Executive Functioning Training in Children with Fetal Alcohol Spectrum Disorder
Project Funded by ACCFCR

Research Team:
Principal Investigator: Dr. Jacqueline Pei
Department of Educational Psychology, University of Alberta

Co-Investigators:
Dr. Kimberly Kerns, Department of Psychology, University of Victoria
Dr. Carmen Rasmussen, Department of Pediatrics, University of Alberta
Dr. Christian Beaulieu, Department of Biomedical Engineering, University of Alberta

Project Coordinator:
Kennedy Denys, Department of Pediatrics, University of Alberta

Research Assistants:
Marnie Hutchison, Department of Educational Psychology, University of Alberta
Sarah Treit, Centre for Neuroscience, University of Alberta
Jennifer MacSween, Department of Psychology, University of Victoria

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**Summary:**

**Background:** Researchers from the University of Victoria and the University of Alberta partnered in a study aimed at improving executive functioning (EF) and attention capacity in children diagnosed with Fetal Alcohol Spectrum Disorders (FASD) through administration of a computerized process approach training program presented as a series of mini-games. Children diagnosed with FASD often have pervasive attention and EF deficits, including impairments in measures of cognitive flexibility, inhibition fluency, abstract thinking, deductive reasoning, hypothesis testing, problem solving, planning, and concept formation. This project is expected to contribute to our understanding of the potential for intervention in this area with children diagnosed with FASD, and may set the course for further intervention efforts of this type. Although it is widely recognized that process approach interventions can be effective for reducing cognitive deficits in aspects of attention and executive function in adults and children with brain injury, the research literature examining interventions with the FASD clinical population is very limited.

**Method:** A pilot study with 18 children ages 6-12 was completed in 2010, and an efficacy study with 21 children ages 6 – 15, was completed June 2011. Children were from the Edmonton, AB school district. The pilot study was a randomized control trial, while the efficacy study used a delayed- treatment control approach. Students received 24 half-hour sessions with the computer program *Cognitive Carnival*, over approximately 12 weeks. *Cognitive Carnival* combines elements of working memory, inhibitory control, and attention training. Participants additionally received one-on-one training of metacognitive strategies. Participants were tested pre- and post- intervention using cognitive measures of attention, executive functioning and academic abilities, as well as parent and teacher behaviour rating scales. Participants were also given diffusion tensor imaging (DTI) MRIs, allowing pre- and post-test maps of white matter tractography.

**Results:** Results indicate a high level of engagement by the children, marked gains on the cognitive training materials, and extremely positive observations with respect to changes in cognitive skills, ability to engage in cognitively demanding tasks over time, and self-regulation within the training sessions. Pilot study results revealed significant group differences, with the control group seeming higher functioning. Both groups made gains pre- to post- test on several measures, possibly due to the control group receiving one-on-one time and an ‘alternative intervention,’ however the intervention group still appeared to make more significant improvement. Preliminary results from the efficacy study revealed some significant gains from pre- to post- intervention, and continued improvement 12 weeks after. With both studies there were difficulties with differences between groups as well as a lot of variability on test results. Analysis of DTI results from the pilot (5 control and 3 intervention students) revealed a significant increase in fractional anisotropy between pre- and post-scans of the genu of the corpus callosum, an area in which diffusion abnormalities are associated with deficits in working memory. The increase observed after the intervention was much greater than would typically be expected with natural variability or typical brain development.
Executive Summary

Cognitive Carnival: 2010 Pilot Study and 2011 Efficacy Study

Background:
Researchers from the University of Victoria and the University of Alberta partnered in a study aimed at improving executive functioning (EF) in and attention capacity in children diagnosed with Fetal Alcohol Spectrum Disorders (FASD). It is widely recognized that process approach interventions can be effective for reducing cognitive deficits in aspects of attention and executive function in children and adults with brain injury, however there is little research examining these types of interventions with the FASD clinical population. A pilot project completed by Dr. Kerns using computerized training materials provided some evidence for positive results from this type of approach in a sample of children with FASD, but was limited by a small sample size and lack of a control group.

Purpose:
The primary purpose of this intervention was to improve EF and attention capacity in children diagnosed with FASD through the use of computerized process approach training and teaching of metacognitive strategies. An additional goal was to develop a way for this computerized training program to be accessed and used by children with FASD, either through schools or at home.

Methods:
Cognitive Carnival is a computerized process-approach training program developed by the University of Victoria; it combines elements of working memory, inhibitory control, and attention training. Cognitive Carnival was designed as a game, using a hierarchically
Based on the results from the 2010 pilot study, some changes were made to the design of the study, and in June 2011 the efficacy study was completed. The efficacy study used a delayed-treatment control, or ‘wait-list’ approach. Twenty-one children ages 6-15 were divided into two groups, both receiving the intervention. Using the new study design, group A received the 12-week intervention while group B served as the control group, and group B then received the intervention once group A had completed it. All students in this study were tested at pre- (0 weeks), mid- (12 weeks), and post-test (24 weeks), allowing group A to demonstrate whether or not there was sustained learning 12 weeks after they completed the intervention.

Measures:

For both the pilot and efficacy study, measures were used to assess working memory such as Spatial Span and Digit Recall. Tests of academics such as reading and math were used, as well as caregiver rating scales. The pilot study used the NEPSY to measure memory, visuomotor, visuospatial, attention, and inhibition changes, and other tasks such as the Day/Night Task and a computerized Go/No-Go task to measure inhibition. For the efficacy study, the NEPSY, Day/Night Task and Go/No-Go tasks were replaced by the TEC (Tasks of Executive Control), which measures attention, inhibition, and working memory, and the KITAP (Test of Attention-Performance-Children’s Version), which measures sustained and divided attention. The TEC was specifically designed for multiple administrations, minimizing practice effects.

In addition to the measures listed above, observational data was collected on the strategies used by group B of the efficacy study while they received the intervention. Interventionists were asked to note when they taught or prompted a metacognitive strategy, or when the student used the strategy spontaneously, without prompting or ‘mastered’ the strategy.

For the pilot study, 5 control and 3 intervention participants were also co-enrolled in a diffusion tensor magnetic resonance imaging (DTI) study undergoing scans before and after the intervention in order to determine if intervention-driven changes in neural architecture could be detected. DTI is a new, non-invasive MRI technique that looks at white matter and structural integrity of the brain.
Key Findings:

### Pilot Study

The most significant finding of the pilot study was that both the control and experimental groups improved pre- to post-test. There are several key factors that may have contributed to this effect:

- The control group may have been ‘higher functioning’ as can be seen in their better performance on many of the pre-tests. The groups did have similar IQ scores, making the differences in EF impossible to predict.

- While the control group did not receive an intervention specifically targeting EF, they may have received benefit from coaching and one-on-one time with the interventionist.

Thus we could consider the control group to actually be an ‘alternative intervention’ group that may have shown a positive effect resulting from the personal component of the intervention.

With that kept in mind, we still saw improvement in the intervention group and in many cases they appeared to be making larger gains than the control group. Unfortunately, many of the results we found were clinically significant but not statistically significant, likely due to small numbers in our groups (9 in each group).

The DTI results were quite positive, with a significant increase in tissue integrity in the genu of the corpus callosum. Abnormalities in this area are associated with deficits in working memory in FASD, and the improvements observed were much greater than typically expected with natural variability or brain development.

### Efficacy Study

The data from the efficacy study is still being analyzed, however we do have some interesting preliminary results from the TEC tests:

- Even though in this study design, both groups received the same intervention, only group A seems to have made improvement.

- Analysis revealed statistically significant reductions in errors and increased in the number of correct responses on the TEC for group A.

- Interestingly, not only did group A improve from pre- to post-intervention, they actually continued to improve after the intervention, with even better results 12 weeks post-intervention.

- We speculate that this post-intervention improvement may be attributed to acquisition and mastery of metacognitive strategies learned during the intervention.

- Results from the observational data looking at the metacognitive strategies revealed that participants were able to increase the number of strategies used over the course of the intervention, and many students were able to master several strategies and eventually use them on their own without prompting from the interventionist.

### Future Directions:

Based on the outcomes of these studies, a new cognitive-training program is being designed to be even more engaging for students. The next step in this study will be to train teachers' aides to deliver the intervention in the schools, and measure not only the learning and progress of the students, but also whether the TAs are benefiting from learning and teaching metacognitive strategies and utilizing scaffolding techniques.

This project is one of the first to develop an evidence-based computerized cognitive intervention aimed at improving underlying function and the developmental, educational, and behavioural outcomes that rely on such functions. Reducing the primary disabilities associated with FASD is anticipated to translate into fewer secondary disabilities. This study also serves as a model for future computerized cognitive intervention studies in FASD and other disabilities.
Executive Functioning Training in Children with Fetal Alcohol Spectrum Disorder

*Final Report*

**BACKGROUND**

Executive Functioning (EF) refers to higher-order cognitive processes under conscious control, necessary for thought and action in complex goal-directed behavior and adaptation to environmental changes and demands (Loring & Meador, 1999; Welsh, Pennington, & Grossier, 1991; Zelazo & Muller, 2002). EF includes flexible thinking, strategy employment, initiating and stopping actions, inhibition, fluency, and planning. Children and adolescents with FASD are impaired on many aspects of EF including: measures of cognitive flexibility, inhibition, fluency, abstract thinking, deductive reasoning, working memory, problem solving, planning, concept formation, and sustained attention (Mattson et al., 1999; Rasmussen & Bisanz, 2009; Schonfeld et al., 2001). The primary deficits in aspects of attention and working memory for individuals with FASD are pervasive, and impact memory and learning, school performance, and behavioral regulation. Despite these significant deficits there has been little research conducted around how to improve these underlying abilities in children with FASD.

Problems in attention, working memory and EF are seen in many childhood conditions, including brain injury (e.g. TBI, stroke, anoxia), developmental disorders (ADHD, learning disabilities, Autism Spectrum Disorders), and other conditions including low birth weight, prematurity, seizure disorder etc. The impacts of these difficulties often include problems with learning, life skills and adaptive functioning at school and at home, difficulty with coping skills, especially when demands, stress, and frustration increase. Self-esteem is often impacted negatively and learned helplessness increases and is reflected in decreased motivation and even avoidance of difficult cognitive tasks (Kit, Mateer, & Graves, 2007). In a school setting, problems in attention, working memory may result in inconsistent learning profiles and knowledge gaps. EF difficulties result in limited self-regulation of learning, impaired ability to plan and organize behavior to complete tasks, and inconsistent self-regulation of mood and emotion (Alloway et al., 2009). Repeated failures at school and home can result in frustration, anger, acting out, withdrawal, avoidance, and low self-esteem.
Researchers suggest that targeted, direct interventions can improve attention, and that this improvement is mediated by changes in underlying brain functions (Sohlberg & Mateer, 2001). One frequently studied method, attention process training, is theorized to repeatedly activate the neural networks underlying specific attentional processes. With massed, increasingly challenging practice, the repeated co-activation of neurons is believed to strengthen the neural pathways responsible for the process, leading to improved attention functioning. This type of intervention is empirically supported and currently well accepted as a treatment in the post-acute stage of TBI and stroke in adults (Rohling et al., 2009; Michel & Mateer, 2006). Likewise, recent work on massed practice of tasks which require increasingly higher levels working memory has also lead to improvement in performance on standardized measures of working memory and other cognitive abilities (Holmes, Gathercole, & Dunning, 2009; Westerberg et al., 2007) and is hypothesized as being secondary to inducing long-term plasticity through either improving the efficiency of neuronal responses or extending the cortical map serving working memory (Westerberg & Klingberg, 2007).

Research over the last decade has also shown attention process training to be a useful intervention method for children and adolescents. Randomized, placebo-controlled research has found that attention process training and/or working memory training improves targeted cognitive functions as well as being associated with improvement in other aspects of cognitive and behavioral functioning in children with TBI (Galbiati et al., 2009; van't Hooft et al., 2005), and children with ADHD (Kerns, Eso, & Thomson, 1999; Klingberg, Forssberg, & Westerberg, 2002; Klingberg et al., 2005; Shalev, Tsal, & Mevorach, 2006). Work by Rueda and colleagues (2005) suggests that attention training in typically developing preschoolers resulted in more ‘adult like’ patterns of electrophysiological functioning on tasks requiring higher levels of attention.

Indeed, Kleim and Jones (2008) suggest that the brain continuously remodels its neural circuitry, and that this is also the mechanism by which the damaged brain re-learns lost behavior or supports new behavior in response to rehabilitation. They define several principles considered critical for experience dependent neural plasticity, including sufficient repetition of a behavior, sufficient time for changes to occur, sufficient intensity of the behavior occurring,
and that behaviors that are more salient by being engaging or rewarding are more likely to produce change.

Process specific cognitive training approaches (Sohlberg & Mateer, 2001) are designed to improve the underlying impaired processes directly through this type of “practice.” Research supports that certain types of cognitive training can actually change underlying brain functioning (Klingberg, Forssberg, & Westerberg, 2002; Rueda et al., 2005). Interventions are designed to specifically improve an underlying impaired process directly, and any change in underlying capacity is assumed to be due to neural plasticity and/or reorganization of the neural system (McNab et al., 2009). While these techniques may not restore or remediate cognitive function to full capacity, they may improve function enough to allow children to engage more in learning or other compensatory strategies. Participants may experience increased sense of self-esteem, which can lead to increased benefit for other interventions and therapies.

Although it is widely recognized that process approach interventions can be effective for reducing cognitive deficits in aspects of attention and EF in adults and children with brain injury and ADHD, the research literature examining interventions with the FASD clinical population is very limited. Indeed, the basis for making decisions regarding secondary and tertiary interventions for individuals with FASD is typically anecdotal reports and clinical wisdom. A pilot project completed by a co-investigator on this project (Kerns, et al., 2010) using a computerized attention training intervention with children with FASD reported increased measures of attention, working memory, processing speed, and academic fluency measures. This study, however, was limited by small sample size and lack of control group. Vernescu (2008) also reported improvements in a small group of children with FASD using attention process training materials.

Direct processes approaches to improving performance on tasks requiring attention and aspects of EF are hypothesized to strengthen underlying neural connections in the networks that support these abilities. Diffusion tensor magnetic resonance imaging (DTI) is a new non-invasive MRI technique that permits the virtual dissection of these networks through investigation of the white matter (i.e. the wiring) in the brain as well as a measure of its
structural integrity (e.g. degree of myelination). DTI is not presently performed in routine clinical MRI scans of the brain although it is one of the most active research topics in MRI. DTI has detected subtle brain abnormalities in others disorders such as multiple sclerosis, Alzheimers disease, epilepsy, and dyslexia, in regions that otherwise appear normal on conventional MRI. A recent study (using DTI) conducted by Dr. Beaulieu and Dr. Rasmussen found that children and adolescents with FASD had significant abnormalities (as compared to normal controls) in a variety of different white matter tracts as well as deep gray matter structures (Lebel et al., 2008). DTI is a neuroimaging technique that has the potential to be sensitive to enhanced neuronal connections and strengthening of neural pathways. As such, it could provide a means of imaging the changes associated with process specific interventions.

This project emphasized a well controlled pilot investigation of early cognitive development and the direct impact of cognitive process training approaches for children with FASD, and sought to develop one of the first evidence-based cognitive interventions aimed at improving underlying function and the developmental, educational, and behavioural outcomes that rely on such abilities. Reducing these primary disabilities is anticipated to translate into fewer secondary disabilities for children with FASD.

METHOD

This study underwent two phases. The first was a pilot phase and the second was an efficacy study that allowed us to truly examine the potential value of the intervention after addressing any concerns identified in the pilot phase.

Phase One: Cognitive Carnival Pilot

Cognitive Carnival is a computerized process-approach training program combining elements of working memory, inhibitory control, and attention training. Cognitive Carnival was designed as a game, using a hierarchically based intervention approach. The Cognitive Carnival consists of several mini-games requiring either higher levels of sustained attention or working memory to succeed, and provides ongoing reward through the gameplay. Cognitive Carnival provides exercises requiring aspects of both visual and auditory working memory.
Cognitive Carnival includes three different mini-games and contains internal and external rewards that are provided throughout game play. Each level requires 90% or higher accuracy to advance, and errors are marked by tones. Levels are hierarchically organized by difficulty and can be re-tried as many times as necessary.

In addition to this exposure to the computerized training program, the participants were paired with an interventionist who taught and helped participants practice metacognitive strategies to improve performance in the game. Metacognition involves monitoring and controlling one’s own cognition, and is necessary for self-regulation (Flavell, 1987). Some of the metacognitive skills taught included deliberate use of goals and strategies to control cognition, procedural knowledge, skills comprised of orientation and planning strategies as well as strategies for monitoring the execution of the planned action and evaluating the outcome (Veenman & Elshout, 1999). The interventionists also used scaffolding to facilitate learning. According to Azevedo and Hadwin (2005), scaffolds are: “tools, strategies, and guides used during learning to enable the development of understanding that is beyond the individual’s immediate grasp.” The scaffolding used in this study had three specific components: 1) it was individualized (tailored to the child and the context); 2) it was calibrated (dynamic and provided at a specific level when needed) and 3) it faded (provided as necessary and reduced over time as competence increased).

Tasks requiring attention and EF are hypothesized to strengthen underlying neural connections in the networks that support these abilities. Diffusion tensor imaging (DTI) is an advanced MRI technique that permits the virtual dissection of these networks through investigation of the white matter (i.e. the wiring) in the brain as well as a measure of its structural integrity (e.g. degree of myelination). DTI has been used to reveal subtle yet widespread white matter abnormalities in FASD (for review, see Wozniak & Muetzel, 2011) and has the potential to be sensitive to subtle changes in brain microstructure associated with process-specific interventions.

Participants were recruited through the Edmonton Public School Board, and from March to June, 2010 we completed the pilot study using Cognitive Carnival. Eighteen children diagnosed with FASD participated in this randomized control trial (see demographics table
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Each child received 24 half-hour sessions over a 12-week span, participating in approximately 2 sessions per week. The children being split into two groups, one receiving the intervention condition (Cognitive Carnival) and the other receiving a control condition (playing computer games focusing on educational materials but not EF).

**Demographics:**

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Control</th>
<th>Experimental</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number (n)</td>
<td>9</td>
<td>9</td>
</tr>
<tr>
<td>Mean Age (range)</td>
<td>9.78 (6-12)</td>
<td>9.12 (6-12)</td>
</tr>
<tr>
<td>Gender (n)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>7</td>
<td>6</td>
</tr>
<tr>
<td>Male</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>IQ</td>
<td></td>
<td></td>
</tr>
<tr>
<td>General</td>
<td>85.22</td>
<td>85.00</td>
</tr>
<tr>
<td>Visual</td>
<td>94.78</td>
<td>92.33</td>
</tr>
<tr>
<td>Verbal</td>
<td>80.22</td>
<td>82.33</td>
</tr>
</tbody>
</table>

During the intervention the students played the computer games with the aid of an interventionist who helped the students apply metacognitive and self-regulation skills, while the interventionists themselves executed the scaffolding techniques, tailoring their supports to each individual student’s needs and abilities then gradually reducing them as the students’ competence increased (fading). The students were explicitly encouraged to try to complete the tasks, even if they made mistakes, and learn from these errors. It was emphasized that students were in a safe environment free from judgment, ridicule, or even a grading system. When students had difficulty performing on certain tasks, the student and the interventionist would brainstorm different strategies for success, or the interventionist would suggest strategies when suitable, and the interventionist would help facilitate the implementation of these strategies. An example of strategies used:

- **Rehearsal:** “Try repeating the numbers as you hear them.”
- **Visualization:** “Picture the shapes in your mind.”
- **Reducing speed:** “Take some time to memorize the items before starting to respond.”
As the students became more effective at implementing successful strategies, the interventionists reduced their supports. The interventionists carefully monitored success, improvement, and especially the use of self-regulation, and gave verbal rewards. Students also received a prize after each session, regardless of their progress, with the emphasis being on trying and working hard.

**MEASURES**

Participants were pre- and post-tested using rating scales and cognitive measures of attention, executive functioning, and academic abilities. The following tests were used during the pilot study:

<table>
<thead>
<tr>
<th>Test</th>
<th>Area Targeted:</th>
</tr>
</thead>
<tbody>
<tr>
<td>NEPSY and NEPSY-II</td>
<td>neuropsych: memory, visuomotor, visuospatial, attention &amp; inhibition</td>
</tr>
<tr>
<td>Spatial Span (WISC IV Integrated)</td>
<td>working memory</td>
</tr>
<tr>
<td>Digit Recall (WMTB-C)</td>
<td>working memory</td>
</tr>
<tr>
<td>WJ-III Reading Fluency</td>
<td>reading</td>
</tr>
<tr>
<td>WJ-III Quantitative Concepts</td>
<td>math</td>
</tr>
<tr>
<td>Animal CPT</td>
<td>attention</td>
</tr>
<tr>
<td>Day/Night Task</td>
<td>inhibition</td>
</tr>
<tr>
<td>Computerized Go/No-Go</td>
<td>inhibition</td>
</tr>
<tr>
<td>BRIEF Parent and Teacher</td>
<td>behaviour</td>
</tr>
<tr>
<td>Conners Rating Scales</td>
<td>behaviour</td>
</tr>
</tbody>
</table>

The NEPSY (Developmental Neuropsychological Assessment) is widely used by school psychologists, neuropsychologists, and research psychologists to assess children ages 3-12 and provides comprehensive assessment over five functional domains: Attention/Executive Functions, Language, Sensorimotor Functions, Visuospatial Processing, and Memory and Learning. We used subtests from the Attention/Executive Functioning domain. The Working Memory Test Battery for Children (WMTB-C) assesses working memory in 5 to 15 year olds. We administered one test of phonological working memory (Digit Recall) and one test of visualspatial working memory (Block Recall). The WJ-III (Woodcock-Johnson III) provides a comprehensive system for measuring general intellectual ability and includes specific tasks.
for academic fluency.

The BRIEF (Behaviour Rating Inventory of Executive Function) consists of two rating forms - a parent questionnaire and a teacher questionnaire, designed to assess executive functioning in the home and school environments. The CRS-R (Conners Rating Scales – Revised) are paper and pencil screening questionnaires completed by parents and teachers to assist in evaluating children for symptoms of attention deficit/hyperactivity disorder (ADHD). Parent and teacher versions of this standardized rating scale were used.

The Day/Night Task is a measure of inhibition (EF). It is composed of one page that contains rows of pictures of 8 suns and 8 moons arranged pseudorandomly. The children are asked to go across the rows and say “sun” for every picture of a moon and to say “moon” for every picture of a sun as quickly as they can. Lastly, the 'Farm Animals Game' CPT Task is a computerized continuous performance task, measuring attention, in which participants are presented with pictures of farm animals and the sounds they make, and asked to respond (by hitting the mouse key) every time a specified target animal is shown, or they hear the sound that animal makes. Outcome variables include number of correct hits for targets, omissions of targets, commissions to distracter animals/animal sounds, and reaction time for correct hits.

Pre- and post-testing was completed by research assistants blind to group assignment. The majority of the participants were co-enrolled in a longitudinal DTI study, and so they also received pre- and post DTI scans. For analysis we looked at differences in raw scores between pre- and post-intervention.

As an adjunct to this study, eight participants underwent DTI scans before and after the intervention in order to determine if intervention-driven changes in neural architecture could be detected. This sub-study was funded by the Canadian Institutes of Health Research (CIHR; PI: Dr. Beaulieu).
RESULTS

The following table lists the subtests that improved from pre- to post-test:

<table>
<thead>
<tr>
<th>Experimental Group</th>
<th>Control Group</th>
</tr>
</thead>
<tbody>
<tr>
<td>NEPSY-II Memory for Names</td>
<td>NEPSY-II Memory for Names</td>
</tr>
<tr>
<td>NEPSY-II Memory for Names Delayed</td>
<td>NEPSY-II Memory for Names Delayed</td>
</tr>
<tr>
<td>WJ-III Quantitative Concepts*</td>
<td>WJ-III Quantitative Concepts</td>
</tr>
<tr>
<td>NEPSY Arrows</td>
<td>NEPSY Visuomotor Precision: completion time</td>
</tr>
<tr>
<td>NEPSY Auditory Attention: total errors*</td>
<td>NEPSY Auditory Attention: total correct</td>
</tr>
<tr>
<td>WMRT-R Word ID</td>
<td>NEPSY Auditory Attention: omission error</td>
</tr>
</tbody>
</table>

* approached significance

The table shows that there were several tests that improved for both groups, not just the experimental group that received the intervention.

Figure 1:

**WJ-III Quantitative Concepts**

* approached significance

In Figure 1 you can see that while both groups improved their performance from pre- to post-test, the experimental group started much lower than the control group and actually improved more. Overall, the experimental group improved by 3.63 points and the control group only by 2.34 points.
In Figure 2 we see that both groups improved their performance from pre- to post-test, and again the experimental group made more improvement on both measures.

While the control group did not make a statistically significant improvement from pre- to post-test, this graph demonstrates how once again, the performance of the experimental group was much lower at pre-test and improved greatly to almost match that of the control group at post-test.
**Figure 4:**

**Nepsy-II: Visuomotor Precision**

![Graph showing completion time and total errors for control (Ctrl) and experimental (Exp) groups pre and post-test.](image)

* control completion time was only found to be statistically significant at p<0.05

Figure 4 contains two very interesting graphs representing the average amount of time it took the students in the control and experimental groups to complete the test as well as the number of errors made. When you look at completion time, you can see that the two groups were equal at pre-test, and that only the control group got faster at post-testing. However, when looking at the number of errors in each group, the control group actually made more errors at post-test, while the experimental group, who had much more errors at pre-test, made a significant reduction in their number of errors at post-test. This suggests that the control group got faster and more careless, while the experimental group did not improve their time but really improved their precision.

**DTI:** Repeated measures analysis of covariance (ANCOVA) was used to determine change in DTI parameters between scans by group (intervention vs sham), using time between scans as a covariate. This revealed a significant increase in fractional anisotropy (a parameter associated with tissue integrity) between scans in the genu of the corpus callosum; a white matter tract that connects the frontal lobes of the right and left hemispheres. Diffusion
abnormalities in the genu have been previously identified (Lebel, et al., 2008; Ma, et al., 2005) and associated with deficits in working memory in FASD (Wozniak, et al., 2009). The increase observed after the intervention was much greater than would typically be expected with natural variability or with brain development in the short time period between scans. This is an interesting preliminary finding that may provide the basis for future studies of intervention-based neural plasticity in FASD.

**DISCUSSION**

We encountered several issues and limitations during the pilot study. One concern was the low numbers and thus lack of statistical power for observing differences, having only 9 students in each group. Additionally, some of the pre- and post-measures seemed not sensitive enough to detect changes, and finally there was also a time-lapse between the intervention and post-testing due to the post-test being conducted along with the post-DTI, which was prone to scheduling backups. In addition, there was significant variability within the participants in the groups, and large differences between the groups (as can be seen the figures above), with the control group seeming to be higher functioning, with better performance on most of the pre-tests. Finally, it is important to note that the control group in this pilot study still received
a computer intervention, and that the one-to-one control intervention may truly be considered an ‘alternative intervention’. While *Cognitive Carnival* is designed to target EF, the personal component of working with a one-on-one interventionist providing coaching and scaffolding, is essential. The control group in this pilot study also received the personal component of the intervention, and improvements in the control group could be due to the positive effect that one-on-one time and coaching may have on students with FASD.

**Phase Two: Cognitive Carnival Efficacy Study**

**MEASURES**

Keeping the successes and issues of the pilot study in mind, the 2011 intervention was designed as a delayed-treatment control, or ‘wait-list’ approach and included 21 children of a larger age-range receiving the 12-week intervention (see demographics table below).

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Group A</th>
<th>Group B</th>
<th>Combined</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number (n)</td>
<td>10</td>
<td>11</td>
<td>21</td>
</tr>
<tr>
<td>Mean Age (range)</td>
<td>11.63 (6-15)</td>
<td>12.17 (7-15)</td>
<td>11.91 (6-15)</td>
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<tr>
<td>Gender (n)</td>
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<tr>
<td>Female</td>
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<td></td>
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<tr>
<td>General</td>
<td>86.2</td>
<td>77.18</td>
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</tr>
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<td>Visual</td>
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<td>84.82</td>
<td>88.90</td>
</tr>
<tr>
<td>Verbal</td>
<td>83.0</td>
<td>76.18</td>
<td>79.43</td>
</tr>
</tbody>
</table>

We included children from the control group of the pilot study, to allow them the benefits of learning *Cognitive Carnival*. The new group of 21 students was randomly assigned to one of two groups (Group A, n=10; Group B, n=11), and all students were tested at three time-points: 0 weeks, 12 weeks, and 24 weeks (see figure below). Group A received the 12-week, 24 session intervention from week 0 to week 12, and group B (wait-list group) received no intervention or interaction during this time. After the 12 week mid-test point, group B received the intervention from week 12 to week 24 and group A received nothing.
Figure: Delayed-treatment control method

<table>
<thead>
<tr>
<th>Group A: Intervention</th>
<th>Group A: nothing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group B: nothing</td>
<td>Group B Intervention</td>
</tr>
</tbody>
</table>

0 weeks | 12 weeks | 24 weeks

Using this wait-list method, group B was the control for both group A and themselves, after 12 weeks with no intervention. Group A measured retention (12 weeks after the intervention). This approach has many benefits over the previous approach of the pilot study because it had larger power with twice as many participants receiving the intervention. We still had a control group without having to use a “sham” intervention, and we were able to eliminate group difference because all 24 children were included in the larger group. We were also able to gather some follow-up data from Group A to determine if changes were sustained.

Statistically this mixed design allowed us to control for change due to development, test-retest, and familiarity with the procedures (Cicerone, Azulay, & Trott, 2009). This design served several important purposes: First, sham intervention controls like the ones used in the pilot study, allow for comparison of interventions but are not inert but truly a ‘different’ intervention with few understood outcome characteristics (Giles, 2010). Given the preliminary nature of this research, a better measure of any treatment effects was necessary. The wait-list control design provided the “best match” control for a population of children who frequently display very diverse cognitive profiles. Second, the design provided the intervention to all participants in a timely manner, which was important for ethical and clinical purposes. Finally, the design preserves sample size in a limited population, providing a more statistically powerful study. As in the pilot study, participants in the efficacy study were also given the opportunity to participate in the ongoing DTI study.
MEASURES

For the 2011 efficacy intervention we retained some of the measures used in the pilot study and also added three new measures. The Tasks of Executive Control (TEC) and Test of Attention Performance – Children’s Version (KITAP) are both standardized computer-administered tests. The Attention Deficit Disorders Evaluation Scale (ADDES) is a standardized rating scale used to detect hyperactive and inattentive behaviors; we used the school and home versions. All of the new measures have excellent psychometric properties, minimal test-retest effects, and have been used successfully in previous intervention studies.

<table>
<thead>
<tr>
<th>Test</th>
<th>Area Targeted:</th>
</tr>
</thead>
<tbody>
<tr>
<td>Spatial Span (WISC IV Integrated)</td>
<td>working memory</td>
</tr>
<tr>
<td>Digit Recall (WMTB-C)</td>
<td>working memory</td>
</tr>
<tr>
<td>WJ-III Reading Fluency</td>
<td>reading</td>
</tr>
<tr>
<td>WJ-III Quantitative Concepts</td>
<td>math</td>
</tr>
<tr>
<td>KITAP</td>
<td>attention (sustained and divided)</td>
</tr>
<tr>
<td>TEC</td>
<td>attention/inhibition/working memory</td>
</tr>
<tr>
<td>ADDES</td>
<td>attention/impulsivity/hyperactivity</td>
</tr>
</tbody>
</table>

Observational data was also collected on the metacognitive strategies used by group B during their intervention period (between weeks 12 and 24), with the interventionist completing a protocol after each Cognitive Carnival session with a student. Modeled after Flavell’s (1970) stages of strategy mastery, the protocol listed several metacognitive strategies, and asked the interventionist to note which strategies had been taught, used with prompting, used spontaneously by the student, or mastered by the student during each session. The frequency of strategy use was not recorded. Instead, the protocol served as a way to track what stage of learning participants were at for each strategy during each session. Taught and prompted strategies were grouped and entered as ‘prompted’ and spontaneous and mastered strategies were grouped and entered as ‘spontaneous’. 720 minutes of intervention were collapsed into 4 approximately equal times periods of 180 minutes each and the highest level of learning for each strategy was recorded for each time period.
RESULTS

Cognitive outcomes

All of the students in group A completed all 24 sessions in the 12-week intervention, while 3 of the students in group B were unable to complete their 24 sessions due to behavioural issues. For statistical analyses, comparing all testing phases, only children who completed the intervention are being included in the analyses. Data from the cognitive tasks are still undergoing analyses at present, but some of the TEC findings are presented here.

The TEC is a standardized computer-administered measure that involves the student responding (or inhibiting a response) by pressing a key to various items that show up on the screen and disappear quickly. The TEC involves 6 different conditions using the N-back paradigm: 0-back, 1-back and 2-back, and each is also presented as a go/no-go task, creating 6 conditions in total. Significant variation was found across the different conditions of the TEC, so the data was collapsed across all 6 conditions for each of the primary outcome measures. The major outcome variables from the TEC include the number of targets and standards correct, total omission errors (failing to respond when a response is required), and commission errors (responding when no response is required).

Given the wait-list control design of this study, it was hypothesized that repeated measures analysis of variance (RMANOVA) would yield a significant interaction between group membership and testing period, as we anticipated that group A would make gains between pre/mid testing and that group B would make gains between mid/post testing. The TEC was designed for multiple administrations, so we anticipated few practice effects on this measure.

The figures below show the results for each of the three time periods for both groups A and B, including the percentage total correct, the total number of incorrect responses, the total number of commission errors, and total number of repeated responses.
Analyses revealed significant interaction effects on the number of incorrect responses (p<.05) and marginally significant (p<=.10) on the total percentage correct and total number of commission errors (note: commission errors only possible on the 3 tasks with inhibition or go/no-go component). Given the small sample sizes reducing the power of the overall analyses, for significant and marginally significant findings, follow-up RMANOVA by group were calculated. For total correct responses to targets and standards (see figure), analyses revealed a significant effect for group A (F=5.78, p=0.011, ES=0.39). In contrast, there were no significant changes for group B, either between pre- and mid-testing (as hypothesized) or between mid- and post-testing where we would expect to see improvement.

For total number of commission errors (see figure), analyses again revealed a significant effect for group A (F= 17.19, p=.006, ES=.66), and again no significant changes across testing sessions were found for group B. No statistically significant changes across the testing sessions were supported for either group A or B on commission errors or total number of multiple responses.

**Metacognitive Strategies**

Results revealed that children and adolescents used any number of 25 different strategies to aid their EF performance. The strategies used were aimed at holding more information in their memory, improving attention abilities, reducing impulsive behaviours, managing negative emotions, goal setting, and focusing on their strengths. Participants were able to increase the number of strategies they used spontaneously and decrease the number of strategies used through prompting over the course of the intervention. By the end of 180 minutes of gameplay, participants could use an average of 9 different strategies spontaneously and by the end of 720 minutes of gameplay, participants could use an average of 12 different strategies spontaneously. By the end of 180 minutes of gameplay an average of 4 strategies were prompted and by the end of 720 minutes of gameplay, an average of 2 strategies were prompted. Furthermore, many of the strategies that were prompted at 180 minutes were used spontaneously at the end of the intervention. Overall, older participants used more spontaneous strategies and significantly fewer prompted strategies than younger children.
DISCUSSION

Cognitive Outcomes

One of the benefits of the wait-list design of this study was to allow us to have larger numbers of students receiving the intervention, however until we have a sense of what is happening with group B, we’ve focused on group A, which shows a nice improvement over time on an attention, inhibition, and working memory test (the TEC). Group B, however, does not show improvement over time, and in some cases seems to be showing worse results after the intervention. While the figures above may show the means scores for group B getting worse, none of these results were statistically significant due to the large amount of variability within the group. Further analyses are being done and inquiries with the interventionists are being done to get a better idea of the participants in group B and how they may be different from those in group A. The improvements made by group A are what we hypothesized we would see, and in some cases is even better, with correct scores improving and errors decreasing from pre- to mid-testing (after intervention), and then even more improvement being made from mid- to post-testing (12 weeks after intervention). We speculate that this improvement from mid- to post-test where we would expect to see retention or maybe even a slight decline may be because the students were able to master some of the tasks and strategies they learned, and then generalize that learning and apply it to other tasks.

Given that the other cognitive tasks used in this study were not explicitly designed for repeated administrations, and that they were administered three times over a period of 24 weeks, analyses for these data are currently underway utilizing a Reliable Change Index (RCI) definition of improvement as there was evidence in most measures of both groups making some gain on repeated administration.

Metacognitive Strategies

These results suggest that children and adolescents with FASD can use a wide and varied battery of executive function strategies that if transferred outside of the intervention setting, could have a potentially positive effect on their daily functioning. This is important because although a child with FASD has incurred brain damage, efforts to equip them with
skills that can improve their independent functioning should not be ignored. This data cannot indicate that these strategies resulted in improvements in memory or executive function performance; however, practicing a variety of strategies that may support executive functioning is considered helpful in the least.

**FUTURE DIRECTIONS**

After seeing the variability of group B in the efficacy study, it is important to consider how we can document these findings. The test re-test performance of children with FASD is much more variable than what is found in typically developing children, and this is something that is discussed by researchers and clinicians working in FASD, but difficult to find in documentation. The research team will analyze the results further to determine whether there is enough evidence of variability to write a publication or if it should be included in an overall publication about the results of the pilot and efficacy studies.

Based on the outcomes of *Cognitive Carnival*, the research team has been developing a new cognitive-training game that was even more engaging based on the feedback from the students in the study as well as the interventionists. The new cognitive training materials are also designed to implement the ability to individually tailor the task difficulty based on each child’s individual performance. Extensive communication during and after the intervention between the treatment team and the computer science team members was essential for providing the needed information to write the design document for the updated web-based cognitive training game, now called the *Caribbean Quest*.

One of the major redesign requirements of *Caribbean Quest* was to provide delivery via multiple operating systems and machines. The technical issues, hardware compatibility, and limited instructions for a nontechnical end user made the *Cognitive Carnival* untenable for broader distribution. Therefore, to target multiple environments, the program was re-created as a web-delivered product. The implementation of low graphic requirements for the *Caribbean Quest* games (2D sprites) meant there was no need for a traditional gaming application with full access to 3D hardware. The installation was designed as a one-click process, allowing a modestly educated user to use the program.
A challenge that emerged in the implementation of *Cognitive Carnival* was finding an appropriate level of difficulty for a large target with high variability in cognitive capacities. To address this in *Caribbean Quest*, a dynamic difficulty system was created to allow for adjusting the working memory span tasks according to the user’s performance, while maintaining distinct characteristics that do not change for each level (e.g., auditory tasks versus visual, etc.). The input system for *Caribbean Quest* was also designed to be flexible and for many types of control methods, again for use in a variety of settings with potentially different hardware devices. As such, the program currently allows players to use a gamepad, keyboard, mouse, or potentially numerous other peripherals.

In addition to extensive changes to the basic software design, additional cognitive exercises have been added to the existing series of ‘games’ based on recent review of the current literature and findings in cognitive remediation. Considerable time was also spend on integrating a ‘reward’ system into the *Caribbean Quest*, with both the ability for short-term rewards (gaining points to be able to play a fun non-cognitively demanding game) and long term rewards (inclusion of a ‘meta-game’ which as participants finish activities provides a higher level of reward for completion of exercises). Unlike the prototype game, *Caribbean Quest* was designed as a more integrated thematic ‘game’ versus just a series of cognitive exercises in a game like environment. With enhanced and original graphic art and music, the game presents much closer to a typical ‘video game’ than educational activity. These types of changes were made to enhance the motivation and interest of the game to children.

Research management suggestions were also taken into consideration while developing *Caribbean Quest*, with significant attention to creating a product that would be useful for a variety of ages of children and while of specific use for children with deficits in aspects of attention and executive function, not specific to a disorder. While the current funding does not allow for investigation of this intervention in populations other than the target population, investigators within this collaborative project were successful in securing some funding to extend the target populations. Research management suggestions for additional cognitive measures for pre/post intervention assessments were also added to the battery of tests to be used to assess *Caribbean Quest*. 
Initially, the plan was to make the intervention available to caregivers to use at home with their FASD diagnosed children, however it was determined that given the intensity of the intervention, delivery within the school setting rather than at home might be most efficacious. Caregivers of children with FASD are often seeking supports and services (Brown, Sigvaldason, & Bednar, 2005) however delivering the intervention at home would require training as well as a weekly time commitment and we did not want to add to the burden that these caregivers of children with disabilities already experience. As most children identified with FASD are already receiving support services with teachers’ aides (TAs), we felt that these aides may be the best providers of the intervention. Likewise, most schools typically have a number of children with FASD or other disorders that affect attention, working memory and executive functions. Thus, training TAs to provide this intervention is an efficient means of providing intervention opportunities for a number of children. In addition, as TAs work with more children, their skills in providing the intervention will also increase, providing an additional valuable training method to their current repertoire of skills.

We intend for phase 3 to first involve a training session for TAs in the schools who are able to do the intervention. Through the training we will inform them as to the purposes of the intervention, the ways in which it meets the unique needs of the FASD population, and the metacognitive strategies that they might employ to support the children in using this training tool.

After their initial training, the aides will engage in the direct administration of the intervention tool with gradually decreasing supports available to them. This means that for their first administration, an experienced interventionist will model strategy use and intervention implementation. For the second session, the interventionist will again attend, but in a purely supportive/observational capacity. The interventionist will be available to attend a third session, if desired by the aides. After this, TAs will independently administer the intervention but will communicate weekly with the trained interventionist to discuss strategies, challenges, and new ideas. This communication may take place at a group level if there is more than one EA working in the intervention at a school site.

Testing of the students, overall research design, and specific strategy approaches
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will not change from the efficacy study at this point in time. Outcomes for the study will again include individual outcomes for each child in the intervention. In addition, a set of outcomes will explore whether there is evidence of increased capacity on the part of the TAs in their ability to respond to the needs of the children within the intervention setting, as well as whether that increased capacity generalizes to their ability to meet the children’s’ needs in the classroom and whether they feel an increased capacity to meet all children’s’ needs. Web-based resources will be explored as a means to facilitate communication between TAs delivering the intervention, and the interventionists. This then has the potential to become a source of ongoing support.

We want to work towards increasing the capacity of the schools to administer the intervention independently so that it becomes more available to students. It is a hope that eventually this tool may become a resources to schools beyond the scope of this research project alone, and that with the TAs could eventually become the mentoring interventionists.

This study serves as a model for future computerized cognitive intervention studies in FASD and other disabilities. Knowledge translation is essential, as it is important for others to learn from our mistakes and our successes in addition to gaining knowledge about our successes. Not only does this study contribute to knowledge around the effectiveness of computerized EF interventions for children with FASD, but it speaks to the experiences of working within schools and partnering with principals, teachers, and TAs to gain positive research outcomes.
REFERENCES


Executive Functioning Training in Children with Fetal Alcohol Spectrum Disorder – Final Report


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Thank You

Please direct correspondence to:

Dr. Jacqueline Pei
Department of Educational Psychology
6-131 Education North, University of Alberta
Edmonton, AB, T6G 2G5
jpei@ualberta.ca

Dr. Kimberly Kerns
Department of Psychology
P.O. Box 3050, University of Victoria
Victoria, BC, V8W 2P5
kkerns@uvic.ca