University of Alberta

Mathematics Intervention for Children with Prenatal Alcohol Exposure and Fetal Alcohol Spectrum Disorders

by

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Dedication

For every child, individual, and family who has been impacted by prenatal alcohol exposure.

"The most turbulent, the most restless child has, amidst all his faults, something true, ingenious, and natural, which is of infinite value and merits every respect."
Felix-Antoine-Philibert Dupanloup

Abstract

Individuals with fetal alcohol spectrum disorders (FASD) experience deficits in behavior, cognition, and academic functioning resulting from prenatal alcohol exposure (PAE). Although receiving intervention for developmental disabilities is a strong protective factor against negative outcomes in FASD, intervention research in this population is in its infancy. The purpose of this study was to replicate and extend a mathematics intervention developed in the USA specifically for children with FASD. Seventeen Canadian children aged 4-10 with confirmed PAE or an FASD diagnosis were assigned to either the math intervention or a contrast intervention. At pre-test, both groups generally exhibited below-average math, executive function, working memory, and visuospatial skills. Following a relatively brief, individualized, one-on-one intervention, children in the math group demonstrated significantly greater changes in math achievement than the contrast group. Significant changes in other cognitive functions were not observed. Older age, lower IQ, and lower socioeconomic status were associated with increased math treatment change in the math group. The replication and extension of the math intervention appears to have significant, positive impact on mathematics achievement scores of children with PAE and FASD.

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CHAPTER ONE

Introduction

Each year, approximately 1 in 8 women consume alcohol while pregnant, and approximately a sixth of these women binge drink (Mattson et al., 2013). Consequently, thousands of children are born prenatally exposed to alcohol, the most widely consumed teratogen in the world (Streissguth, 1997). Approximately 1% to 2 to 5% of children have fetal alcohol spectrum disorder (FASD), an umbrella term that describes a broad spectrum of congenital neurological and physical effects resulting from alcohol-induced organic brain damage (Chudley et al., 2005; May et al., 2009).

Individuals with FASD tend to exhibit significant neurobehavioral deficits, including a range of mild to severe dysfunction in executive functioning, intellectual ability, learning, achievement, memory, attention, visuospatial functioning, language, and adaptive functioning (for a review, see Mattson, Crocker, & Nguyen, 2011). These primary neurobehavioral disabilities, coupled with limited availability of appropriate and effective interventions, likely give rise to the multitude of adverse outcomes common in individuals with FASD. These outcomes include disrupted educational and occupational experiences, confinement (e.g., incarceration, inpatient treatment), inappropriate sexual behaviors, and alcohol and drug abuse (Streissguth, Barr, Kogan, & Bookstein, 1996).

As a result of the expense of multifaceted service delivery efforts to address primary and secondary disabilities coupled with lost individual and caregiver productivity, it has been estimated that the total annual cost associated with FASD in Canada is $5.3 billion (Stade et al., 2009). Specifically, in Alberta, the lifetime cost of supporting an individual with FASD is estimated at $1.1 million (Thanh & Jonsson, 2009). However, early diagnosis and access to services for developmental disabilities have been found to reduce negative outcomes and thus decrease financial burden and other social-emotional tolls that disability may render on individuals and families (Streissguth et al., 2004).

Developing appropriate services to meet the needs of individuals with FASD has been difficult for several reasons. First, the body of research addressing FASD is relatively young—approximately 40 years (Jones & Smith, 1973). Only more recently has understanding of cognitive, behavioral, and adaptive consequences of FASD advanced enough to begin to inform appropriate habilitation methods. Second, individuals with FASD experience a myriad of behavioral and self-regulation problems in addition to cognitive impairments. Many also experience the systemic effects of chaotic family life and involvement in social services (Streissguth et al., 1996). This unique presentation makes it difficult to fit children with FASD neatly into existing treatment or educational intervention platforms. Fortunately, there have been several recent attempts to develop and pilot interventions targeted specifically to children with FASD, such as interventions for improving memory (Loomes, Rasmussen, Pei, Manji, & Andrew, 2007), behavior and school functioning (Stromland et al., 2005), attention (Kerns, MacSween, Wekken, & Gruppuso, 2010), social skills (O'Connor et al., 2006), safety (Coles, Strickland, Padgett, & Bellmoff, 2007), and mathematics (Coles, Kable, & Taddeo, 2009; Kable, Coles, & Taddeo, 2007).

Mathematics, in particular, has been identified as a specific area of difficulty for children with FASD. It has been suggested that deficits in executive functions, working memory, and visuospatial functioning may be related to observed deficits in mathematics in children with FASD (Kable et al., 2007). Therefore, the purpose of the present study was to investigate the effectiveness of a mathematics intervention that aimed to improve math achievement, as well as habilitate underlying cognitive deficits, in young children with FASD and prenatal alcohol exposure (‘PAE’). This project is a replication and an extension of the Math Interactive Learning Experience (MILE) program that was first conceptualized by researchers in Atlanta, Georgia, USA, and studied in a sample of American children with FASD (Kable et al., 2007; Coles et al., 2009). Habilitating cognitive abilities, including mathematics, does not just have the potential to improve academic outcomes. Improved mathematical cognition can greatly impact educational and occupational success as well as everyday wellbeing. The ability to tell and monitor time, manage money, understand cause and effect, monitor sequences of events, solve everyday problems, and respond appropriately are all related in part to mathematical cognition and can be negatively affected by poor math skills and limited understanding of mathematical concepts and reasoning (Kable et al., 2007). Thus, poor math skills may contribute to behavioral and adaptive problems. The findings of this study will impact intervention research, service delivery, and our basic understanding of Albertan and Canadian children with FASD and how to best meet their complex (and often unmet) health, psychological, and educational needs.

CHAPTER TWO

Literature Review

FASD: History, Terminology, and Diagnosis

In 1968, Lemoine and colleagues first noted patterns of abnormal morphogenesis and developmental delays in the offspring of alcoholic parents in France. Fetal alcohol syndrome (FAS) was formally identified by Jones and Smith in 1973 after recognizing a similar pattern of damage in case studies of infants born to mothers who chronically abused alcohol (Jones, Smith, Ulleland, & Streissguth, 1973). Since then, the term fetal alcohol spectrum disorder (FASD) has come to refer to a collection of physical, behavioral, cognitive, and psychosocial impairments that result from prenatal alcohol exposure (PAE). FASD is not a diagnostic designation, but rather an umbrella term used to describe the full range of characteristics experienced by individuals with PAE. Generally, a diagnosis on the fetal alcohol spectrum is assigned based on four main criteria: 1) growth deficiency in height and/or weight; 2) facial dysmorphology that may include an indistinct philtrum, short palpebral fissures, flat midface, short nose with a low nasal bridge, and thin upper lip; 3) damage to the central nervous system (CNS) as indexed by cognitive deficits, attentional problems, deficits in spatial and verbal memory, hyperactivity, motor difficulties, and/or microcephaly (Clarren & Smith, 1978); and 4) a confirmed history of maternal alcohol use during pregnancy.

Several diagnostic categories and systems have been developed to classify individuals exposed to alcohol prenatally. In 1996, the Institute of Medicine (IOM) in the United States proposed five different types of prenatal alcohol effects: FAS with and without confirmed in-utero alcohol exposure; partial FAS (pFAS), alcohol-related neurodevelopmental disorder (ARND), and alcohol-related birth defect (ARBD; Stratton, Howe, & Battaglia, 1996). An FAS diagnosis is applied to individuals prenatally exposed to alcohol who have the characteristic facial phenotype, growth deficiency, and neurobehavioral deficits. Diagnostic terms such as pFAS, ARBD, and ARND are assigned to those who lack some or all of the facial features and growth deficiency, but still experience significant neurobehavioral deficits.

Unfortunately, the IOM's diagnostic criteria are considered by many clinicians and researchers to be vague. Consequently, the Washington State FAS Diagnostic and Prevention Network built upon the IOM criteria to establish a rigorous, objective, and case-defined diagnostic coding system, now referred to as the 4-Digit Diagnostic Code (Astley, 2004). This system ranks diagnostic information in the areas of growth deficiency, facial phenotype, brain dysfunction, and maternal alcohol use. The magnitude of expression of each diagnostic feature is ranked independently on a 4-point scale, with a score of 1 reflecting the complete absence of the feature and a score of 4 indicating a strong or 'classic' presence of the feature. In Canada, Chudley et al. (2005) recommend a multidisciplinary approach to diagnosis where the 4-Digit Diagnostic Code is used to objectively describe and assess the four key FASD features while the IOM terminology (e.g., FAS, pFAS, ARND) are used to describe the resultant clinical diagnosis.

It is important to note that although FAS is considered to be the full clinical syndrome and perhaps the most serious presentation of an FASD, the presence of the facial phenotype and growth deficiency should not outweigh the functional significance of the neurobehavioral sequelae observed across the spectrum. The characteristic FAS facial features occur as a result of alcohol damage during a short period of vulnerability when typical facial structures develop early in the first trimester (Sulik, Johnston, & Webb, 1981). As a result, relatively few children who have been prenatally exposed to alcohol have all of the required facial features for diagnosis (Mattson, Riley, Gramling, Delis, & Jones, 1998). However, the brain develops throughout fetal development, providing a much larger period of time for the teratogenic effects of alcohol to disrupt the development of the CNS (Streissguth & O'Malley, 2000). Since the neuropsychological ramifications of PAE are much more functionally salient than the physical features, Chudley and colleagues (2005) advocate for thorough neuropsychological and multidisciplinary assessment, expanding the requirement of CNS damage to emphasize the importance of neurobehavioral assessment in diagnosing FASDs.

Common Neuropsychological Features in FASD

The majority of research efforts within the past 20 years have focused on the neurobehavioral impact of PAE, with specific attention to delineating the neuropsychological profile of individuals with FASD. Members of all diagnostic subgroups experience some degree of neurobehavioral impairments such as deficits in learning, memory, intelligence, language, visuospatial ability, motor-function, academics, adaptive behavior, attention, and executive function (EF) as a result of brain damage caused by PAE (for reviews, see Kodituwakku, 2009; Mattson et al., 2011). The following sections will review the research documenting patterns of EF, working memory, visuospatial functioning, and mathematics impairments in individuals with PAE and FASD.

Executive functioning and FASD. Executive functioning (EF) has been identified as a core area of deficit in FASD (for a review, see Rasmussen, 2005). EF refers to a number of complex and interdependent cognitive processes that are essential to purposeful, goal-directed behavior. These processes include planning, inhibition, working memory, set-shifting, attention, organization, and self-monitoring (Zelazo & Müller, 2002). Unsurprisingly, individuals with FASD demonstrate deficits in a variety of areas of EF, including cognitive flexibility, response inhibition, verbal reasoning, concept formation (Mattson, Goodman, Caine, Delis, & Riley, 1999; Schonfeld, Mattson, Lang, Delis, & Riley, 2001; Stratton et al., 1996), attention, strategy use, cognitive planning (Kodituwakku, Handmaker, Cutler, Weathersby, & Handmaker, 1995), set-shifting (Kodituwakku, Kalberg, & May, 2001), affective decision-making (Kully-Martens, Treit, Pei, & Rasmussen, 2013), and working memory (Rasmussen & Bisanz, 2009). Deficits in EF exist beyond what would be predicted based on IQ (Carmichael Olson, Feldman, Streissguth, Sampson, & Bookstein, 1998; Kodituwakku et al., 2001; Mattson et al., 1999; Schonfeld et al., 2001; Stratton et al., 1996), are noted in individuals across the fetal alcohol spectrum (Kodituwakku et al., 2001; Mattson et al., 1999; Schonfeld et al., 2001), and predict deficits in moral maturity (Schonfeld, Paley, Frankel, & O'Connor, 2006) and adaptive behavior (Ware et al., 2012). Tests of EF have also been found to be among the most effective in distinguishing children with heavy PAE from their non-exposed peers and from children with ADHD (Mattson et al., 2013). Accordingly, understanding EF deficits in individuals with FASD has become increasingly important in assessment, diagnostic, and intervention efforts.

Working memory and FASD. Baddeley (1992) defined the predominant model of working memory as a three-component system for short-term storage and manipulation of information. The primary component is the *central executive*, which is responsible for the simultaneous processing, manipulation, and preparation of information for storage (Baddeley, 1996). The central executive regulates the secondary memory systems, including the *visuospatial sketchpad* (used for holding and manipulating visual and spatial information; 'visuospatial working memory') and the *phonological loop* (for maintaining and rehearsing verbal information; 'phonological working memory') (Baddeley, 1992). In 2000, Baddeley updated the three-component model to include a fourth component, the *episodic buffer*, which is a limited-capacity subsystem that integrates and temporarily stores information from the other two subsystems and long-term memory. However, the preponderance of FASD working memory research focuses on the first three components. It is important to note that some researchers define working memory as a specific component of EF. However, others consider only the central executive component of working memory to involve EF processes. In this paper, I will generally refer to working memory separately from EF, although at times the central executive component of working memory will be discussed within the framework of EF, particularly as it relates to mathematics.

Evidence of impairment in the components of Baddeley's model of working memory in children with FASD has been documented in several studies (for a review, see Manji, Pei, Loomes, & Rasmussen, 2010). The central executive component of working memory—again, generally considered most related to EF—can be examined using backward digit span tasks. For example, Carmichael Olson and colleagues (1998) found that adolescents with FAS exhibited significant difficulty with backward digit span tasks when compared same-age, IQ-matched controls. Rasmussen and Bisanz (2009) documented backward digit span scores in children with FASD that were significantly below the normative mean, but these scores did not differ significantly from controls.

Phonological working memory dysfunction in children with FASD has been documented in both longitudinal and cross-sectional studies, primarily through use of forward digit span tasks. These deficits seem to be present in both young children and adolescents with FASD, and manifest beyond what would be expected based on IQ (Carmichael Olson et al., 1998). Burden and colleagues (2005) followed a cohort of 337 children with varying levels of PAE from the age of 6 months to 7 years. The amount of PAE was related primarily with poor phonological working memory at age seven as measured by a forward digit span task. Further, in their landmark longitudinal study of nearly 500 children with FASD, Streissguth and colleagues (1990) found that of all subtests on the Wechsler Intelligence Scale for Children: Third Edition (WISC-III), digit span was most related to alcohol exposure. Similarly, Jacobson et al. (1998) found that there were no significant relationships between PAE and IQ scores on the WISC-III among children exposed to moderate levels of alcohol in-utero, yet significant deficits were found on the Digit Span subtest. Rasmussen and Bisanz (2009) examined additional measures of phonological working memory, including memory for words and non-words, and found that children with FASD scored significantly lower than controls.

Children prenatally exposed to alcohol have also demonstrated difficulty with visuospatial working memory. On the Cambridge Neuropsychological Tests Automated Battery (CANTAB), children with PAE scored lower than controls on the Spatial Working Memory and Spatial Span tasks, both of which depend heavily on effective functioning of the visuospatial sketchpad (Rasmussen, Soleimani, & Pei, 2011). However, on a block recall test, Rasmussen and Bisanz (2009) found that although young children with FASD performed in the normative low average range, their performance was not significantly different from that of same-age controls.

Together, the preponderance of the research in this area suggest that children with PAE and FASD may exhibit deficits in visual and verbal working memory, as well as in the central executive component of working memory thought highly related to EF, as measured across a variety of tests.

Visuospatial functioning and FASD. Deficits in visuospatial perception and construction have been documented in children prenatally exposed to alcohol. Studies utilizing simple visuospatial tests of line orientation have yielded mixed results. Kaemingk and Halverson (2000) found that children prenatally exposed to alcohol performed significantly worse than same-age controls on a test of line orientation, whereas Rasmussen and colleagues (2012) found no differences between children with FASD and their same-age peers. On a clock-drawing task, Uecker and Nadel (1996) found that although children with FAS could replicate the essential features of a clock, they disregarded details like number spacing. Similarly, on a hierarchical processing task consisting of large global features made up of smaller local features, Mattson et al. (1996) found that children with PAE had significantly more difficulty processing the detailed local features than the global features. In addition, individuals with PAE have exhibited impaired performance on tests of visuospatial reasoning (Carmichael Olson et al., 1998; Hunt, Streissguth, Kerr, & Carmichael Olson, 1995) and visual abstract reasoning (O'Callaghan, O'Callaghan, Najman, Williams, & Bor, 2007).

Design copying is another area of difficulty for children with PAE, perhaps related to visual perception and visual motor integration deficits. For example, several studies have documented deficient performance on the Beery-Buktenica Developmental Test of Visual Motor Integration (e.g., Jirikowic, Carmichael Olson, & Kartin, 2008; Korkman, Autti-Ramo, Koivulehto, & Granstrom, 1998; Mattson et al., 1998). Tasks demanding more complex design copy, such as the Rey Complex Figure Test (RCFT), allow higher-level functions and the association of visual perception and construction with EF (e.g., attention, planning, and organization) and memory to be assessed. Pei and colleagues (2011) found that children with FASD exhibited significantly poorer RCFT design copy scores and poorer recall of the figure over time compared to same-age controls.

Children with FASD exhibited greatest difficulty with organizing the figural information during the initial copy stage, placing design elements correctly, and recalling the structural components of the figure over time. It appeared that children with FASD struggled to see the complex object as a whole or 'gestalt,' and integrate pieces of the design into a cohesive whole.

Mathematics and FASD. Several studies have documented difficulties with mathematics in children, adolescents, and adults with FASD and PAE (e.g., Carmichael Olson, Sampson, Barr, Streissguth, & Bookstein, 1992; Coles et al., 1991; J. Jacobson, Dodge, Burden, Klorman, & Jacobson, 2011; S. Jacobson et al., 2003; Lebel, Rasmussen, Wyper, Andrew, & Beaulieu, 2010; Nash et al., 2013; Rasmussen & Bisanz, 2010; Streissguth, Barr, Sampson, & Bookstein, 1994; Vaurio, Riley, & Mattson, 2011). Mathematics impairments in individuals with FASD have been a robust finding cross-sectionally and longitudinally, even after controlling for many variables (e.g., maternal drug use/nutrition/education, SES, IQ; see Rasmussen & Bisanz, 2009). Evidence for a specific deficit in mathematics in individuals with FASD is supported by research indicating that individuals with FASD have more difficulty with mathematics than other cognitive domains (Coles et al., 1991; Kerns, Audrey, Mateer, & Streissguth, 1997; Streissguth et al., 1991), and that mathematics impairment is more highly correlated with amount of PAE than other cognitive skills such as reading and spelling (Goldschmidt, Richardson, Stoffer, Geva, & Day, 1996; Streissguth et al., 1994). In a study comparing adolescents with alcohol-related facial dysmorphia with a group of children in special education, Howell et al. (2006) found that the special education students were more impaired in spelling and basic reading, whereas the alcohol-exposed children were more impaired in mathematics. Mathematics deficits in individuals with heavy PAE also tend to be greater than what may be predicted based on global cognitive functioning (i.e., IQ) (e.g., Vaurio et al., 2011), and may be dose-dependent (e.g., Goldschmidt et al., 1996; Streissguth et al., 1994).

Longitudinal research has shown that functional deficits in mathematical cognition and achievement persist from childhood (age four) through to adulthood

In their landmark longitudinal study, Streissguth and colleagues followed 500 offspring selected from 1,529 pregnant women from birth to age 14. Approximately half of the 500 mothers were classified as heavy drinkers, and the other half as infrequent drinkers or alcohol abstainers. Various outcomes (e.g., IQ, academic achievement, neurobehavioral ratings) were assessed at age ages 4, 7, 11 and 14 years. The authors examined the effects of PAE on these various outcomes while controlling for variables such as maternal drug and nicotine use, maternal nutrition, familial socio-demographic and educational factors, and other life stresses. At age four, PAE was most highly correlated with the Arithmetic subtest of the Wechsler Preschool and Primary Scales of Intelligence (WPPSI; Streissguth, Bookstein, Sampson, & Barr, 1989). Similarly, at age seven, the Arithmetic subtest was most strongly related to PAE on both the Wide Range Achievement Test (WRAT) and the Wechsler Intelligence Scale for Children–Revised (WISC-R; Streissguth et al., 1990). At age 11, Carmichael Olson and colleagues (1992) found that of all the national achievement test scores (arithmetic, reading, language, spelling), arithmetic was most related to PAE. The Arithmetic subtest of the WISC-R was again most related to PAE at age 14 (Streissguth et al., 1994). Over 90% of the children with FASD who exhibited poor arithmetic performance as 7-year-olds still experienced difficulty with arithmetic at 14. Finally, as adolescents and adults, Streissguth et al. (1991) found that those with FASD scored at a grade two arithmetic level on the WRAT-R.

In another longitudinal study, Goldschmidt and colleagues (1996) studied academic achievement (using the WRAT-R) in the six-year-old offspring of mothers who reported drinking heavily during pregnancy. After controlling for prenatal and postnatal drug use, maternal psychosocial and sociodemographic characteristics, and family arrangement, the authors found that second trimester drinking predicted difficulties in reading, spelling, and arithmetic. After controlling for IQ, PAE was still significantly related to deficits in arithmetic, but not spelling or reading. Similarly, in the Detroit longitudinal cohort, Jacobson et al. (2004) found that PAE was most strongly related to the WISC subtests of Arithmetic and Digit Span.

Aside from arithmetic, adolescents and adults with FASD have also been found to be impaired on tests of number processing, including measures of cognitive estimation, proximity judgment, and calculation. Kopera-Frye and colleagues (1996) asked participants with FASD and controls matched on gender, age, and education (ages 12 to 44) to read and write numbers, circle the larger of two numbers, and calculate exact and approximate arithmetic problems. Cognitive estimation was examined by asking participants to provide reasonable estimates of qualities of various items (e.g., what is the length of a dollar bill). Finally, the authors measured proximity judgment by asking participants to identify which of two numbers was approximately the same quantity as the target number (e.g., 11: 13 or 21). Despite exhibiting intact number reading, writing, and comparison skills, the FASD group made significantly more errors than controls on tests of cognitive estimation, proximity judgment, approximate subtraction, and exact calculation of addition, subtraction, and multiplication problems. Jacobson et al. (2003) used a similar battery with 13-year-olds and found that PAE was related to exact addition, subtraction, and multiplication, approximate subtraction and addition, proximity judgment, and number comparison. Based on these studies, it would appear that math deficits in FASD extend from calculation and arithmetic to also encompass estimation and magnitude representation.

Both individuals with FASD and those with PAE but no clinical diagnosis appear to show difficulty with mathematics. Coles et al. (1991) compared cognition and academic achievement in three groups of children aged 5 to 9 years: non-exposed controls, children exposed to alcohol throughout pregnancy, and children whose mothers ceased drinking during the second trimester. Children in both alcohol-exposed groups exhibited poorest performance on the math subtest of the Kaufman Assessment Battery for Children (K-ABC). Their math scores were not significantly different from each other, but were significantly lower than that of the control group. Math was the only area in which the group whose mothers stopped drinking during the second trimester performed significantly poorer than the control group. In other words, math was the only achievement measure that appeared to be negatively affected in individuals exposed to lower doses of alcohol. A recent study reviewing neuropsychological assessment files from an FASD diagnostic clinic found that both children with FASD and PAE had below average overall mathematics achievement as measured by the Wechsler Individual Achievement Test (WIAT; Nash et al., 2013). However, children with FASD scored significantly lower and were more likely to have scores in the clinical range in the numerical operations and math reasoning domains.

Relationship Between Mathematics and Other Cognitive Functions in FASD

Deficits in the cognitive functions described above (i.e., EF, working memory, visuospatial functioning) may contribute to the deficits observed in mathematics in individuals with FASD and PAE. Although EF, working memory, and visuospatial functioning have been related to math achievement in typical development and in math deficient groups, most of the research documenting the cognitive correlates of math performance in FASD have focused primarily on various aspects of working memory. In this section, I will briefly describe links between EF, working memory, and visuospatial processing and mathematics both in typical and atypical development and, where possible, in PAE and FASD.

Mathematics and executive functioning. EFs are believed to underlie a number of higher-order cognitive abilities, including mathematics. For example, *set-shifting*, or the ability to switch between sets, tasks, or strategies is hypothesized to aid math performance by supporting alternation between problem-solving strategies involved in multi-step math problems (e.g., Bull, Espy, & Wiebe, 2008), or by shifting between arithmetic operations (Jansen, De Lange, & Van der Molen, 2013). Some studies have found that the ability to shift between mental sets predicts math achievement (Bull et al., 2008) and poor shifting abilities have been documented in children with math disabilities (Bull & Scerif, 2001). *Inhibition* (the ability to suppress prepotent responses in favor of more goal-appropriate responses) has also been connected to math achievement (e.g., Blair & Razza, 2007) and poor inhibition has been found in children with poorer math ability (Bull & Scerif, 2001; Toll, Van der Ven, Kroesbergen, & Van Luit, 2011). Inhibition may assist in math problem solving by suppressing immature strategies and task-irrelevant information. *Attention* is another EF related to math performance in typical and math-deficient populations. Both cognitive and behavioral measures of inattention have been significantly correlated to mathematical performance in children with math difficulties (e.g., Gold et al., 2013; Lindsay, Tomazic, Levine, & Accardo, 2001). Although EFs and math have been theoretically and experimentally connected in the developmental literature, there is no research explicitly documenting the relationship between EF (aside from the central executive component of working memory) and mathematics in individuals with FASD. Only Rasmussen and Bisanz (2009) have examined the connection between a working memory measure of EF and mathematics in FASD, finding a significant relationship between the central executive component of working memory (backward digit span) and mathematics.

Mathematics and working memory. Previous research has shown that working memory is critical to successful mathematics performance (for a review, see Raghubar, Barnes, & Hecht, 2010). Working memory plays an important role in integrating information required for mathematical problem-solving, and 'holding' information required to complete mathematics tasks (e.g., copying off the board, remembering previous steps, entering numbers into a calculator, etc) (Swanson, 2004). Working memory is also required to represent and articulate numbers during counting and calculation, and disruptions during this level may lead to secondary deficits in higher numerical processes.

In typically developing children, longitudinal studies have documented a significant relationship between working memory and mathematics in kindergarten, even when controlling for early numeracy skills (Welsh, Nix, Blair, Bierman, & Nelson, 2010). Furthermore, growth in working memory has been significantly correlated to growth in mathematics in early elementary school children (Van der Ven, Koresbergen, Boom, & Leseman, 2012). In a recent longitudinal study, Toll and colleagues (2011) found that working memory tasks strongly predicted math learning disabilities in early elementary age children, even when preparatory math abilities were considered. The authors suggested that working memory ability might actually aid in the early identification of children at risk for math learning disabilities. Children who exhibit math learning disabilities tend to exhibit deficits on measures of visuospatial working memory and the central executive (Gathercole & Pickering, 2000; McLean & Hitch, 1999). In adolescents with mild to borderline intellectual disability, visuospatial working memory was positively and significantly related to addition and subtraction performance (Jansen et al., 2013).

For children with FASD, deficits in math and working memory appear to be intertwined. As reviewed previously, several longitudinal and cross-sectional studies found that measures of working memory that utilized numerical information (e.g., digit span or arithmetic tasks on the Weschler scales) were significantly impaired in children, adolescents, and adults with FASD. Rasmussen and Bisanz (2009) found that children aged four to six years with FASD were impaired on tests of mathematical problem solving and quantitative concepts. These deficits were highly correlated with measures of working memory, particularly measures of phonological working memory and the central executive, suggesting that mathematical difficulties in young children with FASD may be related to underlying working memory deficits.

Mathematics and visuospatial functioning. The link between visuospatial functioning and math abilities has been reported in typical and special populations (e.g., Assel, Landry, Swank, Smith, & Steelman, 2003). Several visuospatial skills are required for successful math performance, including visuomotor abilities, visuospatial organization, visual discrimination, and long-term visuospatial memory (Assel et al., 2003). Visuospatial skills support the ability to spatially represent and interpret numerical information (Geary, 1993). Particularly in early math acquisition, children interact with objects to acquire eventual internalized mental representations of numeracy. Visuospatial processing deficits may also affect the ability to interpret symbols and understand spatial-representation, both of which are required to process fundamental math concepts (Geary, 1993). For instance, a child with visuospatial difficulties may find it difficult to understand geometry, measurement, place value, and align columns for calculation. However, there is no literature explicating the link between mathematics and visuospatial functioning in children with PAE/FASD.

The Math Interactive Learning Experience (MILE) Program

Intervention literature for children with brain damage suggests that habilitative efforts should target underlying neurocognitive factors of functionally relevant skills within the context of those functional skills (Laugeson et al., 2007; Ylvisaker, 1998, as cited in Kable et al., 2007). Therefore, a mathematics intervention should focus on developing math skills while accommodating and attempting to habilitate underlying problems with cognition (i.e., EF, working memory, visuospatial functioning), rather than repeating tasks aimed solely to improve cognition with the hope improvements will generalize to math achievement (Kable et al., 2007). This theory forms the basis for the Math Interactive Learning Experience (MILE) program, which was developed and evaluated by Kable et al. (2007) and Coles et al. (2009) in Atlanta, Georgia, USA.

MILE was piloted with 61 children with FAS and pFAS aged 3-10 years. Children were first given a neurodevelopmental evaluation, and their parents attended two two-hour training workshops that provided education about FASD and how to support positive behavioral regulation skills in children. Following this, children were randomly assigned to either the MILE group (n = 31) or the standard psychoeducational treatment contrast group (n = 30). The contrast group received the neurodevelopmental evaluation and general educational consultation. In addition to the standard psychoeducational treatment, the MILE group also received six weeks of intensive, interactive, and individualized one-on-one tutoring. Caregivers of MILE participants also received training in how to support math learning at home (e.g., integrating math into play or everyday activities, completing homework assignments, etc). Math proficiency was measured by combining raw scores from the Test of Early Mathematics, selected math subtests from the Bracken Basic Concept Scale Revised, and the Number Writing Task. Post-intervention, both groups demonstrated gains in math knowledge but significantly higher gains were observed in the MILE group. In addition, children in the MILE group were significantly more likely to demonstrate a clinically significant gain (i.e., one standard deviation) from pre- to post-test. Six months post-intervention, children in the MILE group (n = 28) again exhibited significantly greater scores on the math outcome measures than the contrast group (n = 26), and the scores exhibited were higher than at the initial post-test (Coles et al., 2009). In the MILE group, those who made the greatest treatment gains were more likely to be younger. Among the entire sample, those who made gains had higher levels of alcohol-related dysmorphia. Gender, IQ, and SES did not appear to have an impact on treatment outcome.

Present Study

Although it is clear that FASD occurs frequently and leads to challenging neurodevelopmental difficulties, there has been limited attention to developing methods to improve child outcomes. Math achievement, in particular, appears to be significantly negatively affected in children with PAE and FASD. These difficulties tend to persist across the lifespan, likely contributing to a host of behavioral and independent adaptive living problems. Furthermore, little research has examined the connection between mathematics and its cognitive underpinnings in this population.

The original MILE pilot study provided an important first step in better understanding math difficulties in children with FASD, as well as hope that these issues can be remediated. The present study sought to replicate and *extend* the MILE study with a sample of Canadian children prenatally exposed to alcohol. We made several changes to the original MILE study design. First, the original MILE pilot project was not structured in a way that allowed the authors to determine which aspects of the program were more or less responsible for the treatment effect. Kable et al. (2007) acknowledged that one-on-one instruction alone might have been sufficient to effect change, regardless of the extra strategies utilized in the program (e.g., parent training). Understanding the relative contributions of various aspects of treatment is important to determine how MILE can be streamlined to ensure ease of community translation, efficiency, and cost-effectiveness. Therefore, the present study eliminated the significant parental component and utilized child tutoring only. Second, MILE was also developed and evaluated in a laboratory context rather than in an ecological setting (i.e., a school). It is not known what type of effects would be observed if the intervention was implemented in a more relevant context, let alone whether implementation in a different context would even be feasible. In the present study, we conducted all interventions in either the school or home setting. Third, in the original MILE study, the researchers did not examine if the theoretical model (i.e., targeting underlying neurocognitive deficits) was integral to the treatment effect. The present study investigated if underlying cognitive abilities (i.e., EF, working memory, visuospatial functioning) were also improving to attempt to add credence to the theoretical model upon which MILE was designed. Fourth, instead of using a 'sham' intervention contrast group, we used a behavioral comparison intervention. Fifth, the original MILE study utilized an American sample of children with FAS and pFAS. The participants in the present study included Canadian children with diagnoses across the fetal alcohol spectrum, as well as alcohol-exposed children without a clinical diagnosis.

Research Question 1: What are the mathematic and cognitive characteristics of our total alcohol-exposed sample, and how does math achievement relate to other cognitive characteristics?

This primarily descriptive question will establish if we are dealing with a math-deficient group with other cognitive challenges. We would also like to explore possible relationships between math and various cognitive abilities in alcohol-exposed children. This will establish if our sample is comparable to the extant literature, and provide the first examination of the relationship between EF and math in children with PAE/FASD.

Hypothesis. Our sample will exhibit below-average performance in math. Math achievement will be strongly correlated with measures of EF, working memory, and visuospatial functioning.

Research question 2: Do children with PAE/FASD in a modified MILE intervention program improve in mathematics compared to children with PAE/FASD in a different intervention?

Hypothesis: Children with PAE/FASD in the modified MILE intervention program will show greater improvements in mathematics compared to children with PAE/FASD in a different intervention.

Research question 3: Does the MILE intervention also improve other cognitive abilities?

Hypothesis: Children in the MILE intervention will improve other cognitive skills (EF, working memory, visuospatial functioning) from pre- to post-test when compared to children in the contrast intervention.

Research question 4: Are there any specific participant characteristics that seem to influence treatment outcomes?

Hypothesis: Based on the findings of Coles et al. (2009), younger children will show greater change than older children, as will children with a diagnosis of FASD compared to those with just PAE. Gender, IQ, and SES will be unrelated to any observed treatment effects.

CHAPTER THREE

Method

Participants

Seventeen children aged 4 to 10 with PAE and FASD participated: 11 in the MILE program and 6 in the contrast intervention. The groups did not differ significantly on any demographic variables.

Table 1. *Participant Characteristics*

Participant Characteristic	Math Group (n = 11)	Contrast Group (n = 6)	*p*
Age in years (M [range])	7.6 (4-10)	7.5 (6-9)	0.90[a]
Sex [n male (%)]	5 (45.5%)	3 (50.0%)	0.86[b]
Diagnosis [n FASD (%)]	5 (45.5%)	2 (33.3%)	0.63[b]
Full-Scale IQ [M (SD)]	87.8 (13.3)	101.2 (19.7)	0.12[a]
Length of intervention (days) [M (range)]	37.5 (32-6)	37.7 (33-44)	0.94[a]
Time from pre-test to post-test (days) [M (range)]	66.6 (49-85)	66.3 (53-89)	0.97[a]
Current living arrangement [N (%)]			0.48[b]
Adoptive parents	9 (81.8%)	4 (66.7%)	
Foster care	2 (18.2%)	2 (33.3%)	
Lifetime number of living situations [M (range)]	2.7 (1-6)	4.0 (2-6)	0.14[a]
Current caregiver characteristics			
SES [M (SD)]	47.0 (7.2)	38.8 (8.7)	0.06[a]
Annual income [N (%) > than $50,000]	9 (81.8%)	5 (83.3%)	0.21[b]
Education [N (%) one parent graduated high school]	10 (90.9%)	6 (100%)	0.45[b]

Note. SES was obtained from the primary caregiver using the Hollingshead Four Factor Index of Social Status. Possible scores range from 8 to 66. Full-Scale IQ reported as a standard score using the Wide Range Intelligence Test (WRIT).
[a]Analyzed with ANOVA; [b]analyzed with chi-square

Participants were recruited using convenience sampling in a two-pronged approach. Both approaches involved obtaining cooperative activities approvals from Edmonton Public, Edmonton Catholic, and Black Gold school boards. First, children were recruited through the Glenrose Rehabilitation Hospital FASD clinic and Elves Early Education and Respite Programs for children with FASD. Once interested participants contacted us (the researchers), we contacted the potential participants' educators to determine if in-school tutoring was feasible. If in-school tutoring was not permissible, students were tutored in their homes. Second, we contacted various schools within the Edmonton Public School District and Black Gold School Districts to determine if the school had students with PAE/FASD and would like to participate. If the school agreed, identified students received information and could contact researchers if interested. We then made arrangements with the school for tutoring. Informed consent was obtained from legal guardians, and assent was obtained from children.

After consent, it was confirmed if children had a diagnosis of FASD in accordance with the Canadian Guidelines for FASD diagnosis (Chudley et al., 2005). Children without diagnoses established within the Canadian guidelines but with prenatal alcohol exposure (confirmed through affidavit) were labeled as 'PAE.' A diagnosis of FASD is most frequently deferred if a child was too young at time of assessment to fully assess all domains of brain functioning (e.g., EF) with objective standardized testing measures. For this reason, clinics such as the Glenrose FASD clinic generally decline to assess for FASD until at least six or seven years of age. Other reasons for rejecting or deferring a diagnosis of FASD may include lack of evidence of significant brain damage, or because of the confluence of multiple etiological factors, including environmental influences and/or other clinical diagnoses. It is common for children prenatally exposed to alcohol to exhibit other diagnostic comorbidities, particularly ADHD (e.g., Clark, Lutke, Minnes, & Ouellette-Kuntz, 2004; Fryer, McGee, Matt, Riley, & Mattson, 2007; Kully-Martens et al., in preparation; Streissguth et al., 1996). Thus, participant with common mental health comorbidities (e.g., ADHD, ODD, etc) were included. We did not exclude any children from our study, although we would have excluded participants with significant neurological or medical conditions that could have precluded benefit from this intervention (e.g., autism).

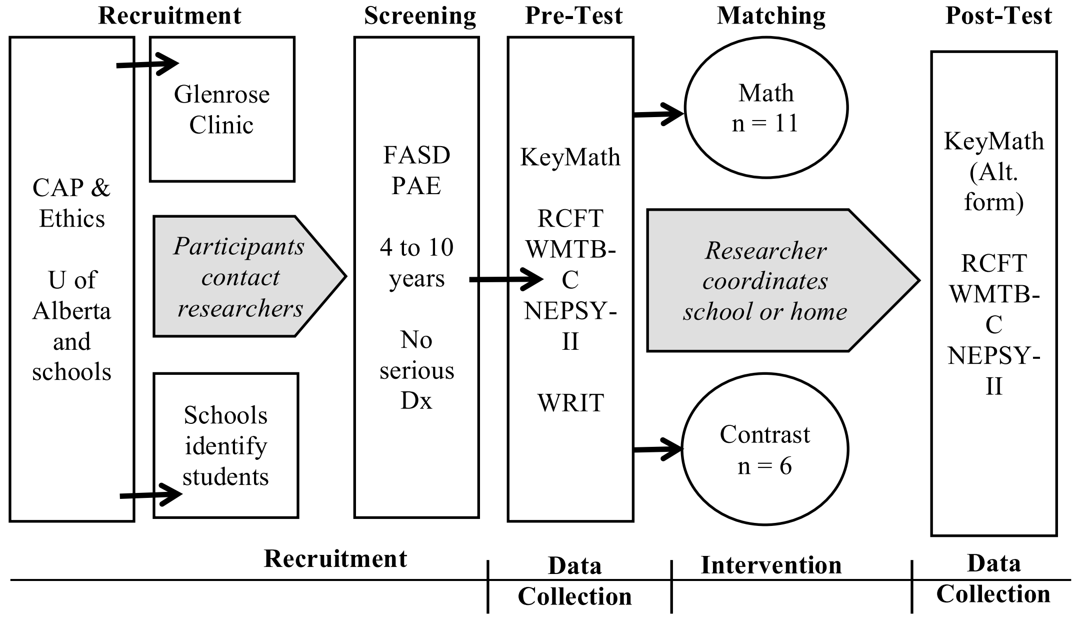

Figure 1. Schematic representation of research methodology.

Procedure

Participants first underwent a three-hour pre-test battery conducted in a research laboratory, which included assessment of IQ, mathematical achievement, and other cognitive abilities. Following pre-testing, children were assigned with matching to two intervention groups, first matching on age, then diagnosis, math achievement score, IQ, and gender where possible.

Both the mathematics and contrast intervention were tailored to individual child needs based on performance on pre-test measures. Intervention sessions began within four weeks of pre-testing. Each child received one-on-one individualized tutoring provided by a graduate student or a trained research assistant; the interventionist was kept consistent throughout the process with the exception of one participant. Intervention sessions were scheduled once or twice a week for a total of 10 sessions over 6-8 weeks. Scheduling incorporated input from the child's teacher and/or parent to minimize disruption. Post-testing occurred within 10 days of the last intervention, on average (range 2 to 21 days). The same measures were used at post-test, with the exception of IQ testing and demographic information. The post-test battery took approximately two and a half hours to complete, and was conducted by a research assistant who was blind to the child's treatment condition. Children received small prizes for participation (e.g., stickers), but no other form of remuneration.

Interventions

Interventionists. Two research assistants and one graduate student ('interventionists') performed the pre- and post-testing, as well as the intervention sessions. Typically, one interventionist would perform a child's pre-testing and intervention, and a different interventionist would conduct the child's post-testing to minimize bias. All interventionists had at least one academic degree in psychology and previous research experience with children with PAE/FASD. Dr. Jacqueline Pei, a registered psychologist, trained each interventionist to use and score (for research purposes) the measures in the neurocognitive battery. Dr. Julie Kable and Dr. Elles Taddeo, co-creators of the math intervention program, trained the three interventionists over the course of two-eight hour days in Atlanta, Georgia (January 2011).

Math intervention. MILE's theoretical premise is that intervention should focus on accommodating and habilitating the underlying neurodevelopmental deficits that contribute to the math difficulties within the context of math tutoring. For example, each intervention session incorporated a slow pace of instruction and an interactive method of teaching to compensate for deficits in EF and working memory and to ensure that important mathematical information (e.g., size, quantity, time, operations) could be fully processed and integrated. Working memory was additionally supported through use of small pieces of information, cues, repetition, and practice, and giving ample time for recall. Visuospatial processing deficits were accommodated by use of manipulatives and tools (e.g., a vertical number line to show that addition results in numbers going up, and subtracting results in numbers going down).

The interventionists played an integral role in the intervention because they were responsible for creating individualized curricula that emphasized strengthening absent and emerging skills to ensure a consistent sense of success.

Based on participant math pre-test performance, the interventionist would create a curriculum using worksheets from a learning bank. Worksheets addressed four different math domains: number and operations, measurement, geometry, and data analysis. Each domain had different areas of focus, and each area of focus had different levels from pre-K to grade 5. The interventionist set goals for a period of three weeks at a time to allow for goal review and amendment. Goals were designed to be inclusive. For instance, if a child was 'emerging' in his/her ability to use size, shape, quantity, counting, and patterns, an inclusive goal of 'patterns' would be selected because within the goal of patterning, instruction on size, shape, quantity, and counting could be easily incorporated.

Since certain preliminary skills need to be in place before a child can progress to doing higher order math, MILE focuses on slowly building a solid foundation of math skills instead of progressing rapidly to advanced levels. Therefore, interventionists worked with children at their respective developmental level, not necessarily their grade level, and more basic weak skills were addressed before higher-order skills. As an illustration, a child needs to be able to recognize the size and shape of an object before skills like sorting and categorization (conceptual precedents for skills such as addition, subtraction, multiplication, division, and eventually algebra and geometry) can develop. Problem-solving skills, understanding key concepts, and 'filling in skill gaps' were emphasized before math fact drills were utilized. This is because children with neurodevelopmental disorders generally do not acquire understanding through rote memorization. In addition, they are prone to forgetting and memory retrieval problems, so performance will be unstable and children may experience difficulty with acquiring future skills that require an understanding of 'lower level' skills.

Contrast intervention. Many individuals with PAE and FASD experience difficulties with various aspects of social behavior (for a review, see Kully-Martens, Denys, Treit, Tamana, & Rasmussen, 2012). In addition, the skills targeted in social skills remediation efforts do not generally overlap with mathematics. For these reasons, we selected a social skills intervention as our comparison to allow for the examination of gains in separate abilities between groups while still providing an intervention to all participants. Our intervention of choice, the Social Skills Intervention System—Intervention Guide, was developed for use with the Social Skills Intervention System (SSIS; Gresham & Elliott, 2010). Based on their SSIS performance (not reported in this paper) each child in the contrast group received an individualized social/behavioral intervention that focuses on several key areas of social skills difficulties and problem behaviors that often manifest in school settings, including communication, cooperation, assertion, responsibility, empathy, engagement, and self-control. More or less focus was given to certain areas based on the child's profile of needs and strengths.

Measures

Demographic questionnaire. Caregivers completed a brief researcher-constructed demographic form at pre-test. The demographic questionnaire gathered information about the child's age, grade, placement history, current living situation, and caregiver factors such as marital status, highest level of education, occupation, and household income bracket.

Mathematics. The Key Math 3 Diagnostic Assessment—Canadian Edition (KeyMath 3 DA; Connolly, 2007) was administered to participants at pre- and post-testing. Individual pre-test scores were used to determine the course of each math participant's mathematics intervention. The KeyMath 3 DA is a standardized and in-depth measure of essential mathematical skills and concepts in school-age children (4-21 years). The battery included 10 subtests measuring basic concepts such as early numeration, algebra, geometry, measurement, and probability awareness; operations such as mental computation, estimation, and written computation; and applications such as problem solving. Two parallel forms of the KeyMath 3 DA were used from pre- to post-test in order to minimize practice effects. Internal consistency, test-retest, and alternate-form reliabilities are high (upper .80s to .90s; Connolly, 2007). The KeyMath 3 DA's content validity is considered adequate, and good construct and concurrent validity is demonstrated by positive correlations (.60 to .90). Validity evidence for use with special populations (e.g., math disability, intellectual disability, etc) is also adequate.

Executive functioning. EF was measured primarily through selected subtests from the NEPSY–II (Davis & Matthews, 2010), which is a standardized neuropsychological battery for children aged 3-16 years. Auditory Attention, Response Set, and Design Fluency were administered to participants at pre- and post-test. The Auditory Attention task measures auditory selective and sustained attention by asking a child to point to stimuli based on auditorily-presented instructions. Response Set involves a similar set of stimuli as the Auditory Attention task, but shifting and inhibition of previous rules from the Auditory Attention task are required. Design Fluency assesses a child's ability to generate novel designs as quickly as possible using pencil and paper on structured and unstructured dot arrays. This provides a measure of initiation and cognitive fluency. Most NEPSY-II subtests demonstrate adequate to high internal consistency, good test-retest reliability (.57 to .84 for selected subtests), and high inter-rater reliability (.98 to .99; Davis & Matthews, 2010). The pattern of correlations between subtests on the NEPSY-II suggests that its construct validity is strong. In addition, the NEPSY-II demonstrates adequate concurrent validity of intellectual functioning and criterion validity in yielding diagnostic information. The NEPSY-II exhibits good discriminant validity across a variety of developmental disabilities, and has been successfully used to detect neurocognitive deficits in a sample of children with FASD aged 6-17 (Rasmussen et al., 2012).

Working memory. The Working Memory Test Battery for Children (WMTB-C; Pickering & Gathercole, 2001) was administered at pre- and post-testing. The WMTB-C assesses the three critical components of Baddeley's model of working memory (the central executive, the phonological loop, and the visuospatial sketchpad) in children aged 5-15. One measure each of the phonological loop (Digit Recall) and the visuospatial sketchpad (Block Recall), as well as two measures of the central executive (Backward Digit Recall, Counting Recall) were used. Test-retest reliability is .81 for Digit Recall, .62 for Backward

Digit Recall, .74 for Counting Recall, and .53 for Block Recall (Pickering & Gathercole, 2001). The WMTB-C has been validated against existing established tests of achievement.

Visuospatial functioning. Two tools were used to assess visuospatial functioning at pre- and post-test. The Block Construction and Geometric Puzzles subtests were selected from the NEPSY-II (described above). The Block Construction subtest measures visuoconstructional skills by asking a child to replicate increasingly complex pictorial representations of three-dimensional designs using blocks. The Geometric Puzzles subtest involves visuospatial analysis and mental rotation. Test-retest reliability is adequate for these selected subtests (.62 to .89). Second, the Rey Complex Figure Test (RCFT; Meyers & Meyers, 1995) was administered at pre- and post-testing as a measure of visuospatial processing, and visuospatial memory. The RCFT requires examinees to copy a complex geometric design with multiple embedded details, and then re-create the figure from memory. The RCFT exhibits strong inter-rater reliability (.94). Good convergent, construct, and discriminant validity has also been established through comparison with other neuropsychological or achievement tests. The RCFT has been used to measure visuospatial processing and memory deficits in a sample of children with FASD aged 6 to 12 (Pei et al., 2011).

Intelligence. The Wide Range Intelligence Test (WRIT; Glutting, Adams, & Shelow, 2000) was used at pre-test only as a brief and reliable measure of verbal and nonverbal cognitive abilities. The WRIT provides verbal (crystallized) IQ and visual (fluid) IQ scores, which together yield a measure of general IQ for individuals aged 4-85. Internal consistency reliability is high for the WRIT main scales and subscales (.84 to .90s; Glutting et al., 2000). Test-retest reliability is also high for the main scales (low to high .90s), but the subscale reliability coefficients are less favorable (.70 and higher). The WRIT has very high inter-rater reliability (.98 to .99). The WRIT has also demonstrated acceptable construct, concurrent, and predictive validity. For instance, although the WRIT is a brief test of IQ, it has been highly correlated with much lengthier IQ tests such as the WISC-III (.90) and moderately to highly correlated with achievement tests such as the WRAT (.36 to .64).

Enhancing Reliability and Validity

In experimental designs, equating of groups through matching helps to minimize internal validity threats of history, maturation, selection, interaction between selection and other threats (Creswell, 2012). Threats related to rivalry, resentful demoralization, and compensatory equalization were addressed by the inclusion of a ‘control’ treatment that was equally attractive as the experimental condition. Thus, benefits were equally distributed and neither group should have felt that they received a less desirable treatment than the other. Since diffusion could have occurred if students at the same school received different treatments and their parents discussed the treatments, students within the same school class received the same condition. To address threats related to testing and instrumentation, we allowed at least two months to elapse between pre- and post-test. We also used alternate test forms when possible (with counterbalancing), as well as consistent instrumentation and testing procedures. Mortality was not an issue, as all participants completed pre- and post-testing.

Threats to external validity were also considered. For instance, interaction of selection and treatment is common in studies with clinically-referred samples. It can be difficult to generalize results based on participants with clinical diagnoses from an urban area to individuals who are not receiving clinical services. We attempted to combat an interaction between setting and treatment by providing intervention to children in many schools and many school districts. We also attempted to minimize the interaction of history and treatment by providing intervention in fall semesters (beginning of school year), winter semesters (end of school year), and summertime.

Ethical Considerations

Ethics approval was obtained from the Human Research Ethics Board at the University of Alberta, and a cooperative activities agreement was approved between the University of Alberta and the Edmonton Public School Board and Edmonton Catholic Schools. This study respected the dignity of participants by sustaining the right to informed and ongoing consent and to privacy and anonymity. The child participants in this study were considered incapable of providing informed consent based on their youth and their cognitive disability. Therefore, legal guardians provided informed consent for children, and the child participants provided assent. Financial inducements were not made, as this could circumvent autonomy. Participant welfare was protected through a risk-benefit analysis. This study was considered at least minimal risk (risks of daily life) because it concerned a vulnerable population (i.e., children, individuals with cognitive disability; Creswell, 2012). The only likely risk of the present study was that children might have felt psychological or mental fatigue. To minimize discomfort, children were continually informed that they could stop for a break at any time during tutoring sessions. Children and guardians were also informed that they could withdraw at any time. This study also strived to treat all participants equitably. Although matched to groups, all participants received an intervention targeted at individual areas of need.

Two additional important ethical considerations were associated with this study. First, the school site where data collection took place had to be respected. We gained permissions to access the sites, respected how the site operated, and endeavored to cause minimal disruption. A second issue was sharing of information from the assessment measures. Cresswell (2012) states that participants have a right to benefit from the study and obtain information, and researchers should work actively to try and give back to them. However, the research battery was not constructed or conducted for clinical purposes, and thus individual scores could not be responsibly interpreted in a clinical manner. In place of this, group result summaries were provided to families and schools.

Data Analysis

Raw scores from each measure were entered into measure-specific computer scoring programs. Relevant data (e.g., raw scores, standard scores) were transferred into an SPSS data file, along with coded information from the demographic questionnaire. The initial phase of data analysis included analyzing the various demographic features of our sample. We compared the two groups for any demographic differences (e.g., age, sex, IQ, SES, number of living arrangements, current living arrangement) by using ANOVA for continuous data and chi-square for categorical data.

To answer the first research question, we first calculated our sample's mean pre-test norm-referenced scores for each measure. We then plotted these in comparison to the normative population mean. To answer the second part of our first research question, we ran correlations within our entire sample between KeyMath Total Achievement standard score and the norm-referenced scores of selected subtests of our cognitive measures (pre-test only).

Questions 2 and 3 were addressed by calculating the difference between pre- and post-test raw scores ('change scores') for each of the relevant measures and subtests. Raw scores were used because they tend to be more sensitive to change over a short period of time than standard scores. Change scores (i.e., changes in raw score over time) were used because there was a significant difference between the group pre-test scores. To investigate whether math performance improved after intervention, we analyzed the KeyMath outcome variables with a step-down procedure. We first analyzed the total KeyMath composite and the three sub-composites (Basic Concepts, Operations, Applications of Problem Solving) separately using four univariate analysis of variance (ANOVA) procedures. If significant group differences were found on any of the three composites, we entered the subtests comprising the composite into a multivariate analysis of variance (MANOVA). Further tests were not performed on composites in which no significant group differences were found. We examined if there were any changes in cognitive measures from pre- to post-test by entering the raw change scores for subtests comprising each cognitive domain into separate MANOVAs.

To answer our fourth and final research question, we examined how particular demographic variables related to the magnitude of the observed math treatment effect. For each intervention group, correlations were conducted between the KeyMath total score and the three KeyMath sub-composites and various predictor variables: age, sex, diagnosis, IQ, and SES. Pearson correlations were conducted for non-dichotomous variables (e.g., age, IQ, SES) and Point Biserial correlations were conducted for dichotomous variables (e.g., sex, diagnosis).

CHAPTER FOUR

Results

Research Question 1

Mathematics. We hypothesized that our alcohol-exposed sample would exhibit pre-test norm-referenced scores that were significantly below (i.e., >1 standard deviation) the normative population mean on each of our math and cognitive measures. On the KeyMath, our sample exhibited scores that were 1 standard deviation (SD) or more below the normative mean on all 10 of the KeyMath subtests, indicating that our sample was indeed math-deficient. Performance was highest on the Geometry subtest, and lowest on the Multiplication/Division, Addition/Subtraction, Mental Computation, and Foundations of Problem Solving subtests.

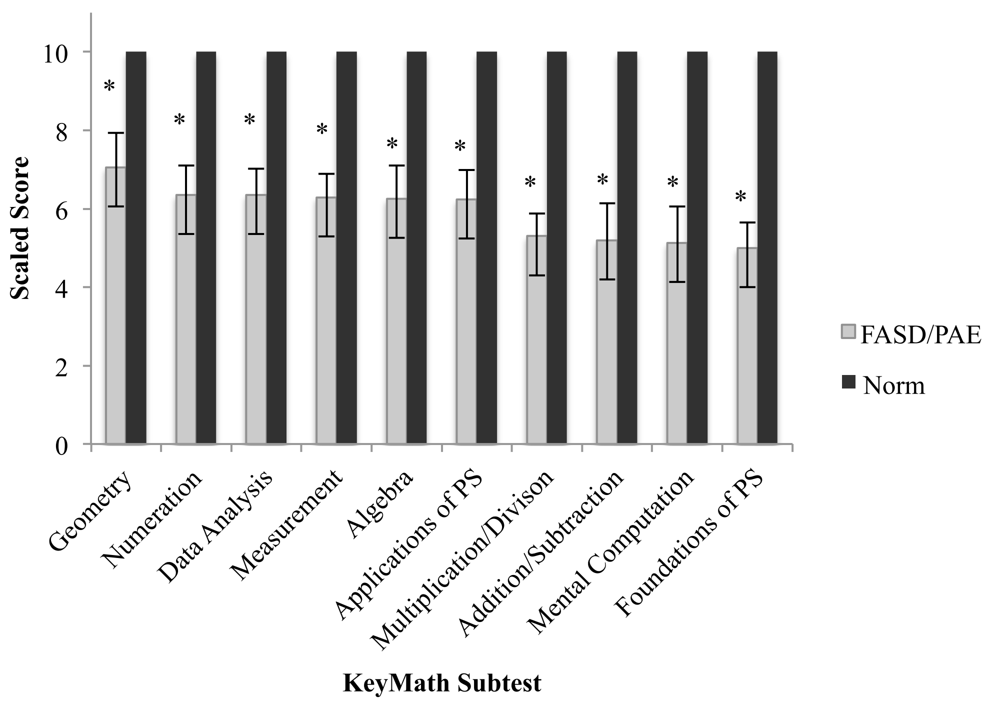

Figure 2. Math Pre-test Performance of Overall Sample vs. Normative Mean. Scaled scores have a mean of 10 and an SD of 3. Error bars represent standard error of the mean. Group subtest scaled score means based on 16 participants, except for Multiplication and Division (10 participants).
* > 1 SD below normative mean

Executive functioning. Three subtests were used from the NEPSY-II to provide a measure of EF: Auditory Attention, Response Set, and Design Fluency. Our sample exhibited scores more than 1 SD below the normative mean on both Auditory Attention and Response Set. However, performance on Design Fluency was within the average range compared to the normative mean.

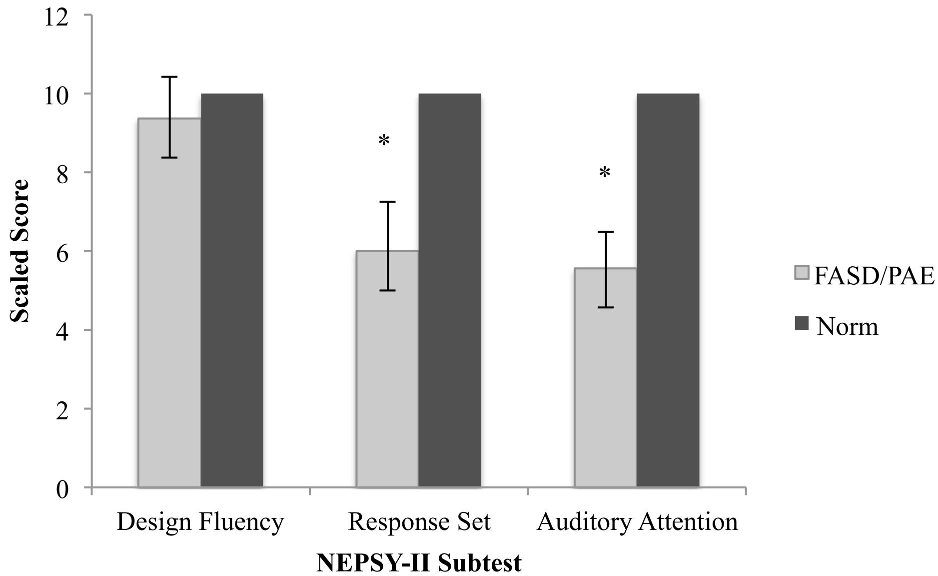

Figure 3. Executive Functioning Pre-test Performance of Overall Sample vs. Normative Mean. Scaled scores have a mean of 10 and an SD of 3. Error bars represent standard error of the mean. Group subtest scaled score for Design Fluency and Auditory Attention means based on 16 participants. Response Set calculated with 10 participants.
* > 1 SD below normative mean

Working memory. Of the four WMTB working memory subtests, our sample achieved highest scores on the measure of the phonological loop (Digit Recall), which was within the average range compared to the normative group. Poorest performance was observed on the two tests of the central executive (Backward Digit Recall, Counting Recall), with our sample performing nearly two SDs below the mean on Counting Recall, and over one SD below the normative mean on Backward Digit Recall. Performance on the visuospatial sketchpad subtest (Block Recall) was within one SD of the normative mean.

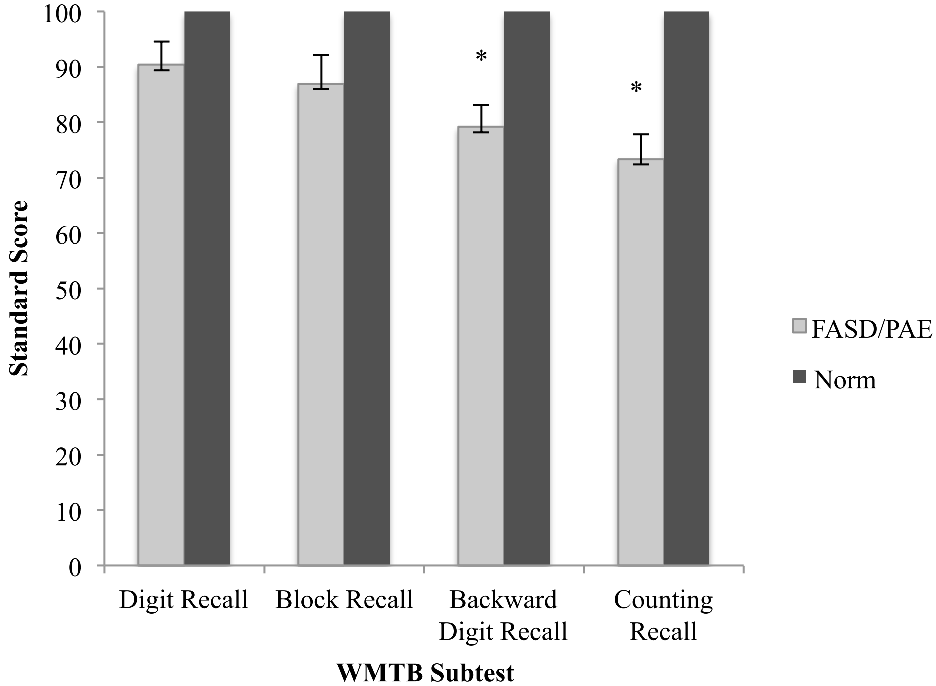

Figure 4. Working Memory Pre-test Performance in Overall Sample vs. Normative Mean. Standard scores have a mean of 100 and an SD of 15. Error bars represent standard error of the mean. All group subtest scaled scores based on 16 participants.
* > 1 SD below normative mean

Visuospatial functioning. Two subtests from the NEPSY-II provided a measure of visuospatial processing (Geometric Puzzles; Block Construction) and three tests from the RCFT estimated visuospatial memory (Immediate Recall, Delayed Recall, Recognition Recall). On the two measures of visuospatial processing, our sample performed quite similarly to the normative mean. Scores on the Immediate Recall and Delayed Recall tasks of the RCFT were more than 1 SD lower than the normative mean; however, the Recognition Recall score was within one SD of the normative mean.

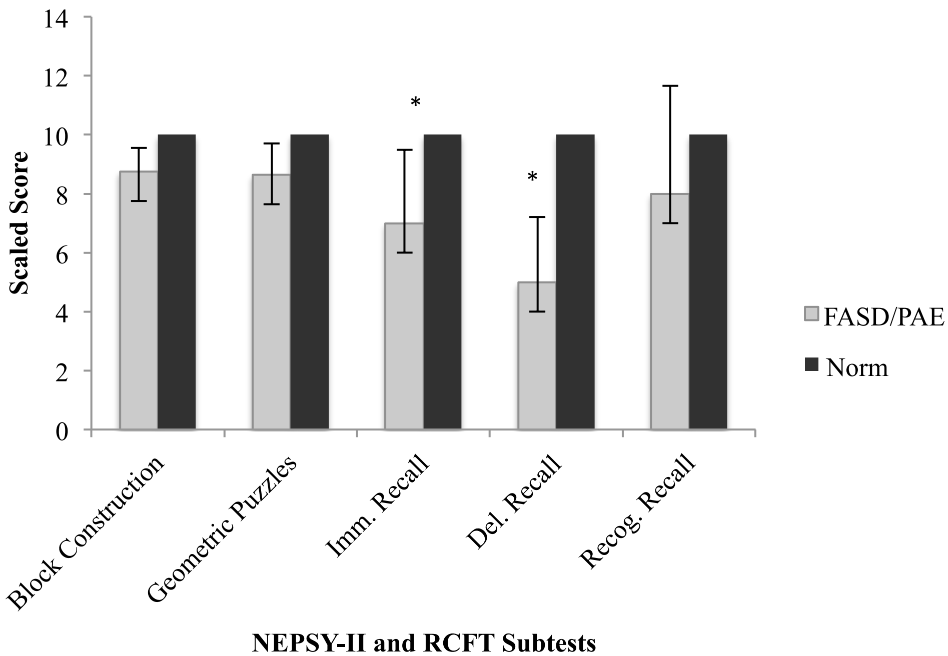

Figure 5. Visuospatial Functioning Pre-Test Performance in Overall Sample vs. Normative Mean. Scaled scores have a mean of 10 and an SD of 3. Error bars represent standard error of the mean. Group subtest scaled score for Block Construction based on 16 participants; Geometric Puzzles based on 11 participants. RCFT scores calculated with 15 participants. RCFT scores transformed from standard scores to scaled scores for ease of comparison.
* > 1 SD below normative mean

Relationship between math achievement and cognitive variables. The second objective of our first research question was to examine the relationship between mathematics, EF, working memory, and visuospatial functioning. All three of the EF measures were strongly and positively correlated with total math achievement (*r*s from .43 to .57; Cresswell, 2012), although the only relationship that was statistically significant was between total math achievement and Auditory Attention, $r(16) = 0.49$, $p < 0.05$. Similarly, strong positive correlations were observed between all of the working memory subtests and total math achievement (*r*s from 0.44 to 0.73). In particular, the two measures of the central executive (Counting Recall, $r(16) = 0.61$, $p < 0.01$; Backward Digit Recall, $r(16) = 0.73$, $p < 0.01$) were significantly correlated with total math achievement. Only one visuospatial subtest (Geometric Puzzles) yielded a strong (but non-significant) correlation with total math achievement, $r(11) = 0.59$, $p > 0.05$.

Table 2. *Correlations Between Total Math Achievement and Cognitive Constructs*

	KeyMath Total Achievement
Executive Functioning	
NEPSY-II Auditory Attention	0.49*
NEPSY-II Response Set	0.57
NEPSY-II Design Fluency (Total)	0.43
Working Memory	
WMTB Digit Span	0.49
WMTB Block Recall	0.44
WMTB Counting Recall	0.61**
WMTB Backward Digit Recall	0.73**
Visuospatial Processing	
NEPSY-II Block Construction	0.07
NEPSY-II Geometric Puzzles	0.59
Visuospatial Memory	
RCFT Immediate Recall	0.01
RCFT Delayed Recall	0.10
RCFT Recognition Recall	0.09

Note. Standard Scores (Pre-Test). Correlations with RCFT conducted with 15 participants. Correlations with WMTB conducted with 16 participants. NEPSY-II Auditory Attention and Response set are combined scaled scores. NEPSY-II Response Set correlations reported for 10 participants. NEPSY-II Geometric Puzzles reported for 11 participants. Other NEPSY-II subtest correlations reported for 16 participants.
** $p < 0.01$; * $p < 0.05$

Research Question 2

We hypothesized that children in the math intervention group would exhibit significantly greater raw point gains on the KeyMath than children in the contrast intervention. From pre- to post-test, children in the math intervention group significantly increased their total KeyMath score by an average of 14.27 raw points, compared to a 1.5 point increase observed in the contrast group, $F(1, 16) = 5.34$, $p < 0.05$, $\eta^2_{partial} = 0.83$. On the Basic Concepts composite, the math group demonstrated a significant gain of 8.36 raw points compared to the contrast group, who gained an average of 1.50 points, $F(1, 16) = 4.69$, $p < 0.05$, $\eta^2_{partial} =$

0.55. The overall MANOVA for the five subtests of the Basic Concepts composite was not significant, $F(5, 11) = 1.19$, $p > 0.05$. The math group did not gain significantly more raw points than the contrast group from pre- to post-test on either of the remaining two composites (Operations and Applications of Problem Solving).

Table 3. *Math Raw Difference Scores by Group*

KeyMath	Math Group (n=11)			Contrast Group (n=6)			p
	Pre	Post	Diff	Pre	Post	Diff	
Basic Concepts	29.8	38.2	8.4	48.3	49.8	1.5	0.04*[a] 0.37[b]
Numeration	8.3	9.9	1.6	11.3	11.8	0.5	
Algebra	3.5	4.4	0.9	7.0	7.2	0.2	
Geometry	8.4	10.3	2.0	12.5	11.5	-1.0	
Measurement	5.2	7.8	2.6	8.1	9.8	1.7	
Data Analysis/Probability	4.6	5.9	1.3	9.3	9.5	0.2	
Operations	5.9	8.4	2.5	14.0	14.2	0.2	0.19[a]
Mental Computation	2.7	3.7	0.8	5.5	5.7	0.2	
Addition/Subtraction	3.2	4.6	1.4	7.8	7.8	0.0	
Multiplication/Division	0.0	0.3	0.3	0.7	0.7	0.0	
Applications of Problem Solving (PS)	8.3	10.9	2.6	12.1	14.3	2.2	0.83[a]
Foundations of PS	3.7	5.7	1.8	5.5	7.3	1.8	
Applied PS	4.6	5.3	0.7	6.7	7.0	0.3	
Total	43.1	57.4	14.3	57.4	58.9	1.5	0.03*[a]

Note. Total possible raw score on the KeyMath3 DA is 372 raw points.
[a]Analyzed with ANOVA; [b] Analyzed with MANOVA
*$p < 0.05$

Research Question 3

Our third research question addressed whether participating in the MILE intervention would be related to improvements in other cognitive abilities, specifically EF, working memory and visuospatial functioning. We hypothesized that MILE participants would show improvements from pre- to post-test on measures of these skills compared to children in the contrast intervention. Raw change scores of three subtests from the NEPSY-II considered to measure EF (Auditory Attention, Response Set, and Design Fluency) were entered into one MANOVA. The overall EF MANOVA was not significant, $F(3, 6) = 1.20$, $p > 0.05$. Similarly, raw change scores from each of the four working memory subtests from the WMTB-C were entered into another MANOVA. Again, the overall working memory MANOVA was not significant, $F(4,12) = 1.30$, $p > 0.05$. The MANOVA for visuospatial processing was calculated using raw change scores from two NEPSY-II subtests (Block Construction and Geometric Puzzles) and the raw change score of the RCFT Copy trial. This overall MANOVA was not significant, $F(3,13) = 1.05$, $p > 0.05$. Finally, we used the three memory trials from the RCFT to calculate a visuospatial memory MANOVA. This MANOVA was also not significant, $F(3,13) = 0.81$, $p > 0.05$.

Table 4. *Cognitive Measures Raw Change Scores by Group*

	Math Group			Contrast Group			*p*
	Pre	Post	Diff	Pre	Post	Diff	
Executive Functioning							0.39
NEPSY-II Auditory Attention	14.1	21.6	7.5	22.0	22.7	0.7	
NEPSY-II Response Set	15.3	23.3	8.0	29.0	28.2	-0.8	
NEPSY-II Design Fluency	14.3	18.0	3.7	19.5	16.0	-3.5	
Working Memory							0.32
WMTB Digit Span	23.1	24.2	1.1	24.3	23.8	-0.5	
WMTB Block Recall	17.8	18.7	0.9	23.2	22.0	-1.2	
WMTB Counting Recall	8.4	10.9	2.5	13.2	12.7	-0.5	
WMTB Backward Digit Recall	6.2	6.6	0.4	7.5	7.8	0.3	
Visuospatial Processing							0.40
NEPSY-II Block Construction	9.9	12.3	2.4	10.7	11.2	0.5	
NEPSY-II Geometric Puzzles	19.6	21.5	1.9	21.8	25.5	3.7	
RCFT Design Copy	13.0	14.6	1.6	15.1	12.4	-2.7	
Visuospatial Memory							0.51
RCFT Immediate Recall	6.2	6.8	0.6	5.9	6.1	0.2	
RCFT Delayed Recall	4.4	5.0	0.6	5.8	4.4	-1.4	
RCFT Recognition Recall	16.9	18.1	1.2	17.5	19.2	1.7	

Note. p-value calculated using MANOVA

For the RCFT, four composite raw change scores were entered into the MANOVA: Copy, Immediate Recall, Delayed Recall, and Recognition Recall. The overall MANOVA was not significant, $F(4,12) = 0.64$, $p > 0.05$. Similarly, the raw difference scores for each of the four WMTB subtests were entered into a MANOVA. Significant group differences were again not observed in the overall MANOVA, $F(4, 12) = 1.30$, $p > 0.05$.

Research Question 4

Fourth and finally, we examined whether specific participant characteristics such as age, sex, or IQ were related to any observed math treatment effects. Based on the findings of Coles et al. (2009), we hypothesized that younger children would exhibit greater raw point gain than older children, and that gender, IQ, and SES would be unrelated to any observed math treatment effects. We also hypothesized that children with a diagnosis of FASD would show greater change compared to those with confirmed PAE only. Correlations between the total KeyMath composite and age, sex, diagnosis, SES, and verbal, visual, and overall IQ were conducted separately for each group.

Math and age. In the math group, a strong relationship between KeyMath total raw change score and age was observed, $r(11) = 0.52$, $p > 0.05$, indicating that older children tended to exhibit greater raw point gains. A weak relationship was observed between KeyMath total raw change score and age in the contrast group, $r(6) = 0.27$, $p > 0.05$.

Math and sex. Sex was not significantly related to KeyMath total raw change score in either the math group or the contrast group (both ps > 0.05).

Math and diagnosis. In the math group, children with a PAE diagnosis exhibited greater raw point gain, $r(11) = -0.53$, $p > 0.05$, than children with FASD. An opposite effect was observed in the contrast group, with children diagnosed with FASD tending to exhibit greater raw point gain, $r(6) = 0.58$, $p > 0.05$.

Math and IQ. In the math group, KeyMath total raw change score was significantly negatively correlated with Verbal IQ, $r(11) = -0.62$, $p < 0.05$, indicating that children with lower verbal IQ tended to exhibit greater raw point gains. In the contrast group, verbal IQ was moderately positively related to total math score, $r(6) = 0.32$, $p > 0.05$. Visual IQ was moderately negatively related to KeyMath total raw change score in the math group, $r(11) = -0.33$, $p > 0.05$, with children with lower visual IQ tending to exhibit higher math change scores. A strong positive relationship was observed between Visual IQ and KeyMath total raw change score in the contrast group, $r(6) = 0.61$, $p > 0.05$. Finally, a strong negative relationship was observed between overall IQ and KeyMath total raw change score in the math group, $r(11) = -0.55$, $p > 0.05$. In comparison, IQ was strongly related to KeyMath total raw change score in the contrast group, $r(6) = 0.53$, $p > 0.05$, with children with higher IQ exhibiting greater raw point gain.

Math and SES. SES was significantly, strongly, and negatively correlated with KeyMath total raw change score in the math group, $r(11) = -0.72$, $p < 0.05$, indicating that children of lower SES in the math group tended to exhibit greater point gain. A moderate positive relationship was observed in the contrast group between SES and math score, $r(6) = 0.34$, $p > 0.05$, indicating that children in the contrast group with higher SES tended to exhibit higher raw point gain.

Table 5. *Correlations Between Demographic Characteristics and Math Achievement (Math Group)*

	Math Composite			
	Basic Concepts	Operations	Problem Solving	Total Math
Age	0.60	0.79**	0.37	0.52
Sex	-0.09	-0.01	-0.31	-0.06
Diagnosis	-0.48	-0.78**	-0.36	-0.53
IQ				
Verbal IQ	-0.48	-0.68*	-0.72*	-0.62*
Visual IQ	-0.34	-0.28	-0.51	-0.33
General IQ	-0.46	-0.56	-0.69*	-0.55
SES	-0.80**	-0.71*	-0.49	-0.72*

**correlation significant at 0.01 level
*correlation significant at 0.05 level

Table 6. *Correlations Between Demographic Characteristics and Math Achievement (Contrast Group)*

	Math Composite			
	Basic Concepts	Operations	Problem Solving	Total Math
Age	0.28	0.24	0.56	0.27
Sex	0.03	-0.11	-0.05	-0.30
Diagnosis	0.27	-0.15	-0.07	0.58
IQ				
Verbal IQ	0.49	0.33	0.85*	0.32
Visual IQ	0.42	0.19	0.91*	0.61
General IQ	0.46	0.25	0.93**	0.53
SES	0.28	-0.45	0.24	0.34

**correlation significant at 0.01 level (2-tailed)
*correlation significant at 0.05 level (2-tailed)

CHAPTER FIVE

Discussion

The purpose of this study was to replicate and extend a mathematics intervention (the Math Interactive Learning Experience; MILE) developed especially for children with FASD. More specifically, we wanted to characterize the profiles of math and related cognitive functions in our sample, and determine if any relationships existed between math performance and related cognitions. Centrally, we sought to determine if the MILE intervention could be streamlined by eliminating a parent training component and providing only individual child tutoring in an ecological (i.e., home or school) setting. We also wanted to determine if the math intervention supported any changes in related cognitive domains such as EF, working memory, and visuospatial skills. Finally, we assessed the relationship between various demographic variables and the magnitude of the math treatment effect in order to help identify qualities of participants who may benefit more from the MILE intervention. In this section, the results from the current study will be interpreted and discussed. Implications, limitations, and directions for future research will also be presented.

Profile of Math Achievement and Other Cognitive Variables

Our sample exhibited math scaled scores that were at least one standard deviation below the normative average across all domains of mathematical functioning tested pre-intervention, indicating a math deficient group. Deficits were most pronounced on tests of mental computation and paper-and-pencil arithmetic, as well as problem solving. These findings add to the body of extant literature documenting math difficulties—particularly arithmetic difficulties—in children with PAE and FASD. Previous studies of math functioning in alcohol-exposed children have inferred mathematical functioning from math-related subtasks of intelligence tests, or from broader achievement batteries. This study provides the most comprehensive profile of mathematic strengths and weaknesses in this population due to use of a math-specific achievement test, which documents a wide range of specific math areas.

In terms of EF, relative strengths were observed on a test of design fluency. However, similar to the findings of Korkman et al. (2003) and Rasmussen et al. (2012), our sample exhibited performance that was significantly below the normative mean on the Auditory Attention and Response Set tasks. Performance was particularly low on Response Set, which is more complex than the Auditory Attention task. Together, this suggests significant difficulty with selective and sustained attention and appropriate response timing and inhibition. These findings are unsurprising given the large body of literature documenting a multitude of EF deficits in children with FASD, including difficulties with cognitive flexibility, inhibition, reasoning, concept formation, attention, strategy use, cognitive planning, set-shifting and working memory (Rasmussen, 2005). All three EF subtests were strongly, positively correlated with total math achievement, although results did not reach statistical significance. Response Set was most highly correlated with math pre-test score, suggesting that difficulty with shifting and inhibition in addition to selective and sustained attention may impede math achievement.

The children in our sample also showed significant challenges with central executive tasks of working memory, which corresponds with previous research with children with PAE/FASD (Carmichael Olson et al., 1998). Performance on the test of phonological working memory, in contrast to previous research (e.g., Rasmussen & Bisanz, 2009), was relatively normal. Perhaps this is not surprising given the relative strengths in basic numeration exhibited by our sample, and significant deficits in arithmetic and problem-solving, which tend to be more related to the central executive. Similar to Rasmussen and Bisanz (2009), our sample demonstrated low-average visuospatial working memory ability that was not significantly different from the normative mean. Although our sample did not show significant impairment in all areas of working memory, working memory performance was strongly, positively related to math achievement, particularly the two measures of the central executive (which were statistically significant). The finding of a strong relationship between working memory and mathematical achievement corroborates research in typical and math disabled populations, as well as the findings of Rasmussen and Bisanz (2009), who documented a connection between working memory and math in children with FASD.

We observed relative strengths on the two measures of visuospatial processing, which was somewhat unexpected given previous research demonstrating visuospatial difficulties among children with PAE/FASD (Mattson et al., 2011). However, similar to what Pei and colleagues (2011) found, on the more complex visuospatial tasks that require the added demand of visuospatial memory and the integration of complex information (RCFT) children in our sample performed significantly lower than the normative mean, particularly on the delayed recall task which required a greater burden on long-term memory. It is possible that a failure to integrate information impedes storage and recall in long-term memory. However, none of the visuospatial memory tasks related to math achievement in our sample.

Intervention Math Outcomes

Following a relatively brief (10 half hour sessions over 6-8 weeks), individualized one-on-one intervention, children in the math group demonstrated an average improvement of 14.27 raw score points compared to children in the contrast group, who on average gained only 1.5 raw points. Effectively, after a two month period, children in the math group were able to demonstrate knowledge of close to 15 new basic math concepts. When the total math score was further broken down into composites, only the Basic Concepts math composite remained significantly different between groups. The Basic Concepts composite contains subtests relating to geometry, measurement, and basic estimation. Interestingly, the majority of the individualized mathematics interventions conducted focused strongly on building basic math skills including geometry, estimation, and measurement/quantity.

The math outcomes observed in this study suggest that a targeted psychoeducational program that focuses on the alcohol-related neurodevelopmental difficulties that are related to poor math learning/performance may help to ameliorate some of the deficits associated with prenatal alcohol exposure. Although the math program was conducted over a relatively short time period, we still observed significant gains in math achievement performance in our math group relative to our contrast group. In addition, the original MILE study (Kable et al., 2007) utilized parent training in addition to child tutoring, and tutoring took place in a laboratory setting. The total intervention equaled less than 20 hours of direct instruction for both children and caregivers combined (Coles et al., 2009). In the present study, we observed a significant gain in math score with an intervention that totaled to approximately five hours of direct child instruction. This suggests that parental support of mathematics learning may be an ideal—but perhaps not wholly necessary—factor in improving math achievement within the context of this intervention.

Cognitive Characteristics and Outcomes

The MILE program was designed not only to strengthen math skills in children with FASD, but to also accommodate deficits in underlying cognitions related to math, such as EF, working memory, and visuospatial functioning. Given the putative role of these processes in supporting math achievement, and the observed relationships between these cognitions and math in our sample, we felt it was reasonable to expect that if we saw changes in math achievement we may also see a transfer effect to other cognitions. Broadly speaking, children in both groups exhibited marginal gains on the cognitive measures, but the change scores were statistically similar for both the MILE and contrast groups. A trend toward greater group differences was observed on the tests of EF. Here, the math group exhibited gains of more than seven raw change points than the contrast group.

Based on the literature, it is not necessarily unusual for an intervention focused on achievement to not show generalized gains to other cognitive domains. For instance, Jansen et al. (2013) studied the effect of a computerized math training program in adolescents with mild to borderline intellectual disability. They found that although adolescents improved in math skill, there was no transfer effect to EF, working memory, or visuospatial processes. However, a recent study by Van der Molen and colleagues (2010) approached treatment of math difficulties inversely, and found that training visuospatial working memory led to improvements in mathematics in children with mild to borderline intellectual disability. This suggests that it may be fruitful to incorporate direct training of various cognitive skills to enhance generalization to math achievement.

It is important to note that although our contrast intervention, which targeted social skills, was not believed to have any overlap with mathematics, the ability to successfully deploy social skills does require the operation of cognitive abilities such as EFs (see Kully-Martens et al., 2013). Thus, it is possible that the MILE intervention did contribute to improvements in EFs, but perhaps not uniquely from another intervention.

Factors Related to Math Treatment Effect

Among the math group, those who made greater math treatment gains were older, with a lower IQ, from a lower SES home, and alcohol-exposed but undiagnosed ('PAE'). Conversely, those in the contrast group who made greatest changes in math performance tended to have higher IQ and an FASD diagnosis. Because these are within-group correlations with very small sample sizes (n = 11 and n = 6, respectively), these results should be interpreted as preliminary.

Coles et al. (2009) reported that those who made treatment gains were more likely to be younger and have greater alcohol-related dysmorphia (i.e., a more severe diagnosis on the fetal alcohol spectrum), and that IQ and SES did not have a significant impact on treatment outcome. However, both the present study and Coles and colleagues found that gender was not related to the treatment outcome. Assuming the present findings represent true relationships, the results may not be completely discordant with the previous MILE studies. For instance, removing the parent component may have 'evened the field' somewhat between children from low income and children from higher income backgrounds. Although our PAE participants were more likely to exhibit a greater treatment effect, participants with lower IQ were also more likely to exhibit significant gains.

The differences found between these two studies could also be due to different sample sizes (roughly 60 in the original MILE study versus 17 in the present study) and different sample compositions. For example, Kable et al. (2007) and Coles et al. (2009) utilized a lower-income, younger, largely African American sample of predominately clinically-referred children diagnosed with the most severe forms of FASD (i.e., FAS, pFAS). In contrast, the present study utilized a more moderate-income, older, predominately Caucasian and Aboriginal sample with a wider spectrum of clinical diagnoses. Additionally, different methodologies were used to determine SES and IQ in the present study and in Kable et al. (2007). Specifically, we used the Hollingshead scales, whereas SES appeared to be estimated based off of household income estimates in the original studies. In addition, we used the WRIT to estimate IQ, whereas the original studies used the Differential Ability Scales (DAS).

Coles et al. (2009) stated that it was logical to assume that children who were younger and more severely affected by alcohol may benefit more from the MILE intervention because they perhaps were less likely to have experienced school failure and academic disengagement. However, the opposite corollary could be proposed in our study. Perhaps older children have greater readiness to learn and thus a greater 'base' upon which to build math knowledge than younger children. Older children in this group could also have better-developed receptive language abilities, which would be important in a highly verbal program with high demands for one-on-one instructor-child interaction such as MILE. The older children in our study did *not* have lower IQ, lower math scores, or lower SES to begin with, suggesting this effect of age is distinct.

Limitations and Future Directions

Although our results are promising, several questions remain unanswered, and several limitations exist pertaining to both the study design and program implementation. First, we do not know the long-term implications of this intervention (although we are in the process of conducting a six-month follow-up). It is possible that there may be delayed treatment effects that we could not detect in such a short time period. Conversely, it is equally possible that the treatment effect observed will not be maintained into the future. Our intervention was also quite brief in duration (6-8 weeks). Future investigations may want to manipulate the length of treatment to ascertain the treatment duration that is most associated with both short and long-term gains.

We attempted to enhance the generalizability of this program to a greater variety of alcohol-affected children by including all children on the fetal alcohol spectrum, as well as those who were undiagnosed (recall, in Kable et al. 2007 only children with FAS and pFAS were used). The MILE program was developed specifically to compensate for alcohol-related difficulties. We still do not know the specificity of the intervention to alcohol-related neurodevelopmental challenges. However, we can speculate that because we observed children with PAE in the math group (who presumably have less neurodevelopmental impairment) improve more than children with FASD that perhaps the methods used in MILE are not entirely specific to the severely alcohol-affected population. To better understand the specificity of the intervention, trials with children with similar neurocognitive issues (e.g., ADHD) or other learning difficulties (e.g., math disability) should be explored. Similarly, although the MILE program was designed to use specific strategies and supportive educational tools to help support alcohol-related neurodevelopmental difficulties, our study could not determine if these strategies were necessary to the treatment effect or if the model was valid. Future studies may want to compare MILE to other one-on-one, individualized math interventions to determine which aspects are integral to the treatment effect. Although our results suggest that one-on-one interaction with an adult is not solely responsible for increasing math performance, it is possible that one-on-one *math* tutoring alone may have been all that was needed to effect change.

There are also several limitations inherent to our sample. One limitation is that our sample is, to some degree, self-selected. The majority of our sample was recruited from a diagnostic clinic; thus, we cannot be sure we would observe similar results in a community-based sample. In addition, there are issues with inclusion of a PAE group. Children exposed to alcohol prenatally are referred to as PAE when they do not have an FASD diagnosis. This can be for several reasons, including having a diagnosis deferred until a child is old enough have all neurobehavioral domains adequately assessed. Therefore, it is possible that children with PAE matched with other PAE participants may actually be diagnosed with FASD in the future. It is also difficult to control for individual participant factors that may contribute to potential treatment effects, such as qualities related to an individual participant's teachers, classroom, programming, school, and family, or unique subjective life history factors.

Statistical limitations also exist. As this is a pilot study, our sample size was very small. This restricted our power to detect group differences. Splitting our sample to correlate math score with demographic factors, in particular, created very small groups. Thus, these correlations should be taken as suggestive, although several strong relationships were observed even in our small sample. Due to these design limitations, we still are not certain which participant characteristics may be most integral to a math treatment effect. Going forward, further expanding our sample size to be more comparable with that of the original MILE studies will help to discern if the relationships found between variables in our study are truly different from those found in the original MILE study. Decreased power to detect group differences was also compounded by calculating change scores instead of using a repeated-measure design. However, because our groups were very different at pre-test (despite matching), a repeated-measure design would likely have cloaked any treatment effects. Because this is an exploratory pilot study, we did not adjust alpha for multiple comparisons. Thus, there is a chance that some of our significant findings are due to chance instead of true relationships.

There are also issues related to testing and measurement. We used the KeyMath for all our participants, even for those whose current age fell below the recommended age for use. During individual testing sessions it generally did not appear to be an issue; however, it could be argued that this test had an insufficient basal level to truly be sensitive to changes in lower level math skills, particularly in younger children. Thus, correlations of math achievement with age could be overestimated. Although we were able to provide an alternate parallel form of the KeyMath to participants at post-test, we did not have different forms of the measures we used to assess EF, working memory, and visuospatial functioning. It is possible that the lack of group differences observed could be due to a practice effect in both groups on these measures. Importantly, our groups exhibited significantly different math scores at pre-test, with our MILE group scoring lower than the contrast group. This means that regression to the mean was more likely to occur in our math group, implying that some of the treatment effect observed could be accounted for by having lower scores to begin with.

There are also several limitations and future directions related to program implementation. One major task of this study was to implement MILE into a school-setting. It can be difficult to apply one-on-one individual instruction in most standard school environments. Because of this, many interventions, for reasons of cost, space, and time, are carried out in a group fashion. In future, we hope to investigate the best way that MILE can be implemented into classrooms, perhaps using a train-the-trainer model with educational assistants or classroom-wide intervention.

Implications

Although the long-term impact of this relatively brief psychoeducational intervention is unknown, our results do suggest that using individualized and targeted teaching methods can foster learning in alcohol-affected children. The results of this study coupled with findings from other intervention studies conducted with this population should help to dispel the belief that individuals with alcohol-related brain damage are resistant to habilitation. In turn, this should generate further interest and study in designing and implementing models and methods of service delivery for individuals with PAE and FASD. Although one-on-one instruction with children does require time, we were able to effect changes in math achievement with an intervention that totaled to only five hours of direct instruction with a child and no significant contact with a teacher or parent. This sort of investment should certainly be feasible to conduct in both clinical and educational settings.

The benefit of learning math concepts is not limited to the traditional math curriculum or the school setting. Over the course of development, math constructs such as sorting, classifying, patterning, adding, and subtracting become integrated into our everyday adaptive life skills and problem-solving behaviors, allowing us to move about successfully in the world, predicting the outcomes of our behavior, the outcomes of others' behavior, and the environmental events that influence us. For example, understanding concepts such as units of time (e.g., minute, hour) and the repetition of days in a calendar can permit a sense of predictability and focus in work, school, and personal lives. Poor understanding of the relationship between time, events, objects, and other people, including event sequencing which is necessary for appropriate causal attribution and accepting time frames and delays in reference to activities and preferred items can contribute to frustration and lead to behavioral problems commonly observed in alcohol-affected individuals (Kable et al., 2007). Accordingly, a mathematic psychoeducational intervention may not just enhance the academic skills that lead to improved educational and occupational outcomes, but enhanced problem-solving skills that will impact everyday functioning.

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