relations. However, direct comparison between the two studies is problematic, because only two participants were included in the LeBlanc et al. study and the IQ levels of those participants were not assessed. Future studies should include larger numbers of participants and adequately characterize the intellectual ability of the participant sample (O’Donnell & Saunders 2003).

In summary, we were able to teach three of the five individuals with FXS to form basic geography relations and one of the five individuals to form basic math relations, using a time-limited stimulus equivalence procedure. In their review of the stimulus equivalence literature over the past 30 years, O’Donnell & Saunders (2003) identified 55 participants with IDs who had received stimulus equivalence training on a variety of skills. Of these, 34 participants completed the training and demonstrated stimulus equivalence at post-test. We agree with these authors that despite ‘a number of important demonstrations . . . in some ways the work has just begun’ (p. 146). We hope that future investigations will further examine the acquisition of stimulus equivalence in individuals with FXS, as well as in individuals with other genetic disorders. Ultimately, we believe that early interventions targeted at specific skill deficits at an early age could also be employed successfully with these populations.

Acknowledgements

The authors would like to thank the parents of the children involved in this study and Reviewer 1 who commented on an earlier draft of this paper. This research was supported by NIH grants MH50047 and MH64708, a Summer Student Fellowship awarded to the second author by the National Fragile X Foundation William and Enid Rosen Research Fund, and the Canell Family Fragile X Research Fund.

References


Stimulus equivalence in fragile X


Accepted 18 November 2005