

Short- and Long-Term Impacts of A Working Memory Intervention for Children with
Prenatal Alcohol Exposure

by

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Abstract

This dissertation consists of two studies designed to: (1) determine the neurobehavioural profile reflecting the strengths and limitations of children with Parental Alcohol Exposure (PAE) and (2) evaluate the short- and long-term impacts of a 25-session Cogmed[©] intervention on improving working memory and other cognitive, learning, and behavioural functioning in children with PAE. The first study examined the neurobehavioural profile of children with PAE. Data was collected from 46 children between 4 and 13 years old who were into a PAE group if they had a confirmation of PAE or a comparison group if they did not have PAE and had a history of typical development. In a clinical setting, children with PAE demonstrated strengths on tasks which required working memory, auditory attention, and cognitive fluency; however, their caregivers had not observed such strengths in day-to-day interaction. The second study involved 38 participants who were in the first study and completed a 25-session Cogmed[©] intervention. Significant improvements on both working memory and attentional control for both the PAE and comparison groups were reported (short-term impact). Most of the participants in the PAE group (65%) improved in at least one area of working memory. The gains on some measures were retained at follow up (long-term impact). The preliminary finding suggested there are short-term gains from the Cogmed[©] intervention and some potential long-term treatment effects.

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Chapter 1: Introduction

Context of problem

Prenatal alcohol exposure (PAE) is preventable, yet it is one of the most common known causes of neurodevelopmental disabilities in North America (National Institute on Alcohol Abuse and Alcoholism, 2015). PAE can result in lifelong permanent damage to the brain and also a potential diagnosis of Fetal Alcohol Spectrum Disorder (FASD). According to new Canadian diagnostic guidelines, FASD is used as an umbrella term as well as a diagnostic term to describe the broad spectrum of functional deficits (e.g., physical, mental, behavioral, and learning disabilities) as a result of maternal alcohol consumption during pregnancy (Cook et al., 2016). Individuals with PAE who fail to meet the criteria for an FASD diagnosis may also be impacted by central nervous system (CNS) damage (Cook et al, 2016). As a result of the CNS damage, individuals with FASD are at risk for neurobehavioural impairments in areas including executive function (EF), intellectual ability, attention, processing speed, language, visuospatial abilities, academics, learning, and memory (Streissguth et al., 1997).

Among the identified neurobehavioural impairments, EF deficits, especially working memory, are recognized as the core impairments for individuals with PAE (Kodituwakku, 2009). Working memory is often found to be a predictor for academic achievement (Nadler & Archibald, 2014). Rasmussen and Bisanz (2011) reported that working memory is highly correlated with mathematics performance in typically developing children, so working memory is crucial for individuals to perform and meet demands in school. Unfortunately, individuals with PAE often experience working memory impairments, especially in verbal and central executive working memory, which may cause the math underachievement in school (Rasmussen & Bisanz, 2011). As a result of working memory and other EF deficits, individuals with PAE often

experience adverse outcomes (e.g., dropping out of school, unemployment, homelessness, and mental health problems) that develop over time due to poor adaptations (Streissguth et al., 1997). The impact of PAE is significant and costly, as the Canadian government spends an estimated \$25,000 per person annually in healthcare, education, social services, and criminal justice systems (Alberta Institute of Health Economics, 2011; Rasmussen, Andrew, Zwaigenbaum, & Tough, 2008).

With the high costs associated with PAE, Petrenko et al. (2013) provided suggestions to remove the barriers that contribute to adverse outcomes in individuals with FASD and to ease the financial stress to the multiple systems. The five major barriers that Petrenko et al. (2013) identified often occur in the diagnostic process (e.g., delayed diagnosis) and in the services and supports for this population (e.g., inability to qualify for services, lack of availability of services, and problems with implementing and maintaining services). Delayed diagnosis is often the first and the most problematic barrier for individuals with FASD possibly reflecting challenges in establishing the highly skill multidisciplinary teams recommended for this diagnosis. In addition, a lack of standard psychological assessment protocols for FASD also contributes to the issue of delayed diagnosis and makes the diagnostic process challenging (Petrenko et al., 2013). It is critical to increase the awareness and knowledge of FASD and to explore assessment protocols that can effectively screen and identify FASD (Petrenko et al., 2013). This paper aims to investigate a neurobehavioural profile of individuals with PAE that provides information about the unique characteristics of these individuals. Sequentially, this can facilitate both diagnostic and intervention planning processes.

Another barrier that contribute to adverse outcomes in individuals with FASD is the limited number of services and interventions available for individuals with PAE (Petrenko et al.,

2013), which increases the risk for developing adverse outcomes. It is crucial to search for an effective, evidence-based intervention for individuals with PAE. To date, only a few interventions exist for individuals with PAE that address the core EF deficits, in particular working memory (Reid et al., 2015). Among the EF interventions, Cogmed[®] intervention is the most researched for improving working memory capacity and attention for a variety of populations, including typically developing children and children with disabilities (e.g., ADHD and learning disabilities) (Pearson, 2014). Studies have shown that the Cogmed[®] intervention can improve working memory capacity (McNab et al., 2009; Roughan & Hadwin, 2011), other EF measures (e.g., attention, inhibition, and non-verbal reasoning) (Klingberg et al., 2005; Klingberg, Forssberg, & Westerberg, 2002), and behavioural problems (e.g., it can reduce inattention and hyperactivity/impulsivity symptoms). Despite promising results, the effect of Cogmed[®] intervention is unclear for children with PAE. This dissertation aims to investigate the impact of the Cogmed[®] intervention on the working memory and other EF for children with PAE.

Statement of purpose

This dissertation consists of three separate articles that review literature about PAE, understand the strengths and limitations of children with PAE, and evaluate the impact of the Cogmed[®] intervention. Chapter 2 reviews the literature to explore the recent understanding of FASD, including the diagnostic criteria, diagnostic outcomes, and impact of PAE. The chapter includes an overview of EF, including its definition and key processes, and the common EF deficits to individuals with PAE. Chapter 2 aims to provide a theoretical foundation for chapters 3 and 4. Chapter 3 explores the neurobehavioural profile of children with PAE. Since the existing neurobehavioral profile research often fails to identify the specific areas of strength for this

population, this chapter intends to close these knowledge gaps by providing an understanding of the relative strengths and limitations of children with PAE in a Canadian context. These findings should help service providers to better understand the unique characteristics of the local population, including the response pattern to different assessment tools. Moreover, the interventions can be tailored to build on the specific strengths and address the specific needs of this local population. Chapter 4 aims to investigate the short- and long-term impacts for individuals with PAE. First, chapter 4 evaluates the short-term impact of a 25-session Cogmed[©] intervention on improving working memory and other cognitive, learning, and behavioural functioning in children with PAE. Second, the chapter examines whether the gain from the intervention is sustained at follow up (long-term impact). With these findings, service providers will have another effective evidence-based intervention option for individuals with PAE if the Cogmed[©] intervention is able to effectively target the core EF deficits.

Chapter 2: Literature review

The following section will include (i) an overview of Fetal Alcohol Spectrum Disorder (FASD), including the FASD phenotype, diagnostic criteria, diagnostic outcomes, and the impact of prenatal alcohol exposure (PAE); (ii) an overview of executive function (EF), including a definition, components, biological underpinnings of EF; and (iii) EF, working memory, and attentional control for individuals with FASD.

Overview of FASD

FASD phenotypes. The adverse impacts of maternal alcoholism on children's development were first documented in 1957 in a thesis by Jacqueline Rouquette, a French graduate student (Sanders, 2009). In 1968, Dr. Paul Lemoine, a French pediatrician, observed and documented the three common impairments in 127 cases of children impacted by maternal alcoholism: (1) developmental delays, (2) physical abnormalities of the face, heart, and limbs, and (3) behavioural problems (Lemoine, 2003). Dr. Paul Lemoine used the term "alcoholic fetopathy" to describe this condition (Public Health Agency of Canada, 2011). In North America, Drs. Ken Jones and David Smith (1973) described similar adverse impacts among 11 children born to alcoholic mothers. Drs. Jones and Smith coined the term Fetal Alcohol Syndrome (FAS), which is interchangeable with "alcoholic fetopathy," to describe the three phenotypes resulting from PAE: (1) distinct facial features, (2) growth deficiency, and (3) central nervous system (CNS) damage (Jones & Smith, 1973).

Distinct facial features. The first visible phenotypes are the two distinct facial features of FAS: a short palpebral fissure and abnormalities in the premaxillary zone (e.g., flat midface, short nose, smooth philtrum, and thin vermilion upper lip) (Chudley et al., 2005; Streissguth, Barr, Kogan, & Bookstein, 1997). Other associated facial features include epicanthal folds, low

nasal bridge, minor ear anomalies, and micrognathia (Streissguth et al., 1997). It is important to note that the exposure to alcohol during days 19 to 21 of gestation often leads to the development of FAS facial features (Clarren, 1999). Having the FAS facial features does not indicate the degree of severity of FASD.

Growth deficiency. Another visible phenotype is growth deficiency in prenatal and/or postnatal height or weight, as observed in low birth weight for gestational age, decelerating weight gain over time not due to other identified causes, and/or a disproportionately low weight-to-height ratio (Chudley et al., 2005). The height and/or weight of individuals with a growth deficiency are typically below the 10th percentile (Chudley et al., 2005). The growth deficiency may continue into adolescence and adulthood (Streissguth, Aase, Clarren, Randels, LaDue, & Smith, 1991).

Central nervous system (CNS) structural and functional abnormalities. As a result of PAE, structural and functional brain damage may occur. Some types of structural brain damage include decreased cranial size at birth, structural brain abnormalities, and neurological hard or soft signs (e.g., poor fine motor skills and/or hand-eye coordination, and loss of hearing) (Chudley et al., 2005). Some of this structural brain damage may lead to functional impairments, including deficits in motor skills, executive functioning, impulse control, intellectual ability, language, memory, attention, processing speed, judgment, learning, affect regulation, adaptive behaviours, and social skills (Cook et al., 2016; Kodituwakku, 2009). These brain impairments often lead to secondary disabilities, such as dropping out of school, unemployment, mental health issues, trouble with the law, and inappropriate social behaviors (Streissguth et al., 1997). The CNS impairment is often less visible at an early age, but will become more visible and pronounced in later years (Rasmussen & Bisanz, 2009a). CNS structural and functional

abnormalities are common among and have a lifelong effect on individuals with FASD (Burd, 2016).

Diagnostic criteria. The 4-digit diagnostic coding system was developed by the Fetal Alcohol Syndrome Diagnostic and Prevention Network (FAS DPN) in Washington, DC, in 1997 to systematically describe the full spectrum of outcomes of PAE (Astley, 2013). This diagnostic system has four criteria: evidence of growth deficiency, FAS facial features, central nervous system (CNS) structural and functional abnormalities, and confirmation of PAE (Astley, 2013). All four categories are rated using a four-point Likert scale with 1 being a lack of FAS phenotype and 4 indicating full expression of FAS phenotypes (Astley, 2013). For example, a rating of 1 may indicate that the individual does not display any growth deficiency, FAS facial features, brain damage, or gestational PAE whereas a rating of 4 may indicate that the individual displays below average (less than the third percentile) height and weight, all three distinct facial features (short palpebral fissure, smooth philtrum, and thin vermilion upper lip), brain damage (structural, neurological, and/or functional impairments), or a high level of PAE with confirmation (a result of heavy alcohol consumption during pregnancy). A total of 102 possible codes is used to describe the outcome of PAE (Astley, 2011). The inter-rater and intra-rater reliability of the 4-digit code ranges from 94% to 100% (Astley, 2013). FAS DPN recommends that a diagnosis of FASD should be formulated by a multidisciplinary team, including physicians, psychologists, speech pathologists, occupational therapists, social workers, and other professionals or individuals (Astley, 2013).

In addition to the 4-digit code diagnostic system, Canadian clinicians also use the Canadian diagnostic guidelines, which were developed by the Public Health Agency of Canada (Chudley et al., 2005) and updated by the Canada Fetal Alcohol Spectrum Disorder Research

Network (CAN FASD) in 2016. These guidelines help clinicians with FASD assessment and diagnostic processes, as shown in Figure 2.1. The new guidelines categorize two of the three hallmark features —distinct facial features (e.g., short palpebral fissure, smooth philtrum, and thin vermilion upper lip) and CNS damage (e.g., structural, neurological, and/or functional impairments) —as the defining features of FASD (Cook et al., 2016). The Canadian guidelines removed the hallmark feature of growth deficiency as a diagnostic criterion because it is no longer viewed as a distinct feature of FASD (Cook et al., 2016). A recent study showed no evidence of a direct association between prenatal alcohol consumption and growth deficiency or preterm birth as maternal smoking mediated the relationship (O’Leary et al., 2009).

The updated guidelines provide special considerations for diagnosing FASD at different developmental stages (e.g., infants, young children, and adults) (Cook et al., 2016). Similarly to FAS DPN, CAN FASD strongly recommends that the diagnostic process should not be performed in isolation, and input from a multidisciplinary team is required. Although both the 4-digit code diagnostic system and Canadian guidelines are popular guidelines in Canada to systematically guide the diagnostic process, there is no recommendation on how to combine the two systems to assess and diagnose clients with FASD as both systems have advantages and disadvantages. Further research is needed to investigate both the convergent validity between these systems and the reliability in diagnosis in order to determine whether one system or the other is more accurate (Coles et al., 2016).

Diagnostic outcomes. When using the 4-digit code diagnostic system (Astley, 2013), the multidisciplinary team often assigns one of the four following diagnoses: 1) FAS with or without confirmation of PAE, 2) Partial Fetal Alcohol Syndrome (pFAS) with or without confirmation of PAE, 3) Alcohol-Related Neurodevelopmental Disorder (ARND), and 4) Alcohol-Related Birth

Defects (ARBD) (Alberta Medical Association, 2003; Stratton, Howe & Battaglia, 1996). These four diagnostic terms fall under the umbrella of FASD, which is used to describe the broad spectrum of disabilities that result from PAE. With the updated Canadian diagnostic guidelines, this umbrella term becomes a diagnostic term. The CAN FASD viewed it unnecessary to use subcategories within the diagnosis as it is unclear whether it is possible to accurately distinguish between subcategories of FASD (Coles et al., 2016; Cook et al., 2016).

Fetal alcohol syndrome (FAS) or FASD with sentinel facial features. The diagnosis of FAS using the 4-digit code diagnostic system can be given with or without confirmation of PAE. A diagnosis of FAS requires an expression of all three FASD phenotypes: evidence of growth deficiency, FAS facial features, and severe brain damage (Alberta Medical Association, 2003). Under the updated Canadian guidelines, a diagnosis of FAS according to the 4-digit code diagnostic system is equivalent to a diagnosis of FASD with sentinel facial features. Since growth deficiency has been removed from the Canadian guidelines, the criteria of a diagnosis of FASD with sentinel facial features include the presentation of the three FAS facial features and evidence of CNS damage in three or more areas (Cook et al., 2016). Only a small portion of individuals who are impacted by PAE display the full spectrum of FAS, and the majority of children do not have the classic facial features (Alberta Learning, 2004). Therefore, FASD is described as an “invisible disability.”

Partial fetal alcohol syndrome (pFAS). The diagnosis of pFAS in the 4-digit code diagnostic system can be made with or without confirmation of PAE. In order to diagnose an individual with pFAS, he/she has to display some evidence of FAS facial features and one of the following FASD characteristics: evidence of growth deficiency, severe brain damage, or behaviour and cognitive abnormalities that are related to brain damage (Alberta Medical

Association, 2003). With the updated Canadian guidelines, this diagnosis does not exist, as the subcategories of FASD have been removed (Coles et al., 2016; Cook et al., 2016).

Alcohol-related neurodevelopmental disorder (ARND) or FASD without sentinel facial features. The diagnosis of ARND in the 4-digit code diagnostic system results from confirmed PAE. An individual with a diagnosis of ARND does not show any evidence of FAS facial features but displays evidence of severe brain damage or exhibits behaviour and cognitive abnormalities that are related to brain damage (Alberta Medical Association, 2003). With the updated Canadian guidelines, a diagnosis of ARND is equivalent to a diagnosis of FASD without sentinel facial features. The criteria of FASD without sentinel facial features include a confirmation of PAE and the evidence of three or more areas of CNS damage (Cook et al., 2016).

Alcohol-related birth defects (ARBD). ARBD also requires confirmation of PAE; however, the individual with a diagnosis of ARBD does not show any evidence of FAS facial features. In order to be diagnosed with ARBD, the individual must have a vision and/or hearing impairment and display specific anomalies, including of the heart, kidneys, and/or bones (Alberta Medical Association, 2003). With the updated Canadian guidelines, this diagnosis does not exist as the growth deficiency criterion has been removed (Cook et al., 2016).

At-risk for neurodevelopmental disorder and FASD (At-Risk). With the updated Canadian guidelines, a new category, At-Risk for Neurodevelopmental disorder and FASD, was added to capture the individuals who did not meet the diagnostic criteria but were still at risk for FASD (Cook et al., 2016). This At-Risk category is not a diagnosis, but a method of identifying individuals who are at risk for FASD because they have a confirmation of PAE at a level known to be associated with CNS damage with or without the facial features (Cook et al., 2016).

Individuals who are assigned to this category have failed to meet the criteria for an FASD diagnosis because they are either too young, unable to complete the neurodevelopmental assessment; or display CNS damage in fewer than three areas (Cook et al., 2016).

Prevalence

All diagnoses of FASD only describe the CNS and physical impairments that are present; they do not imply severity (Clarke & Gibbard, 2003). For example, individuals with partial FAS, who do not display all three FASD phenotypes (e.g., lack of facial features), may not be necessarily less severe than the individuals with FAS because they may experience more severe CNS and/or growth deficits than the individuals with FAS. In Canada, the pooled prevalence of FAS and FASD is estimated to be one out of 1000 individuals and five out of 1000 individuals, respectively (Popova et al., 2017). The new Canadian diagnostic guidelines suggest that the prevalence of FASD has increased to approximately one in 100 individuals with about 330,000 individuals currently affected (Cook et al., 2016). Due to clinical challenges such as limited screening and diagnostic resources, lack of awareness, and missing documentation (e.g., confirmation of PAE) (Healthy Child Manitoba 2012), the prevalence of FASD in Canada may still be underestimated. In the United States (US), the prevalence of FAS and FASD is estimated to be two out of 1000 individuals and 15 out of 1000 individuals, respectively (Popova et al., 2017). In Midwestern US, the prevalence of FAS and pFAS in ranges from 0.6% to 0.9% and 1.1% to 1.7%, respectively (May et al., 2014). A total prevalence of FASD in Midwestern US is estimated to be 2.4% to 4.8% (May et al., 2014). Around the world, an estimated prevalence of both FAS and pFAS ranges from 0.0006% to 0.3% (Ospina & Dennett, 2013). The global prevalence of both ARBD and ARND is estimated to be 1.08% and 0.37% (Ospina & Dennett, 2013). The wide range of global prevalence rates may be due to the different diagnostic methods

used across the world (Ospina & Dennett, 2013). The majority of studies suggest that the prevalence of FAS and/or FASD is often underestimated worldwide due to a lack of awareness and/or misdiagnosis (Popova et al., 2017).

Impact of PAE

In general, individuals with any diagnosis of FASD may experience behavioural and learning difficulties in the classroom, including sensory and motor impairments, poor memory, delayed language development (e.g., difficulty in comprehension and communication), cognitive problems (e.g., low intelligence quotient (IQ), slow processing, and poor use of strategies), difficulties with behavioural regulation (e.g., poor emotional regulation and difficulty shifting attention), and poor adaptive skills (Alberta Learning, 2004; Streissguth *et al.*, 1997). These behavioural and learning difficulties often lead to secondary disabilities or emotional and societal problems (Streissguth et al., 1997). The combination of both primary and secondary disabilities often has a significant impact on the daily function of individuals with FASD (e.g., it limits careers and independent living) (Spohr, Willms, & Steinhausen, 2007). Early interventions which target the cognitive impairments common to FASD will reduce the chance of a child developing secondary disabilities.

Overview of executive function

EF deficits are considered the core impairment for individuals with PAE (Kodituwakku, 2009). This section will review the definition of EF and the impact of EF deficits on children with FASD.

Definition of EF. Historically, the understanding of EF originated with the observation of patients with frontal lobe injury. In 1848, the famous case of Phineas Gage, whose frontal lobe injury caused impairments in emotional regulation, memory, and planning, sparked interest in

examining the relationship between the frontal lobe and EF in the field of neuropsychology (Hunter & Sparrow, 2012). Since then, researchers have continued to examine the function of the frontal lobe in order to gain a better understanding of EF. Anderson, Northam, and Wrennall (2014) suggested that although prefrontal regions play an important role in coordinating a set of basic cognitive processes during complex problem-solving, the input and efficient functioning of other areas of the brain (e.g., subcortical and thalamic pathways) are equally important. The integration of the whole brain allows one to perform optimally on executive tasks (Anderson et al., 2014).

In past decades, many different models and theories have been developed to explain the concepts and components of EF. Because of the wide range of EF abilities observed in the field and the lack of a unified definition, Zelazo, Müller, Frye, and Marcovitch (2003) reviewed three approaches categorizing the current EF models and theories in their review paper. The first model is theoretically constructed based on different EF abilities (Zelazo et al, 2003). For example, EF is considered a higher level cognitive mechanism involving different abilities such as inhibition, working memory, and planning (Miller, 2009; Zelazo et al, 2003). The second model involves conducting factor analytic studies of neuropsychological batteries that measure the functional components of EF in an effort to discover the essential components (McCabe et al., 2010). This method usually yields three to four separate components of EF; however, the factor analyses may also reflect non-EF components (van der Sluis, de Jong, & van der Leij, 2007). The third model conceptualized EF based on the multiple narrow abilities that are required for specific tasks. Unlike the first two approaches, this perspective allows us to understand how different aspects of EF work concurrently instead of viewing EF as a unified construct with multiple components. For example, Miyake et al. (2000) demonstrated both the

diversity (e.g., three distinct factors of inhibition, updating, and shifting) and unity (e.g., the aspects of the distinct factors are related) of EF in their model. This is currently a more popular way to understand EF. Although the understanding of EF is a complex concept and varies across different models (Jurado & Rosselli, 2007; McCloskey, Perkins, & Van Diviner, 2009), most psychologists agree that EF is an umbrella term to describe the top-down neurocognitive processes that are involved in integration of conscious control of thoughts, actions, and emotions to carry out goal-driven behaviours (Miyake et al., 2000; Zelazo & Muller, 2010).

Recently researchers have suggested that EF can be further differentiated into two categories: cool EF (the cognitive aspect of EF) and hot EF (the affective aspect of EF) (Zelazo & Muller, 2010). Cool EF is often considered to be the traditional view of EF, related to cognitive problem-solving. The multi-dimensional construct of cool EF includes cognitive flexibility, inhibition, working memory, planning, organization, and attentional control, which are necessary for adapting to environmental changes and demands (Barkley, 2012; Miyake & Friedman, 2000). Hot EF is a new addition to the perspectives on EF, and relates to affective problem-solving, including the emotional and motivational processes involved in cognitive control (Zelazo, Qu, & Kesek, 2010). This aspect of EF is essential to learning social rules and acting in a socially appropriate manner (Zelazo et al., 2010). Although cool and hot EFs have distinctive characteristics, they also work closely together (Hongwanishkul, Happaney, Lee, & Zelazo, 2005). Deficits in either cool or hot EF are often associated with negative academic outcomes, poor social competency, and high risk for serious maladjustment (e.g., substance abuse and aggression) (Zelazo & Müller, 2010). Based on the development of EF theory, most research and clinical tools (e.g., neuropsychology tests) have historically focused on cool EF (Kodituwakku, 2009). Despite the importance of hot EF, this study is also tailored to understand

the cool EF of children with FASD. The following sections will refer to cool EF as EF and introduce the components, biological underpinnings, and developmental stages of cool EF.

Key EF processes. In order to gain a better understanding of EF, it is important to review the three major EF processes associated with students' academic learning: basic components of EF, working memory, and attentional control (Purdy, 2011).

Basic components of EF. In order to identify the essential and most basic components of EF, the confirmatory factor analysis approach, which compares different EF tasks from the neuropsychological battery, is often used (Zelazo et al., 2003). Miyake et al. (2000) indicated that the three essential separable but interrelated constructs of EF are inhibition, updating, and shifting. Inhibition allows individuals to inhibit dominant, automatic, or prepotent responses. Some classic inhibition tasks are the stop-signal task (Logan, 1994), which requires participants to withhold a response, and the Stroop task (Stroop, 1935), which requires participants to inhibit the dominant response and generate a subdominant response (Friedman et al., 2008). Updating refers to the constant monitoring and updating of information in the working memory either by adding new relevant information or removing irrelevant information. One updating task is the Keep Track Task (Yntema, 1963), in which participants see a series of 15 words from six categories (e.g., animals and countries) on the computer screen and are required to recall the words in categories after the words disappear (Friedman et al., 2008). Shifting allows individuals to switch between mental tasks. One shifting task is the Number-Letter task, in which the participants are asked to remember a number-letter or letter-number pair (e.g., 7G) and are required to switch between two categories in each pair after the stimulus disappears (Friedman et al., 2008). In Miyake et al.'s (2000) model, these three constructs represent distinct abilities but they are also correlated with each other. For example, switching between tasks requires both the

suppression of previous tasks (inhibition) and the cognitive demands to constantly maintain and update mental sets based on feedback (updating). This means individuals have to master the first two components of EF in order to achieve shifting.

Recently, Fisk and Sharp (2004) followed up on Miyake et al.'s work by conducting another factor analysis on executive function measures. In addition to the three components in Miyake and Friedman's model, they identified an additional fourth factor: verbal fluency, which is responsible for the efficiency of lexical access to long-term memory (Fisk & Sharp, 2004, Jurado & Rosselli, 2007). Fisk and Sharp (2004) explained that the three components in Miyake and Friedman's model were derived from analyzing lower level tasks whereas the additional component resulted from analyzing higher level executive tasks.

In addition to the models proposed by Miyake et al. (2000) and Fisk and Sharp (2004), Anderson, Anderson, Northam, Jacobs, and Catroppa (2001) reported that attentional control and planning are also essential components of EF. Attentional control includes selective (or shifting) attention, sustained attention, and response inhibition (Anderson et al, 2001). This component of EF overlaps with Miyake and Friedman's inhibition component and is required for shifting and switching. Planning means to identify materials and steps that are needed to achieve a goal (Anderson et al., 2001). This component requires complex cognitive demands, which involve the three basic components from Miyake and Friedman's model. Recent research supports the theory of essential basic EF components (inhibition, shifting, switching) (Friedman et al., 2008) and some essential advanced EF components (verbal fluency, attentional control, planning) (Anderson et al, 2001; Fisk & Sharp, 2004; Jurado & Rosselli, 2007).

Working memory. Of all the EF processes, working memory is the most researched. Baddeley's working memory model is the most prominent framework used in the field of

psychology (Baddeley, 2003; Miyake et al., 2000). Working memory is described as a cognitive system that is responsible for the short-term storage and manipulation of information (Baddeley, 2003). Working memory serves several functions: 1) encoding incoming information, 2) retaining information, 3) retrieving information from the long-term memory, and 4) processing information (Dehn, 2011). In Baddeley's (2003) model, working memory consists of four components: one central executive system and three slave systems.

The first slave system is the visuospatial sketchpad, which is responsible for helping individuals to remember and manipulate visual and spatial information (e.g., remembering numbers, shapes, or other non-verbal stimulus) (Baddeley, 2003). There are two components within this slave system: a passive temporary storage and an active rehearsal process (Dehn, 2011). Although the visuospatial sketchpad is designed to process visual and spatial information, it is also an important component of reading because it assists with encoding printed letters and words and keeping a visuospatial frame of reference.

The second slave system is the phonological loop, which is responsible for helping individuals to remember and manipulate language-based and phonological information (Baddeley, 2003). Similar to the visuospatial sketchpad, the phonological loop consists of two functions: passively storing and actively rehearsing and processing the phonological information (Dehn, 2011).

The third slave system is the episodic buffer, which was added to the working memory model in 2000. The addition was made to explain the role of active long-term memory representation with limited capacity (Baddeley, 2003). The episodic buffer accesses and searches the long-term memory for representations to facilitate the understanding of the new information (Baddeley, 2003). The episodic buffer can process both visual and verbal information as well as

other information, such as episodic and semantic memory (Dehn, 2011). Initially, the episodic buffer was considered part of the central executive. However, given the importance of the role, Baddeley (2003) later designated it as another slave system.

The central executive is responsible for coordinating the three slave systems, and for controlling and regulating cognitive processes such as planning and organizing information (Baddeley, 2003). The central executive is viewed as the foundation for understanding EF processes (Zelazo & Müller, 2010). The central executive is needed for individuals to perform any basic and advanced EF tasks (Baddeley, 2003; Miyake et al., 2000).

Attentional Control. Another major EF process that is associated with learning is attentional control. Among the competing models and theories, Norman and Shallice's (1986) supervisory attentional system model and Mirsky, Pascualvaca, Duncan and French's (1999) model of attention are the more popular explanations of the attentional control processes.

The Supervisory Attentional System (SAS) model explains the executive attentional control of information processing (Norman & Shallice, 1986). In this model, two types of information processing, contention scheduling and SAS, are described. Contention scheduling is the lower level control mechanism. It is responsible for controlling any routine habits or specific overlearned actions (Norman & Shallice, 1986). This component first receives input from the sensory perceptual systems, which are triggered by physical stimuli in the environment. Then it selects the most appropriate automatic or routine action and inhibits other competing actions (Hunter & Sparrow, 2012). In contrast, SAS is responsible for intentionally overseeing any non-routine or novel action. SAS alters or inhibits the initial action proposed by contention scheduling when encountering novel situations, which allows individuals time to process the situation (Hunter & Sparrow, 2012). SAS is considered as the executive attentional control,

which is crucial when generating new schemas, implementing these schemas, and evaluating the effect of an action.

Mirsky et al. (1999) used a neuropsychological approach to explore different dimensions of attention, and further investigated and compared the pattern of these dimensions in the individuals in their control and experimental groups (e.g., Attention Deficit Hyperactivity Disorder (ADHD)). In this model, they proposed five dimensions of attention: one controlled EF component (focus-execute attention), three attention components (sustain, shift, and encode), and one consistency component (stability) (Mirsky et al., 1999). Focus-execute attention allows individuals to selectively allocate attentional resources to focus on the target task (Mirsky et al., 1999), which is similar to SAS in Norman and Shallice's model.

Sustaining attention is responsible for helping individuals to remaining on task in a vigilant manner (vigilance) (Mirsky et al., 1999). Shifting attention is responsible for helping individuals to shift attentional focus from one task to another task in a flexible and adaptive manner (Zillmer et al., 2008). Encoding attention is responsible for helping individuals to hold information briefly in his/her memory while performing other cognitive actions (Mirsky et al., 1999). This dimension of attention seems to be interrelated with working memory. Stability is responsible for helping individuals to maintain a regular and predictive response over time (Mirsky et al., 1999). Stability is also known as the consistency of attentional effort, which is an important foundation for the first four dimensions (Zillmer et al., 2008).

All essential EF components, working memory, and attentional control often intertwine and work closely together to allow students to learn and function in school.

Biological underpinnings of EF. Both experimental and clinical research indicates that the prefrontal cortex is predominantly responsible for EF. In the 1900s, the terms “frontal lobe

syndrome” (Luria, 1969) and “dysexecutive syndrome” (Baddeley, 1986) were used to describe the deficits associated with damage to the frontal lobe, including problems with goal formulation, planning, organizing, and inhibiting behaviours.

The three primary systems in the frontal lobe that are associated with EF are the dorsolateral prefrontal circuit (DLPC), anterior cingulate circuit (ACC), and orbitofrontal circuit (OFC) (McCloskey et al., 2009; Tekin & Cummings, 2002). DLPC (Brodmann’s areas 9 and 10) is closely connected to the classic characteristics of EF, including anticipation, goal selection, planning and organization, mental flexibility, working memory, attention, self-monitoring and self-regulation (Bonelli & Cummings, 2007). This circuit was found to be predominantly associated with cool EF (cognitive aspect). The ACC (Brodmann’s areas 24) is associated with initiation, inhibition, and motivation of behaviours (Bonelli & Cummings, 2007). The OFC (Brodmann’s areas 10 and 11) is involved in behavioural and emotional regulation, including interpersonal sensitivity, and determines the social appropriateness of behaviour (Bonelli & Cummings, 2007). This circuit was found to be predominantly associated with cool EF.

EF at different developmental stages. The development of EF often aligns with the maturation of the frontal lobe. EF development starts during infancy and continues through late adolescence to early adulthood (Anderson, 2002). Throughout development, there are three growth spurts: i) infancy to early childhood (birth to age six), ii) middle childhood (ages seven to 10), and iii) adolescence (ages 12 to 19) (Anderson, 2002; Welsh, Pennington, & Groisser, 1991). During infancy, babies start to develop basic EF processes, such as simple memory and attentional control (Anderson, 2002; McCloskey, Perkins, & Van Diviner, 2009). As children continue to grow throughout early childhood, they start to be able to resist distraction (Welsh et al, 1991), have better attention control, and perform goal-directed behaviours, including basic

inhibition, planning, and working memory (Anderson, 2002; McCloskey et al, 2009). Later, around seven to 10 years of age, EF starts to develop more rapidly, and set-shifting, goal-setting, and information-processing start to emerge (Anderson, 2002; Welsh et al, 1991). When children reach 12 years of age many EF domains such as verbal fluency, motor sequencing, complex planning skills (Welsh et al, 1991), set-shifting, response-inhibition, and concept-formation (Anderson, 2002) begin to mature. As the prefrontal cortex starts to reach maturation during adolescence and early adulthood, individuals are able to coordinate different EFs to solve a complex task. EF development often begins with skills needed to respond to the external environment (e.g., basic inhibition) and, as children grow, shifts to skills required for internal processing (e.g., verbal fluency).

EF, working memory, and attentional control and FASD. EF, working memory, and attentional control are core deficits in individuals with FASD (Mattson, Crocker & Nguyen, 2011; Mattson et al, 2010; Rasmussen, 2005). Children with PAE display difficulties in cognitive flexibility, response inhibition, planning, concept formation, set-shifting, verbal reasoning, and verbal and nonverbal fluency (Kodituwakku, 2009; Mattson et al, 2011, Mattson et al., 1999). In addition, verbal and nonverbal communication and processing speed deficits are common in individuals with FASD (Vaurio, Riley, & Mattson, 2008). Studies show that some EF tasks that individuals with FASD find challenging are Stroop, the Wisconsin Card Sorting Test, inhibition, and the Tower of London and design fluency subtests (Rasmussen, 2005).

Working memory is significantly impaired in children with FASD regardless of their IQ (Burden, Jacobson, Sokol, & Jacobson, 2005). Although research has been done on verbal memory (e.g., encoding and retrieving information) and spatial memory, which were found to be impaired in individuals with FASD, there has been less research focus on working memory

(Manji, Pei, Loomes, & Rasmussen, 2009). Nonetheless, at this time verbal working memory and functions associated with the central executive are thought to be more impaired in individuals with FASD (Rasmussen, 2005).

A common characteristic of individuals with FASD is a deficit in attentional control, including all four types of attention from the Mirsky model: focus-execute attention, sustaining attention, shifting attention, and encoding attention (Burden, Jacobson, Sokol, & Jacobson, 2005). In comparison to individuals with ADHD, individuals with FASD experience more difficulties in the shifting and encode attention domains (Coles et al., 1997; Mattson et al., 2011). Shifting attention is related to EF whereas encoding attention is related to working memory (Burden et al., 2005). Working memory is the most important mediator of attention deficits in children with FASD (Burden et al., 2005).

Summary

FASD is a lifelong condition that resulting from PAE, and it can be characterized by distinct facial features, growth deficiency, and/or CNS structural and functional abnormalities (Jones & Smith, 1973). Individuals with FASD have some common core deficits in areas including EF, working memory, and attentional control. These core cognitive deficits are typically associated with learning problems in school, which in turn can lead to further adverse outcomes such as dropping out from school, unemployment, homelessness, and mental health concerns (Zelazo & Müller, 2010). In order to effectively support individuals with FASD, it is important to examine effective assessment or screening methods to promote early recognition and accurate diagnosis as well as to search for evidence-based interventions that address the core cognitive deficits. With these goals in mind, the following two chapters aim to 1) explore the neurobehavioural profile of individuals with PAE in order to facilitate assessment, screening, and

intervention planning process (Chapter 3) and 2) evaluate an evidence-based working memory intervention in order to provide an effective intervention for this population (Chapter 4).

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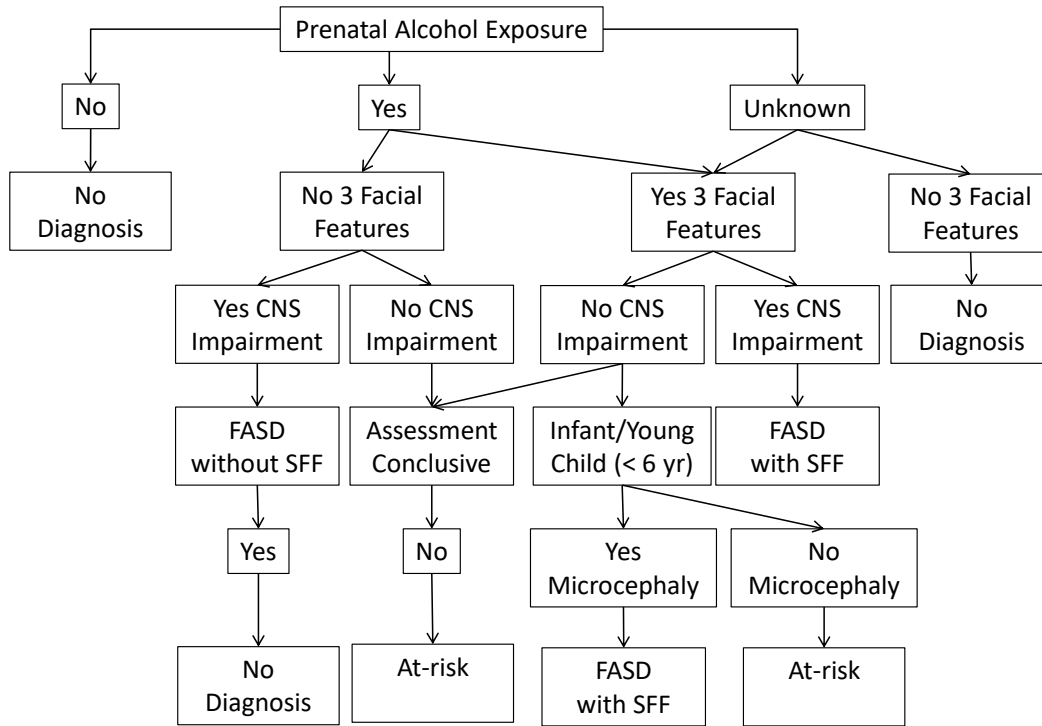


Figure 2.1 Diagnostic algorithm for FASD (Cook et al., 2016)

Chapter 3: Neurobehavioral profile of children with PAE (Study 1)

Prenatal alcohol exposure (PAE) can have significant negative impacts on fetal development, including permanent brain damage (Chudley et al., 2005). Individuals with PAE are at risk for a diagnosis of Fetal Alcohol Spectrum Disorder (FASD), which is characterized by two key criteria: central nervous system (CNS) damage (e.g., structural, neurological, and/or functional brain damage) and distinct facial features (e.g., short palpebral fissure, smooth philtrum, and thin vermilion upper lip) (Cook et al., 2016). As a result of the CNS damage, individuals with PAE often experience executive functioning (EF) deficits that interfere with their daily activities. The most prominent of these are the inability to control impulse behaviour, understand consequences, and make fair judgements (Streissguth et al., 1997). Previous studies identified a spectrum of EF deficits that are common in individuals with PAE, including planning (Kodituwakku, 2009); response inhibition (Mattson et al, 2011; Rasmussen & Bisanz, 2009a); working memory (Burden et al., 2005; Rasmussen, 2005); conceptual and affective set-shifting (Kodituwakku, 2009; Mattson et al, 2011); verbal and nonverbal fluency (Kodituwakku, 2009; Mattson et al, 2011); attention (Mattson et al, 2011); academic underachievement in reading and mathematics (Howell et al, 2005; Rasmussen & Bisanz, 2009a); externalizing problems such as aggression, inattention, and conduct problems (Mattson & Riley, 2000; Nash et al., 2006); and internalizing problems such as depressive symptoms (Mattson & Riley, 2000).

Neurobehavioral profile

This section includes a review of the core EF deficits identified by the previous neurobehavioural profile studies, including inhibitory control, working memory, cognitive flexibility, attentional control, mathematics underachievement, and emotional and behavioural functioning issues.

Inhibitory control. Based on the EF hierarchy model (2000) developed by Miyake et al., inhibition is the foundational EF component, which allows individuals to inhibit dominant, automatic, or prepotent responses (Miyake et al., 2000). Individuals with PAE often experience problems with and/or require more cognitive effort to inhibit their inappropriate habitual responses (Burden et al., 2009; Kodituwakku, Kalberg & May, 2001). As the result of poor impulse control, students with PAE may experience difficulty foreseeing the consequences of their actions and delaying gratification (Schonfeld, Paley, Frankel, & O'Connor, 2006), which puts them at higher risk for experiencing difficulty in social situations (Schonfeld et al., 2006) and engaging in high-risk activities (e.g. delinquent behaviours) (Fast & Conry, 2009).

Working memory. Working memory is the second component of EF (Miyake et al., 2000) and is responsible for short-term storage and manipulation of information (Baddeley, 2003). In Baddeley's (2003) traditional model, working memory can be broken into three components: two slave systems and one central executive system. The first slave system is the phonological loop (verbal short-term memory), which is responsible for temporarily storing and actively rehearsing language-based and phonological information (Baddeley, 2003; Dehn, 2011). The second slave system is the visuospatial sketchpad (visuospatial short-term memory), which is responsible for temporarily storing and actively rehearsing visual and spatial information (Baddeley, 2003; Dehn, 2011). The visuospatial sketchpad is also involved in reading as it assists with encoding printed letters and words and keeping a visuospatial frame of reference (Dehn, 2011). The central executive is responsible for coordinating the slave systems and controlling and regulating cognitive processes, such as planning and organization of both verbal and visuospatial information (verbal and visuospatial working memory) (Baddeley, 2003).

Regardless of intelligence quotient (IQ), working memory is described as a common

impairment in children with FASD (Burden, Jacobson, Sokol, & Jacobson, 2005). Verbal memory (e.g., encoding and retrieving information) and spatial memory are found to be impaired in individuals with PAE (Manji, Pei, Loomes, & Rasmussen, 2009). Specifically, verbal working memory and functions associated with the central executive are thought to be more impaired in individuals with PAE (Rasmussen, 2005). Students with working memory deficits may struggle to learn from their experience and generalize their experience to a new situation, which can lead to adverse outcomes (Rasmussen & Bisanz, 2011; Schonfeld et al., 2006).

Cognitive flexibility. Cognitive flexibility is the last and most complex EF component. It refers to an individual's ability to switch his/her cognitive effort between tasks (Miyake et al., 2000). Cognitive flexibility requires access to both inhibitory control and working memory, as the individual uses both to suppress the tasks and constantly maintain and update mental sets based on feedback (Miyake et al., 2000). Cognitive flexibility impairments, which impact an individual's deductive reasoning and verbal abstract thinking, are common in individuals with PAE (Rasmussen & Bisanz, 2009a).

Attentional control. Attentional control often intertwines with both working memory and cognitive flexibility. The four types of attention from Mirsky's model are focus attention, sustaining attention, shifting attention, and encoding attention (Burden, Jacobson, Sokol, & Jacobson, 2005). Attentional control deficits are common among individuals with FASD, specifically in the shifting and encoding attention domains (Coles et al., 1997; Mattson et al., 2011). Shifting attention is related to cognitive flexibility whereas encoding attention is related to working memory (Burden et al., 2005). In addition, individuals with PAE also display deficits in sustained attention, which is not an EF component even though it is a less prominent issue when compared to shifting attention. Students with attention deficits may struggle to learn in the

classroom and have a lower academic performance (Barriga et al, 2002).

Mathematics achievement. Individuals with PAE often experience difficulty in mathematics achievement, especially in arithmetic (Rasmussen & Bisanz, 2009b). Rasmussen & Bisanz (2011) found that mathematics performance is closely linked to working memory, so individuals with PAE often have difficulty in mathematics due to underlying working memory deficits.

Emotional and behavioral functioning. Emotional and behavioral problems are often common for individuals with PAE throughout childhood and adolescence (Pei, Denys, Hughes, & Rasmussen, 2011). Students with PAE are at a higher risk of experiencing the symptoms of internalizing disorders (e.g., anxiety and mood disorders), externalizing disorders (e.g., ADHD and conduct disorders), suicide, and alcohol and/or drug use (Pei et al., 2011). These mental health problems may not be noticeable at birth but often become more pronounced as the students mature due to the interaction among genetic, neuropsychological (e.g., EF deficits) and environmental factors (Pei et al., 2011). Moreover, it is typical for these mental health problems to persist into adulthood.

Statement of problems

The EF deficits identified above often have a ripple effect and lead to adverse outcomes for individuals with PAE, such as dropping out of school, alcohol and drug addictions, and inappropriate social behavior (Streissguth et al., 1997). Because supporting individuals with FASD is costly, researchers have focused their attention on ways to mitigate these costs, and ultimately support improved outcomes for these individuals. Researchers have explored neurobehavioral profiles to understand the specific areas of weaknesses in individuals with PAE in order to facilitate the diagnostic process and identify priorities for intervention. Through these

efforts, some key cognitive functioning deficits such as language, visual perception, memory, learning, inhibition, social functioning, and attention have been identified (Kodituwakku, 2009; Mattson et al., 2013; Rasmussen, Horne, & Witol, 2006; Rasmussen et al., 2013). Yet, there are two major clinical and research gaps in the existing neurobehavioral profile studies. First, different FASD diagnostic methods used across jurisdictions have identified regional variations as well as different cultural and socioeconomic status (SES) compositions (Petrenko & Davis, 2017). A local neurobehavioral profile study is critical to help identify the unique needs for the local population. This Canadian study aims to replicate the previous findings to Alberta, Canada to provide local data to help with planning services and intervention for the local population.

Second, numerous neurobehavioural profile studies often focus on the deficits of individuals with PAE and fail to acknowledge their relative strengths. To date, there are only a few studies suggesting the relative strengths of affected individuals (McLachlan et al., 2017; Rasmussen et al., 2006; Rasmussen et al., 2013). Recent studies have identified multiple benefits associated with using the strength-based model when working with individuals with disabilities. One strength-based model, “neurodiversity,” has been introduced in the field of Autism Spectrum Disorder (ASD) research, and it describes atypical neurodevelopment as part of a human variation rather than viewing it as a disability (Smith, 2006). The goal is to view children with disabilities as having a different way of thinking and learning instead of pathologizing children (Armstrong, 2012). In one study, researchers found that university students with learning disabilities (LD) who view their difference using the neurodiversity model report a different set of strengths and weaknesses and a higher level of academic self-esteem and career ambition when compared to students with LD who view their difference from a medical model (Griffin & Pollak, 2009). Moreover, a strength-based exercise program in school curriculums was found to

improve life satisfaction and well-being among typical developing students (Proctor et al., 2011). The strength-based model also consistently provides positive outcomes and leads to greater success in different settings such as employee engagement, school achievement, attendance, productivity, and hope (Clifton & Harter, 2003). Since numerous researchers have documented benefits with the strength-based model, it is beneficial to understand the neurobehavioral profile from a strength-based model to capture the strengths of children with PAE while remediating their weaknesses. Therefore, this study aims to provide a balanced neurobehavioural profile indicating both relative strengths and limitations in order to provide insight for strength-based intervention planning.

Besides bridging the clinical and research gaps, this paper also aims to strengthen the current understanding of the tools used to measure EF by providing an explanation of the relationship between objective and subjective measures and suggesting potential screening tools for PAE. Typically, there are two ways to measure EF in individuals with PAE: an objective neuropsychological standardized testing and a subjective caregiver rating scale. Objective measures capture individuals' performances in a clinical setting whereas subjective measures reflect individuals' performances in a real-life setting based on a caregiver's observation. Although different tools exist to measure EF, they don't necessary align. Dekker et al. (2017) suggested that "cognitive EF," which is typically measured by objective standardized testing, often tends to tap into different aspects of EF, in contrast with "behavioural EF," which is measured by subjective observations in a real-life setting. Given the array of cognitive, academic, emotional, behavioural, and social measures used in this study, it is important to understand whether both subjective and objective measures are describing the same construct of EF.

Summary of hypothesis

This study aims to expand upon the previous studies to 1) provide local Alberta, Canada data; 2) gain a balanced understanding of the relative neurobehavioral strengths and weaknesses of children with PAE; and 3) examine the relationships of the measures used in the study. The research hypotheses for this paper are:

1. Children in the PAE group will perform worse on all measures than children in the comparison group, especially on core EF measures identified by Miyake et al. (2000) (e.g., inhibitory control, verbal working memory, and cognitive flexibility).
2. Children in the PAE group will display their relative strengths in EF tasks that require less cognitive demand (e.g., verbal and visuospatial short-term memory and auditory attention).
3. The objective measures will have medium-to-high significant correlations with the subjective measures within the same domain but no-to-low significant correlations with the subjective measures across different domains.

Method

Research design

The present study is part of a larger program of research that aims to evaluate the impact of a computerized working memory intervention (Cogmed[®] intervention) on children with PAE (Experimental Group 1), children who born prematurely (Experimental Group 2), and typically developing children (Comparison Group). This study is a quasi-experimental comparison group research study that focuses on examining the performance of children with PAE (PAE group) and typically developing children (comparison group) at the pre-intervention time point. All

procedures for this study were approved by the Health Research Ethics Board – Health Panel (REB 3) at the University of Alberta, and all caregivers as well as participants who were older than seven years provided informed written consent and/or assent at the time of the pre-intervention.

Participants

In PAE group, a sample of 27 children between the ages of four and 13 years ($M = 9.22$, $SD = 2.26$) participated in the pre-intervention testing. These participants were recruited through the FASD clinic at the Glenrose Rehabilitation Hospital, based on a confirmed history of prenatal alcohol exposure (PAE). Since there is a wide range of IQ among individuals with FASD (Streissguth et al., 1997), children with all intellectual abilities were included in this study. In the comparison group, a sample of 19 typically developing children between the ages of four and 13 years ($M = 6.95$, $SD = 2.70$) who were confirmed as having no PAE participated in the pre-intervention testing. Children were excluded from both PAE and comparison groups if they had any of the following conditions: the existence of any genetic disorders (e.g., Down's syndrome), severe neurodevelopmental disorders (e.g., autism) and/or significant motor/sensory impairments (e.g., cerebral palsy, blindness). The demographic data of both groups including age, SES, IQ, gender, ethnicity, current living arrangement, and number of placements is described in Table 3.1.

Procedure

In this study, three recruitment strategies were employed for the PAE group: (1) recruitment of clients from the FASD clinic at the Glenrose Rehabilitation Hospital, (2) recruitment of participants from previous FASD research studies conducted in the same research lab, and (3) recruitment posters placed throughout the community. The confirmation of PAE was obtained

either from a medical record through the hospital (Method 1) or a confirmation document (e.g., medical report or health record) provided by caregivers (Methods 2 and 3). For the comparison group, the caregivers of the typically developing children were recruited either through recruitment posters or invitations issued from experimental group members from the larger research study (e.g., children with PAE and children born prematurely). The caregivers from both groups contacted the research team if they were interested in participating in this study.

In the larger program of research, data was collected pre-intervention, post-intervention, and at follow-up. This paper focused only on the pre-intervention baseline assessment to examine the pre-intervention characteristics of children with PAE. Pre-intervention data from all participants was collected between April 2014 and September 2016. During the pre-intervention baseline assessment, the participants were tested with three neuropsychology assessments and two ability assessments for three to four hours while their caregivers were asked to complete four caregiver behavioural rating measures in the waiting room.

Measures

Neuropsychology assessments. These assessments are standardized assessments that aim to measure the children's executive functions including inhibitory control, attention, and working memory.

Automated Working Memory Assessment. Working memory, including verbal and visuospatial short-term memories and central executive, was assessed by the Automated Working Memory Assessment (AWMA), a computer-based assessment that measures working memory performance in individuals between four and 22 years of age (Alloway, 2007). This battery consists of 12 subtests with three subtests measuring each of the following four areas: verbal short-term memory (e.g., digit recall, word recall, and nonword recall), visuospatial short-term

memory (e.g., dot matrix, mazes memory, and block recall), verbal working memory (e.g., listening recall, counting recall, and backwards digit), and visuospatial working memory (e.g., odd-one-out, Mr. X, and spatial span) (Alloway, 2007). These subtests were administered to measure two domains in this study: short-term memory and working memory. The scores reported in this test were standardized with a mean of 100 and a standard deviation of 15. Good test reliability (e.g., test-retest reliability) and validity (e.g., convergent and divergent validity) have been reported for the AWMA.

NEPSY II – attention and EF domain. The attention and EF domain of the NEPSY II was used to measure attention and EF. Each subtest measures different aspects of attention and executive function for different age ranges. Based on the age group in this study, three subtests were selected: auditory attention (measuring inhibitory and attentional controls), inhibition (measuring inhibitory control), and design fluency (measuring cognitive flexibility). The scores reported in this test are scaled scores with a mean of 10 and a standard deviation of three. The reliability (e.g., test-retest reliability) and validity for these three subtests are adequate (Brooks, Sherman, & Strauss, 2009).

Test of Variables of Attention. Test of Variable of Attention (TOVA) is a computerized neurophysiology measure of attention often used to assess ADHD behaviours in individuals from four years of age and up (Leark, Wallace, & Fitzgerald, 2004). Response patterns on the TOVA provide information that enables practitioners to better understand the type of deficits that might be present. For example, some response patterns suggest inattentiveness or impulsivity, while others may indicate activation/arousal problems or difficulties maintaining vigilance. The attentional control domain is measured by four composite scores in this test: response time variability, response time, commission errors, and omission errors. The scores reported in this

test are standard scores with a mean of 100 and a standard deviation of 15. The reliability (test-retest reliability) and validity of these four variables are sufficient.

Ability and achievement assessments. These are standardized assessment tools that measure the children's intelligence level and mathematics achievement. The measure for intelligence level was collected only at the baseline as it serves only as a moderator in the study whereas the measure for mathematics achievement was collected throughout all three data collection time points.

Wide Range Intelligence Test. The intelligence level was assessed using the Wide Range Intelligence Test (WRIT), which provides a brief estimate of intelligence level for individuals from four years of age and older (Glutting, Adams, & Sheslow, 2000). The WRIT measures three types of IQ scores: verbal IQ, visuospatial IQ, and general IQ. The scores reported in this test are standard scores with a mean of 100 and a standard deviation of 15. The reliability and validity are at a satisfactory level.

Woodcock Johnson Tests of Achievement – 3rd Edition. The standard battery of the Woodcock Johnson Tests of Achievement – 3rd Edition (WJ-III) includes 13 subtests (Woodcock, McGrew, & Mather, 2001), but participants in this study completed the three subtests that measure the mathematic ability domain: calculation, math fluency, and applied problems. The calculation subtest assesses the individuals' mathematical calculation ability without making use of a time restraint. Math fluency measures the rapid application of basic addition, subtraction, and multiplication. Applied problems measures the ability to analyze and solve math problems. The scores reported in this test are standard scores with a mean of 100 and a standard deviation of 15. Strong reliability and validity for the three subtests have been reported.

Behavioural measures and demographic forms. These measures are caregiver

questionnaires aimed to obtain a better understanding of the participants' demographic backgrounds and behaviours at home.

Demographic questionnaire. Caregivers completed a demographic questionnaire about their relationship to the child (e.g., biological or foster parent), living arrangements in the home (e.g., single parent), type and number of previous home placements, and SES. The Hollingshead's (1957) two-factor index of social position was used to evaluate the families' SES. The SES score ranges from 0 to 66 with a higher score indicating a higher SES.

Behavioral Rating Inventory of Executive Function (BRIEF). The BRIEF measures EF behaviors including inhibition, set-shifting, emotional control, working memory, planning, organizational skills, and monitoring skills based on caregivers' observations at home (Gioia, Isquith, Guy, & Kenworthy, 2000). The BRIEF is a caregiver questionnaire for school-age children (five to 18 years of age). The inhibitory control, working memory, cognitive flexibility, and other EF domains were measured by the following composite scores: inhibition, working memory, shifting, emotional control, initiate, planning, organization of materials, and monitor. The scores reported in this test are T scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment. The reliability and validity are at a satisfactory level.

Conners' Parent Rating Scale – 3rd Edition (short version). Conner's parent rating scale – 3rd Edition (Conners-3) assesses ADHD behaviours including behavioural and attentional difficulties in the past three months for children six to 18 years of age based on information from the caregivers (Conners, 2008). Conners-3 consists of 43 questions, and the responses are scored as sums of values on four subscales: oppositional, cognitive problems/inattention, hyperactivity, and ADHD index. The attentional control, mathematic ability, other EF, and emotional and

behavioural issues domains were measured by the following composite scores: inattention, learning problems, EF, aggression, hyperactivity, and peer relation. The scores reported in this test are T scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment. The reliability and validity are at a satisfactory level.

Behavior Assessment System for Children—2nd Edition: Parent Rating Scale

(BASC-2: PRS). The Behavior Assessment System for Children – 2nd Edition (BASC-2) is an integrated questionnaire designed to examine a variety of emotional and behavioural disorders in children aged four to 18 years (Reynolds & Kamphaus, 2004). The information gained from the BASC-2 focuses on both the strengths and weaknesses of the child's social, emotional, and behavioural functioning. The BASC-2 consists of 134 to 160 items, and the responses are scored as the sums of values on each of the five composite scales: adaptive skills, behavioral symptoms index, externalizing problems, internalizing problems, and school problems. The attentional control and emotional and behavioural issues domains were measured by the following composite scores: attention problems, hyperactivity, aggression, conduct problems, anxiety, depression, somatization, atypicality, withdrawal, adaptability, social skills, leadership, activities of daily living, and functional communication. The scores reported in this test are T scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment. BASC-2 displays high reliability and strong validity.

Data analysis

All statistical analyses in this study were conducted using IBM SPSS Statistics version 23.0 (IBM Corporation, 2015). Standardized scores including standard scores, scaled scores, and T scores were used for all analyses. First, 28 independent t-tests were used to compare the performance of the participants in the PAE group to that of the participants in the comparison

group. This was done for the neuropsychology assessments (AWMA, NEPSY-II, TOVA), ability assessment (WJ-III), and behavioural measures (BRIEF, Conners-3, BASC-2) in order to address Research Question 1. Then, descriptive data was used to investigate the PAE group's pattern of performance and to address Research Question 2. Lastly, Pearson's correlation was used to determine the correlation between the objective measures (both neuropsychology and ability assessments) and subjective measures (caregiver behavioural rating) in order to address Research Questions 3. Since this is an exploratory research study, Type II errors have a more significant impact than Type I errors, as failing to avoid a false negative may not lead to any statistical difference between the groups (Rothman, 1990). Therefore, there was no correction of the multiple comparisons.

Results

Performance difference between PAE and comparison groups

Overall, children in the comparison group performed at an average level on all measures except response time variability and omission from TOVA. Children in the PAE group performed significantly lower than children in the comparison group on most of the measures except for the auditory attention and design fluency subtests from the NEPSY as listed in Table 3.1. The overall effect of age and SES between groups was significant as shown in Table 3.1 as children with PAE were older and had lower SES than children in the comparison group. Matching the age and SES between the groups was done by removing three participants from each group. After making that adjustment, the age and SES were no longer significantly different between the groups. However, the pattern of performance across the tests remained the same because there were still significant differences between the groups. Due to the small sample size in this study, the original data was used for all analyses to obtain a higher statistical power.

Neurobehavioral profile of PAE group

As children in the PAE group often performed in the lower-than-average range, relative strengths were defined as any performance in the average range because children with PAE were able to perform as well as their peers. Relative weaknesses were defined as any performances that were at least two standard deviations below the mean.

Demographic information. In general, as shown in Table 3.1, children in the PAE group had a wider range of IQ scores within the sample, more members identifying as Aboriginal, more members living in either adoptive or foster homes, and more placements than children in the comparison group. In terms of IQ, children in the PAE group scored in the low average range for the general intelligence quotient (GIQ) ($M = 87.63$, $SD = 13.49$) and the verbal intelligence quotient (VIQ) ($M = 85.81$, $SD = 15.62$). In contrast, the visuospatial intelligence quotient (VSIQ) ($M = 93.26$, $SD = 10.58$) score, which was in the average range, reflected that visuospatial intelligence is an area of relative strength for children in the PAE group.

Inhibitory control. Inhibitory control was measured by auditory attention and inhibition (naming, inhibition, and switching conditions) subtests from NEPSY-II and an inhibition subscale score from BRIEF. On average, as shown in Table 3.2., children in the PAE group performed at one standard deviation below the mean in the following areas: inhibition (naming condition) ($M = 6.07$, $SD = 3.34$), inhibition (inhibition condition) ($M = 6.81$, $SD = 3.83$), and inhibition (switching condition) ($M = 6.50$, $SD = 2.81$) subtests from NEPSY-II and the inhibition subscale score from BRIEF ($M = 70.59$, $SD = 11.14$). Children in the PAE group performed at an average level on the auditory attention subtest from NEPSY-II ($M = 10.20$, $SD = 4.49$).

Working memory. Working memory was measured by verbal short-term memory,

visuospatial short-term memory, verbal working memory, and visuospatial working memory from AWMA and a working memory subscale from BRIEF. Children in the PAE group performed at an average level on assessments for verbal short-term memory ($M = 94.67$, $SD = 15.95$), verbal working memory ($M = 91.14$, $SD = 12.90$), and visuospatial working memory ($M = 95.75$, $SD = 12.84$) from AWMA. In contrast to their performance on the AWMA ($M = 87.69$, $SD = 12.02$), their performance on the working memory subscale from the BRIEF ($M = 72.96$, $SD = 8.2$) was approximately one to two standard deviations below the mean.

Cognitive flexibility. Cognitive flexibility was measured by design fluency subtest from the NESPY-II and a shifting subscale from the BRIEF. Children in the PAE group performed in the average range on design fluency from NESPY-II ($M = 8.61$, $SD = 3.91$) whereas they scored one standard deviation below the mean on the shifting subscale from BRIEF ($M = 69.00$, $SD = 12.82$).

Attentional control. Attentional control was measured by response time variability, response time, commission errors, and omission errors from TOVA; the inattention composite from Conners-3; and the attention problems composite from BASC-2. Children with PAE performed at more than three standard deviations below the mean on response time variability ($M = 54.65$, $SD = 19.13$) and omission errors ($M = 50.92$, $SD = 19.01$) from TOVA. Moreover, they scored at two standard deviations below the mean on commission errors from TOVA ($M = 65.58$, $SD = 23.07$) and the inattention subscale from Conners-3 ($M = 78.40$, $SD = 11.11$). Lastly, they scored at one standard deviation below the mean on response time from TOVA ($M = 78.85$, $SD = 20.91$) and the attention problems subscale from BASC-2 ($M = 67.33$, $SD = 5.82$).

Other EF domains. Other EF domains were measured by emotional control, initiate, planning, organization of materials, and monitor composites from BRIEF and the EF composite

from Conners-3. Children with PAE performed at two standard deviations below the mean on the planning ($M=71.41$, $SD = 6.80$) and monitor subscales ($M=70.48$, $SD = 8.19$) from BRIEF and the EF subscale from Conners-3 ($M=75.00$, $SD = 13.55$). On other composites, they scored at one standard deviation below the mean: emotional control ($M=65.81$, $SD = 12.82$), initiate ($M=66.37$, $SD = 8.39$), and organization of materials ($M=60.41$, $SD = 9.34$) composites from BRIEF.

Mathematics achievement. Academic achievement was measured by the calculation, math fluency, and applied problems subtests from WJ-III and the learning problem subscales from Conners-3. Children in the PAE group scored at two standard deviations below the mean on the learning problem subscale from Conners-3 ($M=77.84$, $SD = 9.97$) whereas they scored at one standard deviation below the mean on calculation ($M=77.00$, $SD = 19.78$), math fluency ($M=72.13$, $SD = 15.36$), and applied problems ($M=80.88$, $SD = 17.93$) subtests from WJ-III.

Emotional and behavioral functioning. Emotional and behavioral problems were measured by the hyperactivity, aggression, and peer relation subscales from Conners-3 and the hyperactivity, aggression, conduct problems, anxiety, depression, somatization, atypicality, withdrawal, adaptability, social skills, leadership, activities of daily living, and functional communication subscales from BASC-2. The caregivers in the PAE group rated their children in the average range on anxiety ($M=53.89$, $SD = 12.38$), somatization ($M=54.89$, $SD = 15.72$), and withdrawal ($M=59.52$, $SD = 13.63$) subscales from BASC-2; the scores suggest that these areas are relative strengths for children in the PAE group. On the other hand, the caregivers in the PAE group rated their children at one standard deviation below the mean on the following subscales from the BASC-2: aggression ($M=62.15$, $SD = 13.50$), conduct problems ($M=66.80$, $SD = 15.36$), depression ($M=61.56$, $SD = 16.05$), atypicality ($M=67.15$, $SD = 15.03$), adaptability (M

=37.22, $SD = 8.76$), social skills ($M = 38.15$, $SD = 7.77$), leadership ($M = 36.92$, $SD = 7.02$), and activities of daily living ($M = 32.26$, $SD = 7.71$). Similarly, the caregivers also rated the aggression subscales from Conners-3 ($M = 68.44$, $SD = 12.41$) at one standard deviation below the mean. The caregivers in the PAE group rated their children at two standard deviations below the mean on the following BASC-2 subscales: hyperactivity ($M = 71.11$, $SD = 13.24$) and functional communication ($M = 29.81$, $SD = 8.76$). Similarly, the caregivers also rated the hyperactivity ($M = 77.64$, $SD = 13.47$) and peer relation ($M = 72.12$, $SD = 17.04$) subscales from Conners-3 at two standard deviations below the mean.

Understanding objective and subjective EF measures

For the inhibitory control domain, as shown in Table 3.4, there were moderate positive significant correlations between the inhibition (switching condition) subtest from NEPSY-II, the metacognitive index from BRIEF, $r(20) = .52$, $p < .05$, and the global executive composite from BRIEF, $r(20) = .53$, $p < .05$. Yet subtests from NEPSY-II measuring inhibitory control including auditory attention and inhibition (naming and inhibition conditions) were not significantly correlated with any composite scores from BRIEF. On the other hand, as shown in Table 3.3, there were moderate negative significant correlations between inhibition (inhibition condition) subtests from NEPSY-II and both the inattention subscale score, $r(23) = -.42$, $p < .05$, and learning problem subscale score, $r(23) = -.41$, $p < .01$, from the Conners-3. Moreover, there were moderate positive significant correlations between the inhibition (switching) subtest from NEPSY-II and the externalizing composite from BASC-2, $r(20) = .52$, $p < .05$.

For the working memory domain, there were no significant correlations between the AWMA and the BRIEF. In spite of this, as shown in Table 3.3, there was a moderate negative significant correlation between the verbal short-memory subscale scores from AWMA and the

learning problem, $r(23) = -.45, p < .05$, subscale scores from Conners-3. There were also moderate negative significant correlations between visuospatial short-term memory from AWMA and both inattention, $r(23) = -.43, p < .05$, and learning problem, $r(23) = -.54, p < .01$, subscale scores from Conners-3. Lastly, there was a moderate positive correlation between the verbal working memory score from AWMA and the externalizing problem composite score from BASC-2, $r(25) = .47, p < .05$.

For the cognitive flexibility domain, there were no significant correlations between the design fluency subtest from the NEPSY-II and the set-shifting subscale from the BRIEF. In contrast, there were moderate negative significant correlations between the design fluency and learning problem subscale score, $r(19) = -.47, p < .05$, from Conners-3. For the attentional control domain, there were moderate negative significant correlations between the response time variability from TOVA and the hyperactive, $r(22) = -.44, p < .05$, and peer relation, $r(22) = -.42, p < .05$, subscale scores from Conners-3. Moreover, there were moderate positive significant correlations between the response time from TOVA and the inattention subscale score, $r(22) = -.42, p < .05$, from Conners-3. There were moderate positive significant correlations between omission errors from TOVA and both the behavioural regulation index from BRIEF, $r(24) = .51, p < .01$, and the behavioural index from BASC-2, $r(24) = .40, p < .05$. There was no significant correlation between commission errors from TOVA and any other measures. For the mathematics achievement domain, all three subtests from WJ-III were negatively correlated with the learning problem index from Conners-3: calculation, $r(22) = -.43, p < .05$, math fluency, $r(22) = -.45, p < .05$, and applied problem, $r(19) = -.53, p < .01$.

Discussion

Performance differences between the PAE and comparison groups

In general, the findings suggest that participants in the PAE group performed below average on most measures and generally performed more poorly than the participants in the comparison group, as expected for Hypothesis 1. This is consistent with the previous research (Kodituwakku, 2009), indicating that individuals with PAE often have generalized deficits in higher-order cognitive functioning. The results demonstrate that a group difference was found in all EF measures, except for auditory attention and design fluency from NESPY-II. As expected, one of the most prominent problem areas identified in the PAE group was EF (e.g., inhibition, shifting, emotional control, and working memory from BRIEF). Participants from the PAE group also experienced the following problems: academic underachievement (e.g., learning problem from CONNERS-3), externalizing problems (e.g., hyperactivity and inattention from CONNERS-3 and BASC-2), and adaptive functioning (e.g., social skills, activities of daily living, and functional communication from BASC-2). The majority of the problem areas were identified by the caregivers and did not show in the objective test results. One plausible explanation for the gap between the results is that participants with PAE encountered more challenges in their daily lives than in clinical settings, and the caregivers reported the daily challenges. Another explanation is that this pattern may result from raters' frustration when working with this population.

Relative strengths of the PAE group

Relative strength areas are defined as those tasks for which participants' performance fell within the average range. Participants with PAE showed relative strengths on the following measures: working memory, auditory attention, cognitive fluency, and internalizing behaviours.

Working memory. For most working memory measures, including verbal short-term and working memory and visuospatial working memory, participant performance fell in the low end

of the average range. This indicates that these individuals may experience some difficulties in remembering and manipulating information but not to a level that impacts their performance as compared to their peers. This finding contradicts Hypothesis 2, as it posited that working memory tasks are expected to be more cognitively demanding in terms of EF. Although the participants with PAE seemed to be able to perform tasks that required working memory skills in a one-to-one clinical setting, the subjective measure from the BRIEF indicated that they experience moderate-to-severe difficulty in their day-to-day lives as they performed approximately at one-to-two standard deviations below the mean. Caregivers reported that participants with PAE experienced significant working memory deficits throughout their lives even though they were capable of working on tasks that required working memory skills.

Auditory attention and cognitive flexibility. Participants in the PAE group were able to perform the tasks required to sustain auditory attention and cognitive flexibility; they scored in the average range in these two domains (e.g., auditory attention and design fluency from NEPSY-II) as expected in Hypothesis 2. In contrast, the caregivers reported that their children with PAE often experience mild-to-moderate difficulty applying their cognitive flexibility skills in real life. The results suggest that in a clinical setting, children with PAE are able to work well on tasks that require simple, sustained auditory attention and cognitive flexibility, which does not appear to happen in real-life settings.

Internalizing behaviours. Based on the caregiver's rating, participants with PAE did not exhibit internalizing behaviours; the composite score from BASC-2 (parent rating scale) is in the average range as are the anxiety, somatization, and withdrawal scale scores. In contrast, children with PAE exhibit more externalizing problems as they scored at least one standard deviation below the mean. This finding is consistent with previous research: the caregivers of the children

with FASD were more likely to rate their child's externalizing behaviours within the clinical range than they were to rate the internalizing behaviours that way (Pei et al, 2011). The finding may be a result of caregivers' underreporting internalizing behaviours or because the children exhibit fewer symptoms of those behaviours. Moreover, the parent rating scale was used to measure internalizing symptoms instead of the self-rating scale in this study. The absence of internalizing symptoms may also be due to the lack of self-report instrument as the self-rating scales are typically used to capture internalizing symptoms.

Relative weaknesses of PAE group

Relative weaknesses are defined as the areas that the participants struggled with as they performed at least two standard deviations below the mean. Participants with PAE showed relative weaknesses in the following areas: visual attention; observed EF; and behavioural, learning, and adaptive skills.

Visual attention. Participants with PAE experience severe problems with their attentional control as demonstrated by the objective measures from TOVA, which range from one to three standard deviations below the mean. In short, these children experience severe difficulty sustaining visual attention, and exhibit symptoms of inattention. Moreover, they experience moderate level of impulsive symptoms and mild difficulty with their response time in a clinical setting. The caregivers also reported that their children experience clinically significant attention problems in daily life based on the subjective measure, Conners-3. The results are consistent with previous research (Mattson, Crocker & Nguyen, 2011; Pei et al, 2011; Tsang et al., 2016).

Observed EF. In addition to attention issues, participants with PAE tend to experience more problems with inhibition, working memory, and planning in real-life settings; the caregivers reported scores in the clinically significant range in these domains in both the BRIEF

and Conners-3 rating scales. The impairments in these fundamental EFs often have a significant negative impact in the children's daily lives.

Behavioural, learning, and adaptive skills. Other areas of weaknesses are externalizing behaviours, learning problems, peer relations, and functional communication. The caregivers reported that their children with PAE tend to exhibit inattention and hyperactivity symptoms; have difficulty with reading, writing, and math; have trouble relating or interacting with their same age peers, and communicating with others effectively in daily life. Caregivers' reports on these scales place their children in the clinically significant range in both Conners-3 and BASC-2 assessments. The results are consistent with previous research; it is more likely for children with FASD to score within the clinically significant range. When compared to children without PAE who have similar intelligence levels, children with FASD tend to encounter more mental health problems or have poorer outcomes (Tsang et al., 2016).

Summary. Overall, the findings from this study are consistent with previous studies (e.g., Kodituwakku, 2009). Children with PAE may be able to employ strategies to complete simple tasks (e.g., auditory attention) at an average level; however, they may experience difficulties when performing complex tasks (e.g., inhibition) or applying EF in real life. Across domains, it was noted that there were differences between objective testing and subjective testing as raters often described concerns greater than were measured in objective testing. One explanation is that the individuals with PAE were able to work on tasks that required one specific EF skill at a time in the clinical setting but may struggle when asked to integrate different EFs in real-life settings. More research is needed to explore this performance pattern: the subjective measures are rated to be more severe than the objective measures. In general, clinicians should be cautious when they encounter this pattern.

Understanding objective and subjective EF measures

Within domains. In general, most objective measures of EF do not correlate with subjective measures. This suggests that objective measures may address different aspects of core EF, and that the objective and subjective measures tap into different aspects of the EF, as suggested by Dekker et al. (2017). In contrast, the correlations between the objective and subjective measures of attentional control and mathematic achievement are significant. For attentional control, response time from TOVA and the inattention index from Conners-3 were moderately negative correlated, suggesting that individuals who show less inattention symptoms on the Conners-3 tended to react faster on TOVA. One explanation is that when an individual can focus better, he/she can be more on task and react more quickly. For the mathematic ability domain, all three subtests from WJ-III correlated well with the learning problem index from Conners-3. This indicates that individuals whose caregivers reported more learning problems performed worse on all three math subtests from WJ-III. The results suggest that the “measured mathematical underachievement” (WJ-III) and the “observed learning problem” (Conners-3) tapped into the same construct.

Between domains. In addition to the expected correlations, there were unexpected correlations between specific objective and subjective measures, which were opposite than expected. For example, inhibition (switching) from NEPSY-II correlated positively with the metacognitive index from BRIEF, which is comprised of inhibition, working memory, planning, organization, and monitor composites, as well as the externalizing index from BASC-2, which is comprised of hyperactivity, aggression, and conduct problems composite scores. One explanation for the findings may be that individuals with metacognitive impairments and more externalizing problems are better able to inhibit the proponent responses and provide an

alternative response. Moreover, verbal working memory from AWMA was positively correlated to the externalizing index from BASC-2. These findings seem to suggest that the individuals experiencing more externalizing problems are better able to perform on tasks that require verbal working memory. Omission errors from TOVA correlated positively with both the behavioural regulation index from BRIEF, which is comprised of inhibition, shifting, and emotional control; and the behavioural index BASC-2, which is comprised of atypicality, withdrawal, and attention problems. A plausible explanation may be that the more behavioural problems an individual is experiencing the less likely he/she is to be inattentive. Overall, these unexpected findings may suggest that individuals who behave badly may be able to pay attention on tasks, but they may experience difficulty with the tasks, and in turn misbehave as an escape strategy. More research is needed to investigate these unexpected findings to provide a better understanding of how to integrate different EF measures.

Potential screener for PAE. Based on this sample of participants with PAE, Conners-3 seems to be a good subjective measure to screen any potential cognitive deficits experienced by individuals with PAE. The correlation is shown in Table 3.3. First, learning problems were correlated with inhibition, working memory, cognitive flexibility, and mathematic achievement. This suggests that individuals whose caregivers reported more learning problems often experience impairments in all three fundamental components of EF from Miyake et al.'s (2000) model (inhibitory control, working memory, and cognitive flexibility) and mathematic achievement. Second, inattention correlated with inhibition and working memory, suggesting that individuals with more inattentive symptoms often experience the first two fundamental EF deficits. Lastly, hyperactivity and peer relations correlated with response time variability (sustain attention) from TOVA, indicating that individuals with more hyperactivity and peer relationship

problems tend to experience more problems with sustained attention. In short, the results suggest that the deficits reported in Conners-3 (e.g., learning problems, inattention, hyperactivity, and peer relations) correlate closely with the core EF (e.g., inhibition, working memory, and cognitive flexibility), as well as attentional control and mathematic achievement.

Limitations and future studies

Although the study provides neurobehavioral profile findings consistent with previous research, there are several limitations. First, this study aims to describe individuals with PAE in Alberta, Canada, so service providers should be cautious when generalizing the local findings to other jurisdictions. Second, the small sample size restricts the statistical analysis; future studies should include a larger sample size as it will add statistical power to the conclusions. When examining the correlation between the measures, the composite scores in the subjective measures (e.g., BRIEF and BASC-2) were used instead of scale scores in order to reduce the Type I errors due to multiple comparisons. Unfortunately, combining the multiple scale scores into one composite score may increase the risk of a cancelling effect. Future research with a larger sample size should investigate the relationship between different scale scores.

Moreover, the effect of a diagnosis was not explored in this study because the information was not available for some participants, as some who had confirmed PAE were on the waitlist to be assessed for FASD by the FASD clinic. Children with a diagnosis of FASD tend to display more severe cognitive deficits in the areas of verbal reasoning, memory, language functioning, math reasoning and calculation than children who have PAE without a diagnosis (Nash et. al, 2013). Therefore, it is important for future studies to explore how an FASD diagnosis affects cognitive performance.

Lastly, although we tried to account for SES and IQ when comparing groups, a few

confounding factors have not been accounted for in this study. Compared to the comparison group, a higher number of participants in the PAE group were not living with their biological parents and had multiple care placements. It is typical for children with PAE to be raised in adoptive or foster families and to have multiple care placements. These living arrangements may be associated with an unstable home environment, more familial conflicts, and an increased risk of mental health problems, which in turn impact the children's cognitive performance (Streissguth et. al, 2004; Rasmussen, McAuley & Andrew, 2007). Future studies should examine the impact of home environment and multiple care placements on neurobehavioral outcomes in individuals with PAE.

Conclusion

This study highlights children with PAE's strengths and weaknesses in a neurobehavioral profile. In general, children with PAE demonstrated the ability to work on tasks which required working memory, auditory attention, and cognitive fluency in a clinical setting. Thus, clinicians can develop a strength-based intervention to capture these children's strengths (e.g., auditory attention) while remediating their weaknesses (e.g., visual attention). For example, the teacher or caregiver can provide instruction in an audio format as well as a visual format to help building on a child's strengths in auditory attention while strengthening his/her weaknesses in visual attention. In this way, the teacher or caregiver can help the child to develop the necessary confidence to learn in class or at home. Children with PAE often experience more visual attention problems, mental health issues (e.g., inattention and hyperactivity), learning problems (e.g., in reading, writing, and math), and social and communication problems, so it is important to provide proper services for these students and their families. These services include counselling or mental health workshop for the children and caregivers, academic supports in

school (e.g., tutoring or strategy learning), social skills training, and speech therapy or functional communication training.

The finding suggests that the symptoms measured by the “observed EF” (subjective measures) tends to be more severe than the ones in “measured EF” (objective measures), so clinicians and researchers should be cautious when analyzing their data and future studies should continue to investigate the meaning of the measures. In addition, some objective measures map well on subjective measures within and between domains whereas some do not. Thus, more research is needed to gain a better understanding of the relationship between different EF measures. Such an understanding can eventually be used to streamline the assessment process and achieve a standard assessment protocol for FASD.

This study also indicates that individuals who experience significant learning problems tend to have impairments in all three of the fundamental areas of EF (e.g., inhibition, working memory, and cognitive flexibility) as well as lower math achievement scores. This suggests that observed learning problems in the classroom often are an indication of EF impairments. If the teacher or caregiver is able to detect learning problems in students, he/she can refer the students for further screening to assess any potential EF or cognitive problems. Based on the findings of this study, the CONNERS-3 rating scales is useful for detecting the cognitive deficits in individuals with PAE, so it is a potentially effective screener for FASD. Future research should continue to investigate this preliminary finding. With proper diagnosis, students with PAE may receive effective intervention to target their EF deficits at an early age in effort to reduce the risk of adverse outcomes.

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Table 3.1. *Demographic data for the PAE (PAE) and comparison (COM) groups*

Demographic variables	PAE (<i>n</i> =27)	COM (<i>n</i> =19)	df	t-value (2-tailed)
Age [<i>M</i> (<i>SD</i>)]	9.22 (2.26)	6.95 (2.70)	43	-2.30*
SES [<i>M</i> (<i>SD</i>)]	42.24 (12.12)	49.86 (8.68)	44	3.10**
GIQ [<i>M</i> (<i>SD</i>)]	87.63 (13.49)	109.37 (10.93)	44	-5.80**
Verbal IQ	85.81 (15.62)	104.16 (10.57)	44	-4.75**
Visuospatial IQ	93.26 (10.58)	112.47 (12.70)	44	-5.58**
Gender				
Female (<i>n</i>)	12	10		
Male (<i>n</i>)	15	9		
Ethnicity				
Caucasian (<i>n</i>)	7	13		
Aboriginal (<i>n</i>)	20	1		
Current living arrangement				
Biological Parents (<i>n</i>)	0	17		
Adaptive Parents (<i>n</i>)	20	1		
Foster Parents (<i>n</i>)	6	0		
Others/Unidentified (<i>n</i>)	1	1		
Number of Placements [<i>M</i> (<i>SD</i>)]	3.59 (2.59)	0.28 (1.12)	43	5.81**

* Statistically significant at $p < .05$ ** Statistically significant at $p < .01$

Table 3.2. *Pre-intervention descriptive data*

	PAE Group [<i>M</i> (<i>SD</i>)]	Comparison Group [<i>M</i> (<i>SD</i>)]	df	t-value (2- tailed)
AWMA				
Verbal STM	94.67 (15.95)	106.82 (11.36)	44	-2.85**
Verbal WM	91.14 (12.90)	102.23 (14.18)	44	-2.76**
Visuospatial STM	87.69 (12.02)	102.64 (12.40)	44	-4.10**
Visuospatial WM	95.75 (12.84)	109.96 (15.95)	44	-3.34**
TOVA				
Response Time Variability	54.65 (19.13)	86.11 (20.13)	43	-4.92**
Response Time	78.85 (20.91)	95.74 (17.82)	43	-2.84**
Commission Error	65.58 (23.07)	95.59 (24.75)	43	-3.37**
Omission Error	50.92 (19.01)	82.53 (17.55)	43	-5.69**
NEPSY-II				
Auditory Attention	10.20 (4.49)	9.56 (3.54)	39	0.77
Design Fluency	8.61 (3.91)	9.87 (3.31)	36	-1.03
Inhibition - Naming	6.07 (3.34)	9.94 (3.73)	41	-3.51**
Inhibition - Inhibition	6.81 (3.83)	9.88 (3.65)	41	-2.58*
Inhibition - Switching	6.50 (2.81)	11.50 (2.98)	28	-4.25**
BRIEF				
Inhibition	70.59 (11.14)	48.31 (9.63)	38	6.18**
Shifting	69.00 (12.82)	46.85 (7.79)	38	6.76**
Emotional Control	65.81 (12.78)	48.85 (11.75)	38	4.03**
Initiate	66.37 (8.39)	47.46 (10.63)	38	6.12**
WM	72.96 (8.20)	49.69 (11.17)	38	7.46**
Planning	71.41 (6.80)	51.46 (11.93)	38	5.61**
Organization	60.41 (9.34)	51.69 (8.85)	38	2.81**
Monitor	70.48 (8.19)	48.08 (14.75)	38	5.11**
Behavioral Regulation Index	71.30 (11.00)	48.00 (10.39)	38	6.38**
Metacognition Index	71.89 (7.08)	49.23 (12.06)	38	6.28**
Global Executive Composite	73.19 (7.42)	48.77 (10.73)	38	7.40**
CONNERS-3				
Inattention	78.40 (11.11)	53.67 (13.86)	36	6.23**
Hyperactivity	77.64 (13.47)	54.10 (10.13)	36	6.29**
Learning Problem	77.84 (9.97)	51.50 (10.00)	36	7.98**
EF	75.00 (13.55)	55.10 (15.29)	36	4.05**
Aggression	68.44 (12.41)	51.31 (7.42)	36	4.55**
Peer Relation	72.12 (17.04)	49.85 (10.20)	36	5.03**
BASC-2				
Hyperactivity	71.11 (13.24)	45.16 (9.95)	44	7.22**
Aggression	62.15 (13.50)	44.89 (6.60)	44	5.75**
Conduct Problems	66.80 (15.36)	45.18 (8.30)	44	5.45**
Anxiety	53.89 (12.38)	46.47 (10.06)	44	2.16**

Depression	61.56 (16.05)	45.53 (6.53)	44	4.67**
Somatization	54.89 (15.72)	45.16 (6.37)	44	2.90**
Atypicality	67.15 (15.03)	47.79 (8.54)	44	5.54**
Withdrawal	59.52 (13.63)	47.26 (7.35)	44	3.93**
Attention Problems	67.33 (5.82)	44.95 (9.23)	44	9.35**
Adaptability	37.22 (8.76)	56.58 (7.21)	44	-7.92**
Social Skills	38.15 (7.77)	56.79 (8.64)	44	-7.65**
Leadership	36.92 (7.02)	55.73 (8.73)	44	-6.87**
Activities of Daily Living	32.26 (7.71)	53.11 (9.66)	44	-8.13**
Functional Communication	29.81 (8.76)	56.42 (6.66)	44	-11.70**
Externalizing	68.48 (12.28)	43.74 (6.93)	44	7.93**
Internalizing	59.15 (13.16)	44.58 (7.25)	44	4.37**
Behavioral Index	69.07 (10.76)	44.68 (7.92)	44	8.40**
Adaptive	32.11 (6.09)	57.26 (7.99)	44	-12.12**
WJ-III				
Calculation	77.00 (18.78)	104.92 (10.90)	34	-4.75**
Math Fluency	72.13 (15.36)	94.10 (14.86)	32	-3.84**
Applied Problem	80.88 (17.93)	103.26 (9.72)	43	-4.93**

* Statistically significant at $p < .05$

** Statistically significant at $p < .01$

Table 3.3. *Correlation between Conners' Parent Rating Scale (subjective measures) and cognitive deficits (objective measures)*

	Conners' Parent Rating Scale					
	Inattention	Hyperactivity	Learning Problems	EF	Aggression	Peer Relation
AWMA						
Verbal STM	-.35	.03	-.45*	-.20	.22	-.07
Verbal WM	-.35	.21	-.39	-.15	.25	-.07
Visuospatial STM	-.43*	.06	-.54**	-.19	.23	-.14
Visuospatial WM	-.35	-.04	-.31	-.02	.04	-.27
TOVA						
Response Time Variability	-.38	-.44*	-.32	-.17	-.09	-.42*
Response Time Commission Error	-.42*	-.36	-.25	-.22	.10	-.37
Omission Error	.13	.38	.10	-.22	-.04	-.14
	-.02	.04	-.25	-.02	.35	.27
NEPSY-II						
Auditory Attention	-.04	.05	-.04	-.34	.31	.12
Design Fluency	-.17	-.08	-.47*	.20	-.33	-.22
Inhibition - Naming	-.24	.01	-.30	-.15	-.11	-.30
Inhibition - Inhibition	-.42*	.04	-.41*	-.29	.07	-.28
Inhibition - Switching	.19	.24	.02	.35	.02	.06
Inhibition - Total Error	-.27	.01	-.34	-.08	-.05	-.16
WJ-III						
Calculation	-.28	.04	-.43*	-.13	-.16	-.28
Math Fluency	-.10	.13	-.45*	.04	-.19	-.26
Applied Problem	-.19	.11	-.53**	.10	-.19	-.24

* Statistically significant at $p < .05$ ** Statistically significant at $p < .01$

Table 3.4. *Correlation between BRIEF and BASC-2 (subjective measures) and cognitive deficits (objective measures)*

	Behavioral Rating Inventory of Executive Function			Behavior Assessment System for Children Second Edition			
	Behavioral Regulation Index	Metacognition Index	Global Executive Composite	Externalizing Problems	Internalizing Problems	Behavioral Symptoms Index	Adaptive Skills
AWMA							
Verbal STM	.13	.13	.19	.29	.26	.22	-.15
Verbal WM	.18	.14	.23	.47*	.21	.34	.08
Visuospatial STM	.32	.02	.23	.34	.38	.35	-.06
Visuospatial WM	.14	.25	.27	.15	.32	.03	-.02
TOVA							
Response Time Variability	.03	-.11	-.03	-.08	.18	-.19	-.16
Response Time Commission	-.04	-.26	-.18	.22	.15	-.01	-.09
Error	.09	-.07	.02	.05	-.34	-.15	-.32
Omission Error	.51**	.10	.38	.31	.23	.40*	-.27
NEPSY-II							
Auditory Attention	.21	.06	.18	.21	-.06	.13	-.10
Design Fluency	-.31	-.08	-.23	-.19	-.16	-.35	.40
Inhibition – Naming	.23	.16	.27	.31	.14	.18	-.07
Inhibition - Inhibition	.25	.05	.21	.34	.18	.25	-.08
Inhibition - Switching	.34	.52*	.53*	.52*	-.11	.41	-.30
Inhibition – Total Error	.30	.21	.31	.31	.06	.25	-.15
WJ-III							
Calculation	-.20	.00	-.09	.08	-.10	-.17	.33
Math Fluency	-.22	.05	-.09	.11	-.05	-.17	.28
Applied Problem	-.03	.22	.14	.18	.16	.06	.15

* Statistically significant at $p < .05$ ** Statistically significant at $p < .01$

Chapter 4: A working memory intervention for children with PAE: A pilot study (Study 2)

Fetal Alcohol Spectrum Disorder (FASD) is a lifelong disability that results from prenatal alcohol exposure (PAE) (Streissguth *et al.*, 1997). In Canada, the estimated number of individuals affected by FASD is approximately one percent (Cook *et al.*, 2016). Individuals with PAE may suffer from primary disabilities which reflect the underlying brain and central nervous system (CNS) damage. These include poor executive functioning (EF), memory problems, impaired judgment, inability to control impulse behaviour, inability to understand the consequences of actions, impaired mental functioning, and inability to internally modify behavior control (Streissguth *et al.*, 1997). Specifically, working memory (verbal working memory and central executive in particular) is significantly impaired in children with PAE (Rasmussen, 2005). Working memory is a cognitive system that is responsible for the short-term storage and manipulation of information (Baddeley & Hitch, 1974). It is an essential tool for children to effectively meet the demands of the classroom (e.g., remembering multiple-step instructions to perform a task). Many studies have shown that working memory is a key predictor of cognitive performance and academic achievement in children (Nadler & Archibald, 2014). Kodituwakku (2009) suggested that domain-specific cognitive impairments such as those affecting working memory have a generalizing effect to other cognitive functioning because the different brain regions develop interdependently. Since working memory is a fundamental cognitive function that links closely with other EF components and learning, emotional, and behavioural functioning, improvements in working memory may have a generalized effect on other functional areas (Dahlin, 2013; Green *et al.*, 2012; Thorell *et al.*, 2009). Therefore, it is critical to search for an evidence-based intervention to improve working memory in children with PAE to help these children to succeed in school and minimize the adverse outcomes.

Interventions for children with PAE

In the past decade, there has been growing interest in intervention for individuals with PAE; however, there is limited research focusing on evidence-based interventions to address primary disabilities such as those affecting EF (Peadon, Rhys-Jones, Bower, & Elliott, 2009; Reid et al., 2015; Ricco & Gomes, 2013). Interventions studied to date include metacognitive approaches, specific academic and learning initiatives, structured environment supports, and computerized working memory programs.

Metacognitive interventions. Metacognitive interventions are geared towards teaching children strategies to observe, monitor, and manage their own performance during daily activities (Ricco & Gomes, 2013). This type of intervention includes teaching children to monitor and regulate their behaviour through self-talk and using external devices such as written cues to help self-regulate and organize their work (Ricco & Gomes, 2013). The most promising metacognition training method for children with FASD is Cognitive Control Therapy (CCT), which helps students to build competencies by understanding their own learning style and learning challenges and monitoring their own cognitive processes (Kalberg & Buckley, 2007). According to Kalberg and Buckley (2007), after completing CCT children have improvement in body position, movement and awareness, attention, information processing, and organization. The Alert Program for Self-Regulation®, is another metacognition intervention that aims to help children to identify, monitor, and change their level of alertness. It uses the metaphor of a car engine (e.g., changing engine speeds) (Nash et al., 2015). The results from the neuropsychological tests and parental reports reflected that students had better self-regulation (e.g., inhibitory control) after using the Alert Program for Self-Regulation® (Nash et al., 2015).

Moreover, a follow-up study showed evidence of an increase in gray matter, indicating potential brain development after intervention (Soh et al., 2015).

GoFAR is a three-stage computerized game-like metacognition intervention. The stages are Focus and Plan, Act, and Reflect (Coles et al., 2015). This intervention aims to teach children to regulate their attention through repeated trials so that instead of acting in a hurried and impulsive manner, children strengthen their cognitive inhibition (Coles et al., 2015). The goal is to target disruptive behaviours by improving self-regulation and adaptive skills for children with PAE (Coles et al., 2015). A study showed that children's disruptive behaviours decreased after the intervention (Coles et al., 2015). The GoFAR study has added a parent-training component to its subsequent study to educate parents about metacognitive learning strategies and parenting techniques (e.g., how to avoid triggers and prepare for difficult situations) (Kable et al., 2016). The goal of the parental training component is to teach parents to better support their children when training with the GoFAR program. Parental compliance and completion of homework were shown to be indicators of children's success in the training program (Kable et al., 2016). This combined metacognitive intervention led to positive change in children's self-regulation skills (Kable et al., 2016).

Specific academic and learning intervention. Mathematics intervention is another common type of training that addresses EF deficits, especially working memory, because working memory is found to correlate strongly with mathematics performance (Rasmussen & Bisanz, 2011). Mathematics interventions, such as the Math Interactive Learning Experience (MILE), aim to improve a child's mathematical achievement as well as to promote a child's learning readiness and improve behavioural outcomes (Coles, Kable, & Taddeo, 2009). Researchers indicated that MILE leads to improved mathematical outcomes (Coles et al., 2009);

Kully-Martens, 2013) as well as significant behavioural changes. Rehearsal training is another learning intervention that targets working memory deficits by teaching children to rehearse material in order to reduce the chance of forgetting (Loomes, Rasmussen, Pei, Manji, & Andrew, 2008). The researchers found this intervention to be effective in improving working memory and behavioural outcomes (Loomes et al, 2008). Another type of learning intervention is social skills training, such as Promoting Alternative Thinking Strategies (PATH) and Parent-Assisted Child Friendship Training (CFT). These types of interventions are often used to help children recognize and manage feelings and understand and apply knowledge about appropriate social behaviour (Diamond & Lee, 2011; Peadon et al, 2009). They are also used to promote social pretend play. In sum, social skills trainings aim to address deficits in affective and interpersonal problem solving (Peadon et al, 2009; Reid et al., 2015; Zelazo, Qu, & Kesek, 2010).

Structural environment supports. Having structures and supports in the environment is often helpful to address EF challenges (e.g., set-shifting, working memory and attention) for children with PAE (Kalberg & Buckley, 2007). In the classroom, teaching functional routines (e.g., identifying skills, routines, or activities), using visual materials (e.g., individualized daily schedules), and structuring specific tasks (e.g., providing a clear system that helps the child to understand the steps of the tasks) often decreases both visual and auditory distraction and provides structure for the child to follow in order to complete the tasks (Kalberg & Buckley, 2007).

Computerized working memory programs. Computerized programs used to address EF deficits, especially working memory and attention, are based on neuroplasticity principles (Klingberg, 2010). Through repeated practice and reinforcement during training, the child is able to improve his/her working memory capacity, which suggests that associated structural changes

in the brain have occurred (Klingberg, 2010). Computerized program interventions such as Cognitive Carnival (Pei & Kerns, 2012) and Cogmed[©] (Pearson, 2014) use computer games to increase working memory capacity by progressively increasing demands on working memory. Some advantages of using computerized programs are treatment integrity (explicit control of the intervention) and flexibility of time and location (Klingberg, 2010). Computerized programs are more accessible for children with FASD and their families as they can access the training anytime anywhere. This is especially advantageous for families living in rural settings with limited resources and supports or families with busy schedules.

Rationale of Cogmed[©] intervention. The Cogmed[©] intervention is currently the most researched intervention targeting core EF (e.g., working memory) in populations including typically and atypically developing children (e.g., children with attention deficit/hyperactivity disorder [ADHD] and learning disabilities) (Pearson, 2014). The effect of the Cogmed[©] intervention has been observed based on three types of data: neurochemical, neuropsychological, and behavioural. In neuroimaging studies, researchers found that the density of dopamine D1 receptors had been altered (McNab et al., 2009) and brain activity in the prefrontal and parietal areas increased (Olesen, Westerberg, & Klingberg, 2004; Westerberg, & Klingberg, 2007) in healthy young adults who had completed the Cogmed[©] intervention. This is associated with an improvement in working memory capacity (McNab et al., 2009; Westerberg, & Klingberg, 2007). The improvement of working memory capacity is also observed in neuropsychological studies. Researchers found that young people with social, emotional, and behavioural difficulties showed improvement in working memory tasks (e.g., digit span and spatial span) post-intervention (Roughan & Hadwin, 2011). In addition to improvement in working memory, Cogmed[©] intervention also led to a generalizing effect for other cognitive functions. For

example, children with Attention Deficit Hyperactivity Disorder (ADHD) improved on measures of attention, inhibition, and non-verbal reasoning after the intervention (Bigorra, Garolera, Guijarro, & Hervás, 2016; Klingberg et al., 2005; Klingberg, Forssberg, & Westerberg, 2002). Researchers also found positive behavioural changes in children with ADHD; caregivers reported a decrease in inattention and hyperactivity/impulsivity symptoms after intervention (Klingberg et al., 2005). Söderqvist and Nutley (2015) found that after two years after intervention, typically developing students were still performing well in math and reading. Based on these results, Cogmed[©] intervention seems to be effective for improving working memory as well as other cognitive, behavioural, and learning functions.

Given that working memory deficits are common in individuals with PAE (Rasmussen, 2005), it is logical to examine the results of applying this intervention to individuals with PAE. To date, it is unclear how effective the Cogmed[©] intervention is for children with PAE, so this pilot study investigated the short-term impact of the intervention on both targeted (e.g., working memory) and non-targeted EF (e.g., inhibitory control, cognitive flexibility, and attentional control), learning, emotional, and behavioural measures for children with PAE. There is evidence indicating the maintenance effects of the Cogmed[©] intervention from three to six months follow-up in various population including children with ADHD (Holmes et al, 2010), post-secondary students with ADHD (Mawjee, Woltering, & Tannock, 2015), and typically developing adults (Brehmer, Westerberg, & Bäckman, 2012). Yet, Shinaver, Entwistle, and Söderqvist (2014) argued that the evidence is considered speculative regarding long-term effects, as there are not enough studies to warrant a conclusion. Therefore, it is also important to examine the long-term impact of the intervention in this pilot study.

Potential moderators

Researchers have shown that gender (Rasmussen, Horne, & Witol, 2006), age (Rasmussen & Bisanz, 2009a), intelligence level (Connor, Sampson, Bookstein, Barr & Streissguth, 2000), socioeconomic status (SES), and training program choice (Vaurio, Riley, & Mattson, 2008) can impact performance. Although these studies were not intervention studies, these factors may potentially affect the impact of the intervention. By understanding the effect of these moderators on the impact of the intervention, it is helpful in determining who benefit the most from this intervention.

Gender. Based on caregivers' responses to questionnaires, Rasmussen, Horne, and Witol (2006) found that girls with PAE displayed more EF deficits than boys with PAE. Panczakiewicz et al. (2016) reported that boys performed better in language and visual spatial measures and experienced less somatic complaints than girls; however, this finding was not unique for children with PAE as the control group displayed the same pattern. On the other hand, McLachlan et al. (2017) identified no independent moderator effect on gender, but did find a three-way interaction between gender, age, and group on tasks requiring inhibition, verbal short-term memory, and visual working memory. Boys with PAE tended to have more cognitive deficits when they were younger (five to nine years old) than girls with PAE. As they matured (10 to 14 years old), they exhibited fewer cognitive deficits than girls. When they reached late teen years (15 to 18 years old), they once again seemed to have more cognitive deficits than girls. Due to the small sample size, this study will only focus on the independent moderator effect (main effect) and not the interaction effect. With the mixed findings, this paper supported that gender does not influence treatment effect.

Age. As children with PAE get older, their EF deficits tend to become more pronounced. Compared to younger children with PAE, older children with PAE often experience more difficulty with EF tasks, especially verbal EF tasks, relative to the norm (Rasmussen & Bisanz, 2009a). Panczakiewicz et al. (2016) found that younger children exhibited fewer deficits in language and adaptive skills (e.g., communication and socialization) measures; however, this finding again was not unique for children with PAE. As mentioned, McLachlan et al. (2017) found a three-way interaction between gender, age, and group but not an independent moderator effect on age. Based on previous research, this paper supported that younger children are likely to show better treatment effect than older children with PAE (independent moderator effect) as younger children tend to experience fewer EF deficits.

Intelligence Level. The Intelligence Quotient (IQ) of individuals with FASD ranges from 29 to 142, with a mean IQ ranging from 79 to 90 (Streissguth et al., 1997). Within this wide range, 16% of individuals with FASD are considered to have “mental retardation” (IQ < 70) [The term “mental retardation” has since been replaced by “intellectual disability”; however, that term was used in the article referenced]. Research has shown that most EF tasks are not correlated with IQ in either typical developing individuals (Welsh, Pennington, & Groisser, 1991) or individuals with PAE (Connor et al., 2000; Quattlebaum & O’Connor, 2013). Based on these findings, this paper proposed that IQ does not influence treatment effect.

Socioeconomic status. Previous researchers indicated that SES has no impact on EF functioning in individuals with PAE (Vaurio, Riley, & Mattson, 2008), and this paper adopted that premise.

Training program. The Cogmed[®] training programs are different for preschoolers and school-age children: The preschooler’s version is called JM and the school-age children’s

version is RM. Research has shown that both the JM (Thorell et al., 2009; Dongen-Boomsma, Vollebregt, Buitelaar, and Slaats-Willems, 2014) and RM versions (Klingberg et al., 2002) are effective at improving working memory in participants, so there was no reason to expect any difference in outcomes for either version.

Summary of hypothesis

This study examines both the short- and long-term impact of Cogmed[®] intervention on children with PAE. The potential moderating roles of gender, age, intelligence level, and SES were also considered. The research hypotheses for this paper were:

1. Children in both the PAE and comparison groups will show improvement in targeted executive functions (working memory) after the intervention. However, the children in the PAE group will show greater improvement in their working memory abilities when compared to children in the comparison group.
2. Children in both the PAE and comparison groups will show improvement in other non-targeted EFs (e.g., inhibitory control, cognitive flexibility, and attentional control), mathematical achievement, and emotional and behavioral problems after the intervention. However, the children in the PAE group will show greater improvement in the areas of EF and attentional control when compared to children in the comparison group.
3. Younger children (four to six years old) will show greater improvement in both targeted and other non-targeted EFs compared to older children (seven to 13 years old). Gender, IQ, and SES will not influence the treatment effect.

4. The gain in targeted (e.g., working memory) and non-targeted (e.g., inhibitory control, cognitive flexibility, and attentional control) measures from the Cogmed[®] intervention will be sustained at the follow-up for both the PAE and comparison groups.

Method

Research design

The present study is part of a larger program of research evaluating the impact of a computerized working memory intervention (Cogmed[®] intervention) on children with PAE (Experimental Group 1), children who were born prematurely (Experimental Group 2), and typically developing children (Comparison Group). This study is a quasi-experimental comparison group research study that examines the impact of an intervention on participants in PAE and comparison groups at three time points. All procedures for this study were approved by the Health Research Ethics Board – Health Panel (REB 3) at the University of Alberta. All caregivers and participants who were older than seven provided informed written consent and/or assent prior to intervention.

Participants

The PAE group was comprised of 27 children between the ages of four and 13 years old. These children had also participated in the neurobehavioural profile study described in Chapter 3. Seven participants dropped out during the intervention due to school and home schedules, so they completed only the pre-intervention testing. The remaining 20 participants ($M = 9.20$, $SD = 2.59$) completed the 25-session intervention and the testing at three time points: pre-intervention, post-intervention, and follow-up. In the comparison group, 19 children between the ages of four and 13 participated. These participants were typically developing children who were confirmed have no PAE, and who had also participated in the neurobehavioural profile study described in

Chapter 3. One participant dropped out during the intervention due to school and home schedules, and as a result completed only the pre-intervention testing. The remaining 18 participants ($M = 6.78$, $SD = 2.67$) completed the 25-session intervention and the testing at three time points: pre-intervention, post-intervention, and follow-up. Table 4.1 shows the demographic data for both groups, including age, SES, IQ, gender, ethnicity, current living arrangements, and number of placements.

Cogmed[®] intervention

The Cogmed[®] intervention is a computerized working memory training program designed to help children with ADHD to improve their attention and working memory (Pearson, 2014). The Cogmed[®] intervention consists of 25 “video game” training sessions, which the children complete either at home or in school, with the supervision of their caregiver or support person. The Cogmed[®] intervention offers different versions of training programs for different age groups: the JM version for preschoolers (four to six years old) and the RM version for school-age children (seven to 13 years old). During the intervention, the children completed five training sessions a week over a five-week period. Each JM session lasted approximately 15 minutes and each RM session lasted approximately 30-45 minutes. During each session, users participated in three out of seven interactive computer games (JM version) or eight out of 12 interactive computer games (RM version) that targeted different aspects of working memory, specifically verbal and visuospatial short-term memory and verbal and visuospatial working memory. The difficulty level for each game was programmed to automatically adapt to the user’s performance in order to constantly challenge the users to improve their working memory capacity during each session.

Procedure

In this study, there were three data collection time points: pre-intervention, post-intervention, and follow-up, as shown in Figure 4.1. All data for each time point was collected between April 2014 and September 2016.

Pre-intervention. Participants in both the PAE and control groups completed the pre-intervention baseline assessment, which lasted between three and four hours. The battery of baseline assessments consisted of 1) three neuropsychology assessments (Automated Working Memory Assessment (AWMA), NEPSY-II, and the Test of Variables of Attention (TOVA)) to measure inhibitory control, working memory, cognitive flexibility, and attentional control; 2) two ability and achievement assessments (Wide Range Intelligence Test (WRIT) and Woodcock Johnson Tests of Achievement – 3rd Edition (WJ-III)) to measure the intelligence level and mathematics performance; and 3) four caregiver behavioural rating measures (Demographic questionnaire, Behavioral Rating Inventory of Executive Function (BRIEF), Conners' Parent Rating Scale-3rd Edition short version (Conners-3), and Behavior Assessment System for Children-Second Edition (BASC-2)) to measure the child's behaviours at home. On the same day of the baseline assessment, all participants and their caregivers underwent a training session with a research assistant to learn about the Cogmed[®] intervention. In this training session they learned how to access the intervention and play the "games"), how to create a training schedule (e.g., time and location of training), and how to set up the training reward system. After the training session, all participants started the Cogmed[®] intervention either at home or in school for five weeks.

Post-intervention. Once the participants had completed all 25 sessions, they underwent a post-intervention assessment. The battery of post-intervention assessments consisted of tasks

similar to those in the baseline assessment: 1) three neuropsychology assessments (AWMA, NEPSY-II, and TOVA) to measure inhibitory control, working memory, cognitive flexibility, and attentional control; 2) one achievement assessment (WJ-III) to measure mathematics performance; and 3) three caregiver behavioural rating measures (BRIEF, Conners-3, and BASC-2) to measure the child's behaviours at home and to report on the training experience.

Follow-up. Participants received no training for the five weeks after the post-intervention assessment. After the five weeks, they were tested again. The battery of follow-up assessments was similar to the post-intervention assessment, which consisted of 1) three neuropsychology assessments (AWMA, NEPSY-II, and TOVA) to measure inhibitory control, working memory, cognitive flexibility, and attentional control; 2) one achievement assessment (WJ-III) to measure mathematics performance; and 3) three caregiver behavioural rating measures (BRIEF, Conners-3, and BASC-2) to measure the child's behaviours at home.

Measures

Neuropsychology assessments. This type of objective assessment measured the children's EF, including inhibitory control, working memory, cognitive flexibility, and attentional control. The data for the following assessments was collected throughout all three data collection time points.

Automated Working Memory Assessment. The AWMA is a computerized assessment that measures four areas of working memory: verbal short-term memory, visuospatial short-term memory, verbal working memory, and visuospatial working memory (Alloway, 2007). AWMA consists of a total of 12 subtests with three subtests measuring each area. The test scores are standardized with a mean of 100 and a standard deviation of 15. Good test reliability and validity have been reported for the AWMA.

NEPSY II – attention and EF domain. The attention and EF domain of the NEPSY-II measures a range of skills: inhibition; monitoring and self-regulation; vigilance; selective and sustained attention; working memory; nonverbal problem solving; planning and organization; and figural fluency (Korkman, Kirk, & Kemp, 2007). Three of the six subtests were selected based on the age groups in this study to measure different aspects of attention and EF including the inhibitory control, cognitive flexibility, and attentional control domains (auditory attention, inhibition, and design fluency). The test scores are scaled scores with a mean of 10 and a standard deviation of three. Good test reliability and validity have been reported for these three subtests (Brooks, Sherman, & Strauss, 2009).

Test of Variables of Attention. The TOVA is a computerized assessment that measures four major areas of attentional control: response time variability, response time, commission errors, and omission errors (Leark, Wallace, & Fitzgerald, 2004). The test scores are standard scores with a mean of 100 and a standard deviation of 15. Good test reliability and validity of these four variables have been reported.

Ability assessments. This type of objective assessment measured the children's intelligence level and mathematics ability. The intelligence assessment was only used at the baseline assessment whereas the data from the mathematics ability was collected at all three time points.

Wide Range Intelligence Test. WRIT provides a brief estimate of the intelligence level for individuals using three IQ scores: Verbal IQ, Visuospatial IQ, and General IQ (Glutting, Adams, & Sheslow, 2000). The test scores are standard scores with a mean of 100 and a standard deviation of 15, and the reliability and validity are at a satisfactory level.

Woodcock Johnson Tests of Achievement – 3rd Edition. The three subtests of WJ-III

measure the mathematics ability domain: calculation, math fluency, and applied problems (Woodcock, McGrew, & Mather, 2001). The test scores are standard scores with a mean of 100 and a standard deviation of 15. Strong reliability and validity for the three subtests have been reported.

Behavioural measures forms. The caregivers completed all questionnaires, the use of which was intended to obtain a better understanding of the child's demographic background and behaviours at home. All caregiver questionnaires except the demographic questionnaire were administered at all three time points. The demographic questionnaire was used only during the baseline assessment.

Demographic questionnaire. Demographic information about the guardianship of the child (e.g., biological or foster parent), living arrangements in the home (e.g., single parent), type and number of previous home placements, and SES was measured. Then, the family's SES was calculated based on Hollingshead's (1957) two-factor index of social position. The score ranges from 0 to 66 with a higher score indicating a higher SES.

Behavioral Rating Inventory of Executive Function. BRIEF measures the behaviours that caregivers observe at home and which indicate EF impairment. The scale scores include inhibition, set-shifting, emotional control, working memory, planning, organizational skills, and monitoring skills (Gioia, Isquith, Guy, & Kenworthy, 2000). The scale scores can be further summarized into three composite scores: behavioural regulation, metacognition, and global executive composite. Good reliability and validity have been reported. The test scores are T-scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment.

Conners' Parent Rating Scale – 3rd Edition (short version). Conners-3 assesses the

caregiver's perspective of the child's behavioural and attentional difficulties (Conners, 2008). The attentional control, mathematics ability, other EFs, and other emotional and behavioural issues domains were measured using the following scale scores: inattention, learning problems, EFs, aggression, hyperactivity, and peer relation. The test reliability and validity are at a satisfactory level. The test scores reported are T-scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment.

Behavior Assessment System for Children – Second Edition: Parent Rating Scale. The BASC-2 examines a variety of attentional, emotional, and behavioural problems including the following scale scores: attention problems, hyperactivity, aggression, conduct problems, anxiety, depression, somatization, atypicality, withdrawal, adaptability, social skills, leadership, activities of daily living, and functional communication. The scale scores can be further summarized into five composite scores: adaptive skills, behavioral symptoms index, externalizing problems, internalizing problems, and school problems. Good reliability and validity have been reported. The test scores are T-scores with a mean of 50 and a standard deviation of 10, with a higher score indicating more impairment.

Statistical analysis

All statistical analyses in this study were conducted using IBM SPSS Statistics version 23.0 (IBM Corporation, 2015). Standardized scores including standard scores, scaled scores, and T-scores were used for all analyses. The first series of analyses focused on examining short- and long-term treatment effects on the targeted EF (working memory). First, a set of four pairwise t-tests were computed for each group (PAE and comparison groups) between pre- and post-intervention time points to answer the within-group difference aspect of Research Question #1. This analysis determined whether completing the Cogmed[®] intervention improves the four areas

of working memory measured by AWMA: verbal short-term memory, visuospatial short-term memory, verbal working memory, and visuospatial working memory. Second, for any significant t-tests between pre- and post-intervention time points, further pairwise t-tests were conducted between post-intervention and follow-up. This analysis addressed Research Question #4 by investigating whether there was a sustained gain on the working memory measures from the post-intervention. Third, four independent t-tests were computed to explore the group difference of the treatment effect to address the between-group difference aspect of Research Question #1. Fourth, the difference in the four working memory measures of AWMA between pre- and post-intervention was first calculated for each individual. Then, that difference was categorized into an increment of 7.5 points, which is 0.5 standard deviation of the tool. For example, if the difference were 10 points, it would fall under the category of 0.5 to 1.0 changes. This analysis used to examine the individual treatment effect in both groups to further explore Research Question #1. Lastly, Pearson's correlations were used to determine the correlation between moderators (age, gender, IQ, SES, and training programs) and the score difference between pre- and post- intervention time points. This analysis answered Research Question #3.

The second series of analyses focused on examining the short- and long-term training effects on the non-targeted EFs (e.g., inhibitory control, cognitive flexibility, and attentional control), mathematics ability, and emotional and behavioral problem measures. First, a set of 24 pairwise t-tests was computed for each group (PAE and comparison groups) between pre- and post-intervention time points to address the within-group difference of Research Question #2. Second, for any significant t-tests between pre- and post-intervention time points, further pairwise t-tests were conducted between post-intervention and follow-up to address Research Question #4. Third, 24 independent t-tests were computed to explore the group difference in the

treatment effect to address the between-group difference aspect of Research Question #2. Lastly, Pearson's correlations were used to determine the correlation between moderators and the score difference to answer Research Question #3. The significance level was set at .05 for all analyses. In this exploratory study, Type I errors were not corrected despite multiple comparisons in order to avoid false negatives on any statistical difference between the groups (Rothman, 1990).

Results

Intervention impact on targeted executive functions

This section examines the short- and long-term training effects on the targeted EFs (working memory). Four pairwise t-tests were calculated to reveal the intervention impact on working memory for each group between pre- and post-interventions.

PAE group. A significant improvement in three out of four areas of working memory measured by AWMA was found in the PAE group after 25 sessions of Cogmed[®] intervention. There was a significant improvement in verbal short-term memory before ($M = 99.42$, $SD = 14.96$) and after intervention ($M = 108.21$, $SD = 14.03$); $t(19) = -3.921$, $p = .001$. Similarly, there was a significant improvement in visuospatial short-term memory before ($M = 89.89$, $SD = 12.79$) and after intervention ($M = 100.01$, $SD = 16.04$); $t(19) = -3.152$, $p = .005$. Lastly, there was a significant improvement in verbal working memory before ($M = 93.25$, $SD = 14.10$) and after intervention ($M = 99.41$, $SD = 16.00$); $t(19) = -2.972$, $p = .008$.

At the five-week follow-up, visuospatial short-term memory continued to improve, showing improvement from post-intervention ($M = 100.01$, $SD = 16.04$) to follow-up ($M = 106.00$, $SD = 13.27$); $t(19) = -2.411$, $p = .026$. This was the only working memory improvement that continued to get better. On the other hand, there was a significant decrease in verbal short-term memory from post-intervention ($M = 108.21$, $SD = 14.03$) to follow-up ($M = 103.04$, $SD =$

16.34); $t(19) = 2.539, p = .020$. Although there were no significant improvements at the follow-up time point for verbal short-term memory, verbal working memory, and visuospatial working memory, the mean scores at follow-up were at least the same or better compared to the pre-intervention time point, as shown in Figure 4.2.

Comparison group. After the intervention, there were significant improvements in the comparison group in all four areas of working memory measured by AWMA. A significant improvement in verbal short-term memory was found between pre- ($M = 106.92, SD = 11.68$) and post-intervention ($M = 117.96, SD = 10.56$); $t(17) = -3.985, p = .001$. Likewise, there was a significant improvement in visuospatial short-term memory before ($M = 103.68, SD = 11.88$) and after intervention ($M = 116.25, SD = 9.64$); $t(17) = -3.986, p = .001$. Another significant improvement in verbal working memory was found between pre- ($M = 102.69, SD = 14.44$) and post-intervention ($M = 110.04, SD = 10.14$); $t(17) = -2.522, p = .022$. Lastly, there was a significant improvement in visuospatial working memory between pre- ($M = 111.35, SD = 15.19$) and post-intervention ($M = 122.72, SD = 9.88$); $t(17) = -3.940, p = .001$.

At the five-week follow-up, no working memory improvement was found. However, all mean scores at the follow-up were higher than at the pre-intervention time point, as shown in Figure 4.3.

Difference between groups. Between the pre- and post-interventions, there was a significant group difference in the treatment effect in one of four areas of working memory measured by AWMA. The significant difference occurred in the visuospatial working memory between the PAE ($M = 3.06, SD = 10.30$) and comparison groups ($M = 11.37, SD = 12.25$); $t(36) = -2.272, p = .029$. This finding suggested that the comparison group improved more than the PAE group on the visuospatial working memory measure.

Individual intervention impact on targeted executive functions

The individual treatment effect in four working memory measures of AWMA was described in an increment of 0.5 standard deviation of the tool. Any changes that are greater than 0.5 standard deviations are defined as improvements. In the PAE group, 5% of participants ($n=1$) improved their verbal short-term memory whereas 45% of participants ($n=9$) improved their visuospatial short-term memory after the intervention. Moreover, 30% of participants ($n=6$) improved their verbal working memory whereas 20% of participants ($n=4$) improved their visuospatial working memory after the intervention, as shown in Figure 4.4. An analysis of the participants who improved after the intervention revealed that 35% ($n=7$) improved in one of four areas of working memory as shown in Table 4.2., 25% ($n=5$) improved in two areas, and 5% ($n=1$) improved in three areas. Overall, 65% of the participants ($n=13$) improved in at least one area of working memory.

In the comparison group, 6% of participants ($n=1$) improved their verbal short-term memory whereas 44% of participants ($n=8$) improved their visuospatial short-term memory. Moreover, 56% ($n=10$) improved their verbal working memory whereas 22% ($n=4$) improved their visuospatial working memory as shown in Figure 4.5. Of the participants who improved after intervention, 33% ($n=6$) improved in one of four areas of working memory as shown in Table 4.2, 39% ($n=7$) improved in two areas, and 6% ($n=1$) improved in three areas. Overall, 78% of participants ($n=14$) showed at least one area of working memory improvement.

Intervention impact on non-targeted executive functions

This section examines the short- and long-term training effects on non-targeted EFs (e.g., inhibitory control, cognitive flexibility, and attentional control), mathematics ability, and other emotional and behavioral problem measures.

PAE group. In the PAE group, significant improvement was found in four of 24 pairwise t-tests after the intervention. First, significant improvement was reported in both areas of attentional control measured by TOVA. There was a significant decrease in commission errors before ($M = 66.95$, $SD = 23.84$) and after the intervention ($M = 78.74$, $SD = 27.18$); $t(18) = -3.143$, $p = .006$. Similarly, there was a significant decrease in omission errors before ($M = 54.79$, $SD = 21.04$) and after the intervention ($M = 65.11$, $SD = 22.92$); $t(18) = -2.696$, $p = .015$. Second, a significant improvement in mathematics achievement, especially in applied problems (WJ-III), was detected from pre- ($M = 81.90$, $SD = 18.22$) to post-intervention ($M = 85.60$, $SD = 16.67$); $t(19) = -2.820$, $p = .011$. Lastly, there was a significant improvement in adaptive skills measured by BASC-2 from pre- ($M = 31.40$, $SD = 6.00$) to post-intervention ($M = 33.50$, $SD = 6.94$); $t(19) = -2.483$, $p = .023$. A significant increase in externalizing problems (e.g., hyperactivity, aggression, and/or conduct problems) was found as measured by BASC-2 from pre- ($M = 68.10$, $SD = 13.87$) to post-intervention ($M = 70.65$, $SD = 13.628$); $t(19) = -2.495$, $p = .022$.

Between post-intervention and the five-week follow-up, only adaptive skills measured by BASC-2 were reported to continue showing improvement from post-intervention ($M = 38.05$, $SD = 10.03$) to follow-up ($M = 33.50$, $SD = 6.94$); $t(19) = -2.417$, $p = .026$. Although there was no significant improvement in commission and omission errors (TOVA) and applied problems (WJ-III) at the follow-up, the mean scores at the follow-up were at least the same as or better compared to the pre-intervention time point as shown in Figure 4.6. Externalizing problems were found to be worse at the follow-up than at pre-intervention, but there was no significant difference from post-intervention to the follow-up.

Comparison group. In the comparison group, a significant improvement was found in three out of 24 pairwise t-tests after the intervention. First, a significant improvement in

commission errors measured by TOVA was found before ($M = 90.31, SD = 26.51$) and after the intervention ($M = 97.06, SD = 23.79$); $t(15) = -2.220, p = .042$. Second, a significant improvement was reported in the two subtests from the inhibitory control domain measured by NEPSY-II. There was a significant improvement in the naming subtest before ($M = 9.93, SD = 3.86$) and after the intervention ($M = 12.73, SD = 3.99$); $t(14) = -3.761, p = .002$. Likewise, there was a significant improvement in the inhibition subtest between pre- ($M = 9.80, SD = 3.76$) and post-intervention ($M = 12.60, SD = 3.68$); $t(14) = -2.758, p = .015$.

At the five-week follow-up, no significant improvement was found. However, the mean scores of the three reported measures at the follow-up were higher than at both the pre- and post-intervention time points, as shown in Figure 4.7.

Difference between groups. The intervention impact in two of 24 non-targeted EFs was significantly different between the two groups between the pre- and post-interventions. A significant group difference in adaptive skills improvement measured by BASC-2 was found between the PAE ($M = 2.10, SD = 3.78$) and comparison groups ($M = -0.94, SD = 5.10$); $t(36) = 2.106, p = .042$. Moreover, a significant group difference in the inhibition subtest of the NEPSY-II between the PAE ($M = -0.52, SD = 2.04$) and comparison group ($M = 2.80, SD = 3.93$); $t(34) = -3.312, p = .002$. This finding suggested that the PAE group showed more improvement in adaptive skills whereas comparison group showed more improvement in inhibitory control measure.

Potential moderators

There are five potential moderators to be considered in this study: age at the time of training, gender, IQ at the time of training, SES, and type of training programs. Multiple Pearson's correlations were calculated to determine the potential moderator effects. In the PAE

group, there were multiple significant correlations between the treatment effect and moderators (age, gender, IQ, and type of training programs). First, there were significant correlations between age and the change in both verbal, $r(18) = -.463, p=.04$, and visuospatial, $r(18) = .448, p=.048$, short-term memory measured by AWMA from pre- to post-intervention. This finding suggested that the older children improved more on verbal short-term memory measure whereas the younger children improved more on visual spatial short-term memory. There was also a significant correlation between age and the intervention impact on response time measured by TOVA, $r(17) = -.544, p=.016$. Second, there were significant correlations between gender and the improvement in both hyperactivity, $r(16) = -.553, p=.017$, and inattention, $r(16) = -.710, p=.001$, scale scores measured by Conners-3. Third, there was a significant correlation between General Intelligence Quotient (GIQ) and the treatment effect in the calculation subtest measured by WJ-III, $r(15) = .514, p=.035$. Lastly, there were significant correlations between the type of training program and the improvement in the behavioral symptoms index measured by BASC-2, $r(18) = -.466, p=.038$, and the calculation subtest measured by WJ-III, $r(15) = -.494, p=.044$.

In the comparison group, there were also multiple significant correlations between the treatment effect and moderators (age, gender, IQ, and SES). First, there was a significant correlation between gender and the improvement in the behavioral symptoms index measured by BASC-2, $r(17) = -.472, p=.048$. This finding suggested that the girls showed more improvement in behavioural symptoms than boys. Second, there was a significant correlation between GIQ and the intervention impact on auditory attention measured by NEPSY-II, $r(13) = -.655, p=.008$. Third, there were significant correlations between SES and the improvement in both response time, $r(13) = .805, p<.001$ and commission errors, $r(13) = .524, p=.045$, measured by TOVA and the design fluency subtest measured by NEPSY-II, $r(11) = -.717, p=.006$. Moreover, from the

pre- to post-intervention, there were significant correlations between SES and the change in the learning problem scale scores, $r(10) = -.758, p=.004$, and peer relation $r(10) = .690, p=.013$, scale scores measured by Conners-3 and the behavioural regulation composite scores from BRIEF, $r(10) = -.589, p=.044$.

Discussion

This pilot study examined the short- and long-term impacts of the 25 sessions of Cogmed[®] intervention on targeted and non-targeted EFs in children with PAE and typically developing children.

Intervention impact on targeted executive functions

After 25 sessions of the Cogmed[®] intervention, both the PAE and comparison groups showed significant improvements in working memory. In general, the PAE group improved in three areas of working memory: verbal short-term memory, visuospatial short-term memory, and verbal working memory. Specifically, after the intervention most participants in the PAE group showed the most improvement in their visuospatial short-term memory, followed by verbal working memory, visuospatial working memory, and verbal short-term memory. Overall, 65% of participants in the PAE group improved in at least one area of their working memory.

Likewise, the comparison group showed significant improvement in all four areas of working memory: verbal short-term memory, visuospatial short-term memory, verbal working memory, and visuospatial working memory. In particular, after the intervention most participants in the comparison group showed the most improvement in their verbal working memory, followed by visuospatial short-term memory, visuospatial working memory, and verbal short-term memory. Overall, 78% of the participants in the comparison group improved in at least one area of their working memory. The findings are consistent with previous intervention studies

examining working memory improvement in atypically and typically developing preschoolers and children (Chacko et al., 2014; Roughan & Hadwin, 2011). Contrary to the hypothesis, children in the comparison group showed more improvement in visuospatial working memory than children in the PAE group, who did not show improvement in this area. One possible explanation is that visuospatial working memory deficits are at the root of working memory difficulties for children with PAE, and will take longer to remedy. Thus, future interventions may need to be geared towards that area of function in order to help support this skill. Overall, the findings suggest that the Cogmed[®] intervention is effective at improving at least three areas of working memory in both groups after 25 sessions of training, as posited in Hypothesis #1. Moreover, more than half of the sample in both groups showed improvements in at least one area of working memory.

Follow up. At the five-week follow-up, the participants from the PAE group continued to show significant improvements in visuospatial short-term memory; however, their performance in verbal short-term memory decreased when compared to their post-intervention scores. Despite this, their performance in all four areas were at least the same as or better than their pre-intervention scores. Similarly, the participants from the comparison group had scores that were better than those from their pre-intervention assessments, even though they did not show continuous improvement from post-intervention to the follow-up. The findings suggest a potential long-term treatment effect, as posited in Hypothesis #4.

Intervention impact on non-targeted executive functions

After 25 sessions of the Cogmed[®] intervention, both the PAE and comparison groups showed significant improvements in attentional control, as posited in Hypothesis #2. Specifically, participants from the PAE group improved on both commission and omission

errors, suggesting that the intervention helped them to reduce impulsivity and improve their visual attention. Likewise, participants from the comparison groups improved on their commission errors, suggesting that they reduced their impulsivity after the intervention. The findings are consistent with previous studies showing that both atypically and typically developing children improve on objective measures of attentional control after an intervention (Green et al., 2012; Thorell et al., 2009).

In terms of inhibitory control, the treatment effect was observed in the comparison group. The participants in the comparison group improved on both naming and inhibition subtests after the intervention; however, this effect was not observed in the PAE group. One possible explanation is that children with PAE experience more severe deficits in this area compared to other EF deficits, as described in Chapter 3 and previous research (Rasmussen et al, 2013). Therefore, more intense interventions may be needed.

In terms of mathematics achievement, effects of the treatment were observed in the PAE group but not in the comparison group. The participants from the PAE group showed improvement on the applied problems subtest. Since working memory is closely linked to mathematics performance (Rasmussen & Bisanz, 2011), the improvement in the working memory deficits for participants with PAE may lead to improved mathematics performance.

In terms of subjective measures, the effects of the treatment effect were found only in the PAE group, where there was improvement in adaptive composite scores, suggesting that the caregivers viewed the participants to have improved their adaptive skills after the intervention. There was also an increase in externalizing problems in the PAE group, suggesting that the caregivers described the participants as exhibiting more externalizing problems (e.g., hyperactivity, aggression, and/or conduct problems) after the intervention. Overall, based on

subjective caregiver ratings, most non-targeted EF improvements were observed through objective measures in an experimental setting, but the improvements were not generalized to real-life settings. According to Shipstead, Redick, and Engle (2010), the generalization effect is unclear for the Cogmed[®] intervention as previous studies encountered similar problems. Future studies need to further investigate the generalization effect.

Children in the comparison group improved their inhibitory control after intervention, whereas children in the PAE group did not. This finding suggests that inhibitory control is an area of challenge for individuals with PAE. Children in the PAE group showed more improvement in adaptive skills than children in the comparison group but this could be because before the intervention, deficits in adaptive skills were observed only in the PAE group.

Follow up. At the five-week follow-up, the participants in the PAE group continued to show significant improvements in adaptive skills. Although they did not show a significant change in commission errors (TOVA) and applied problems (WJ-III) between post-intervention and the follow-up, the follow-up scores in these areas were better than the post-intervention scores. Omission errors (TOVA) is another area in which there was not a significant change at the follow-up, but the follow-up score was better than the pre-intervention score. Thus, the improvements in impulsivity, visual attention, and mathematics performance are signs that the intervention was effective, as attentional control and mathematics performance are closely linked to working memory (targeted EF). Moreover, the caregivers reported that the participants improved in their adaptive skills. On the other hand, the caregivers viewed the participants' externalizing problems as worse than at the pre- and post-interventions.

In the comparison group, the participants did not show continuous improvement from the post-intervention to the follow-up, but on the non-targeted measures, they performed better than

they had at the pre- and post-interventions. The findings suggest potential long-term treatment effects, as the treatment effects on some non-targeted measures seem to be sustained after an intervention.

Potential moderators

In this paper, there are multiple significant correlations between moderators and targeted and non-targeted EF measures, suggesting that these moderators have an impact on the treatment effect. As predicted in Hypothesis 3, age moderates the effects of an intervention; however, this moderator effect is only observed in certain measures, such as working memory and attentional control. The other moderators such as gender, GIQ, SES, and training programs also seem to have a potential effect. The following section will discuss how the moderators influence the effects of intervention in both targeted and non-targeted measures.

In terms of targeted measures (working memory), age is the only moderator that affected the change between pre- and post-intervention time points in the PAE group. Older children showed more improvement in verbal working memory than younger children, whereas younger children improved more in the area of visuospatial working memory than older children. One plausible explanation is that children tend to rely more on their visuospatial working memory when they are young and to slowly switch to verbal working memory as they mature (Manji, Pei, Loomes, & Rasmussen, 2009). Thus, younger children may show more gains in their predominant working memory after intervention, while the opposite is true for older children. There was no moderator effect in the comparison group. The mixed findings on age in the targeted measures suggest a possible interaction moderator effect as presented in McLachlan et al. (2017), instead of a simple independent moderator effect. Future studies are needed to clarify the potential interaction effect.

In terms of non-targeted objective measures (standardized testing), age, GIQ, SES, and the type of training program correlate significantly with intervention gains in four main areas: inhibitory control, cognitive flexibility, attentional control, and math achievement. First, GIQ correlates significantly with inhibitory control in the comparison group, as children with lower GIQ improved more in the area of auditory attention than children with a higher GIQ. This effect did not appear in the PAE group as posited in the hypothesis. Unlike previous studies of typically developing individuals (Welsh et al., 1991), these findings suggest that GIQ moderates the effect of intervention for the inhibitory control in the comparison group. The mixed findings on GIQ may suggest an interaction moderator effect as GIQ has an impact on the comparison group but not on the PAE group. Future studies are needed to further examine how IQ impacts the effects of treatment.

Second, SES is significantly correlated with the change in cognitive flexibility in the comparison group between the pre- and post-interventions; however, it did not have a significant impact on the PAE group. The findings confirm those of a previous study which found that SES is not a moderator in the PAE group (Vaurio et al, 2008). In contrast, SES had a significant impact on the effect of intervention for cognitive flexibility in the comparison group, as children with a lower SES improved their cognitive flexibility more than children with a higher SES. The findings may suggest an interaction moderator effect; thus, future studies are needed to investigate the moderating role that SES has on the effects of treatment for cognitive flexibility.

Third, both age and SES play a significant role in the effects of treatment for attentional control. In the PAE group, younger children had more improved reaction time than older children, consistent with Hypothesis 3. In the comparison group, children with higher SES improved more on the impulsivity measure than children with lower SES whereas children with

lower SES had more improved reaction time than children with higher SES. The mixed results suggest that SES influences the effects of intervention for attentional control; however, the moderator effect is unclear. Future studies are needed to examine this issue. Lastly, both GIQ and the type of training program correlated significantly with intervention gains in mathematics performance for the PAE group. These effects were not observed in the comparison group. In the PAE group, children with a higher GIQ had more improved calculation subtests than children with a lower GIQ. A plausible explanation is that children with a higher GIQ have potential for greater improvement in academic tasks. Similarly, children in the JM version (ages four to six) showed more improvement in calculation subtests than children in the RM version (ages seven to 13). The findings do not support the previous studies that looked at training programs (Dongen-Boomsma et al, 2014; Klingberg et al., 2002; Thorell et al., 2009) but support previous studies that looked at age (Rasmussen & Bisanz, 2009a) as the treatments had a better effect on younger children in the PAE than on older children in that group.

In terms of non-targeted subjective measures (caregiver ratings), gender, SES, and type of training programs correlated significantly with intervention gains. First, the findings from this study contradict with the previous studies (McLachlan et al., 2017) as gender moderates the effect of intervention in both groups. Girls in the PAE group showed less hyperactive and inattentive symptoms whereas girls in the comparison group showed less behavioural problems reported by the caregivers. Second, SES seemed to influence behavioural and learning changes in the comparison group between the pre- and post-interventions; however, it did not have a significant impact on the PAE group. In the comparison group, children with higher SES showed more improvement in peer relations than children with lower SES whereas children with lower SES improved more on the learning problems and behavioural regulation scales than children

with higher SES. The findings about SES in the PAE group are consistent with those from a previous study, which found no SES impact on the effects of intervention in the PAE group (Vaurio et al, 2008). Despite this, there were mixed findings in the comparison group. To gain a better understanding of this, the moderating role of SES needs to be further investigated in future studies. Lastly, the type of training program correlated significantly with the behavioural problems reported by the caregivers in the PAE group; children in JM version (ages four to six) showed more improvement in behaviour than children in the RM version (ages seven to 13). This effect was not found in the comparison group. The findings contradict previous studies about types of training programs (Dongen-Boomsma et al, 2014; Klingberg et al., 2002; Thorell et al., 2009) but are consistent with previous studies about age (Rasmussen & Bisanz, 2009a) as the treatment had a better effect on younger children with PAE who had behavioural problems than on older children with PAE. In conclusion, evidence from this study suggests that the intervention is effective at improving working memory and other functioning. Therefore, Cogmed[®] intervention should be implemented into educational systems or provided as a targeted intervention in a rehabilitation setting that provides services for individuals with PAE.

Limitations and future directions

Despite the promising preliminary findings presented in this paper, there are three major areas of limitations: research design, intervention program, and methodology. This quasi-experimental study lacks a control group, who does not receive the intervention; therefore, the children's maturation may play a role in these positive findings. In order to rule out the maturation and support the robustness of the findings, future studies should address these limitations by involving a blind control group and random assignment.

Another limitation is the Cogmed[®] intervention. Because it is computer-programmed, it

allows researchers to have explicit control of the intervention, but it also has problems with treatment integrity when used with individuals with PAE. Only 50% of the participants ($n=10$) in the PAE group completed the training within 25 days, and most of those had participated in the RM version. Although each session of the RM version is designed to be completed in 30 to 45 minutes each day, caregivers reported that the participants in the PAE group typically took more time to complete each session because of several hurdles that lengthened the training sessions: 1) the computer program suggested frequent breaks in response to sequential incorrect answers; 2) the participants had limited ability to control their attention; and 3) the school-aged participants had a busier schedule compared to the preschool participants. For these reasons, some participants decided to break up each training session over a few days and some only participated in one or more sessions but fewer than five per week. A high percentage of participants dropped out of the study for the same reasons. This may have a great impact on treatment integrity, so future studies should examine how violating the training protocol influences the effect of the treatment. Moreover, future studies should consider using the shortened version of the Cogmed[©] intervention as the shortened version may be more practical for this population. Since most research on the Cogmed[©] intervention has used the original version of the training program, the effectiveness of the shortened version should be investigated.

The small sample size in this pilot study is another limitation as it has reduced the statistical power (e.g., increased the likelihood of Type II errors), which limits both the generalization and confidence of the findings. Thus, it is important to increase the sample sizes in future studies to generate greater statistical power to support the findings. Another issue is the lack of correction on Type I errors in this pilot study, so the positive findings presented in this paper should be interpreted cautiously. Future studies should replicate this study with a larger sample to further

examine the positive findings. Moreover, this study explores the measures at the composite score level instead of the scale score level in order to gain an overview of the effect of treatment.

Future studies may examine the scale score of each measure (e.g., BRIEF and BASC-2) to gain a better understanding of the effect of treatment on specific areas of cognitive, learning, emotional and behavioural functioning. Lastly, this study is limited to investigating the independent moderator effect (main effect) on the intervention due to the small sample size. As suggested in McLachlan et al. (2017), there is an interaction moderator effect instead of a main effect. Future studies should explore how the interaction moderator effect influences the treatment effect.

Conclusion

These preliminary findings suggest that the Cogmed[®] intervention is an effective intervention for both children with PAE and typically developing children to improve working memory and attentional control. There are potential gains in other EFs, learning, behavioural, and adaptive measures after intervention, suggesting a potential transfer effect to non-targeted functioning. There are also potential long-term treatment effects. The promising findings may spark clinicians' interest in using the Cogmed[®] intervention as an evidence-based intervention for PAE. It may also inspire researchers to explore effective interventions geared toward the unique needs of individuals with PAE experience. First, the Cogmed[®] intervention can bridge the service gap for families who live in rural settings that have limited resources. Second, this intervention can be introduced in the school setting as the findings suggest that children with PAE had improved cognitive (e.g., working memory and attentional control), adaptive, and learning (e.g., mathematic achievement) functioning as a result. Since the cognitive deficits in individuals with PAE often lead to adverse outcomes such as academic failure, poor adaptive functioning, impaired social skills, and mental health disorders, it is important to offer the

Cogmed[®] intervention as part of an early intervention in school to facilitate proactive support and minimize possible adverse outcomes and societal costs. Lastly, researchers should continue to investigate effective interventions that specifically target the problem areas identified in the PAE group in this study (e.g., visuospatial working memory and inhibition) as these areas may need more intensive training or a longer training time in order for treatment to be effective.

Although this pilot study suggests that the Cogmed[®] intervention is a potential effective treatment option for children with PAE, clinicians need to be cautious when interpreting the findings, and future studies are needed to strengthen the findings.

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Table 4.1. *Demographic data for the PAE (PAE) and comparison (COM) groups*

Demographic variables	PAE (<i>n</i> =20)	COM (<i>n</i> =18)	df	t-value (2-tailed)
Age [<i>M</i> (<i>SD</i>)]	9.20 (2.59)	6.78 (2.67)	36	2.834**
SES [<i>M</i> (<i>SD</i>)]	43.73 (10.20)	49.97 (8.94)	36	1.990
GIQ [<i>M</i> (<i>SD</i>)]	88.55 (13.83)	109.89 (11.00)	36	5.224**
Verbal IQ	86.10 (15.45)	104.89 (10.37)	36	4.350**
Visuospatial IQ	94.60 (11.36)	112.67 (13.03)	36	4.567**
Gender				
Female (<i>n</i>)	9	9		
Male (<i>n</i>)	11	9		
Ethnicity				
Caucasian (<i>n</i>)	4	12		
Aboriginal (<i>n</i>)	16	1		
Others/ Unidentified (<i>n</i>)	0	5		
Current living arrangement				
Biological Parents (<i>n</i>)	0	16		
Adaptive Parents (<i>n</i>)	15	1		
Foster Parents (<i>n</i>)	4	0		
Others/Unidentified (<i>n</i>)	1	1		
Number of Placements [<i>M</i> (<i>SD</i>)]	3.50 (2.71)	0.29 (1.21)		

* Statistically significant at $p < 0.05$ ** Statistically significant at $p < 0.01$

Table 4.2. *Number of areas of working memory (WM) improvement in both PAE (PAE) and comparison (COM) groups measured by AWMA between pre- and post-interventions*

Number of areas of WM improvements	PAE	COM
0	<i>n=7</i>	<i>n=4</i>
1	<i>n=7</i>	<i>n=6</i>
2	<i>n=5</i>	<i>n=7</i>
3	<i>n=1</i>	<i>n=1</i>
4	<i>n=0</i>	<i>n=0</i>

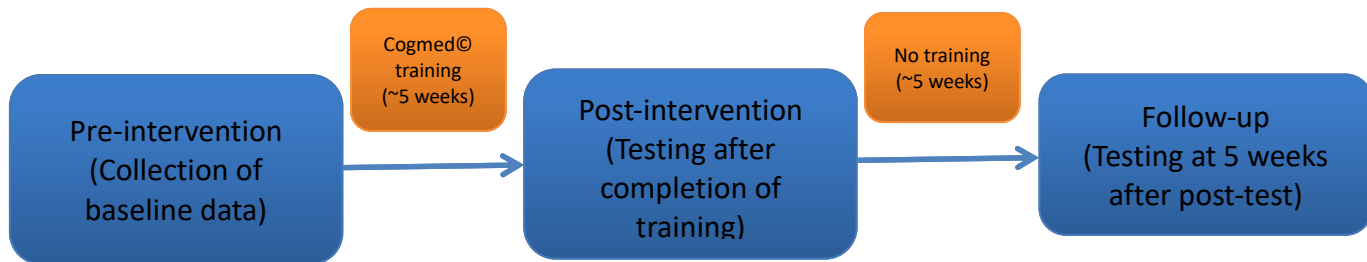


Figure 4.1. Study design

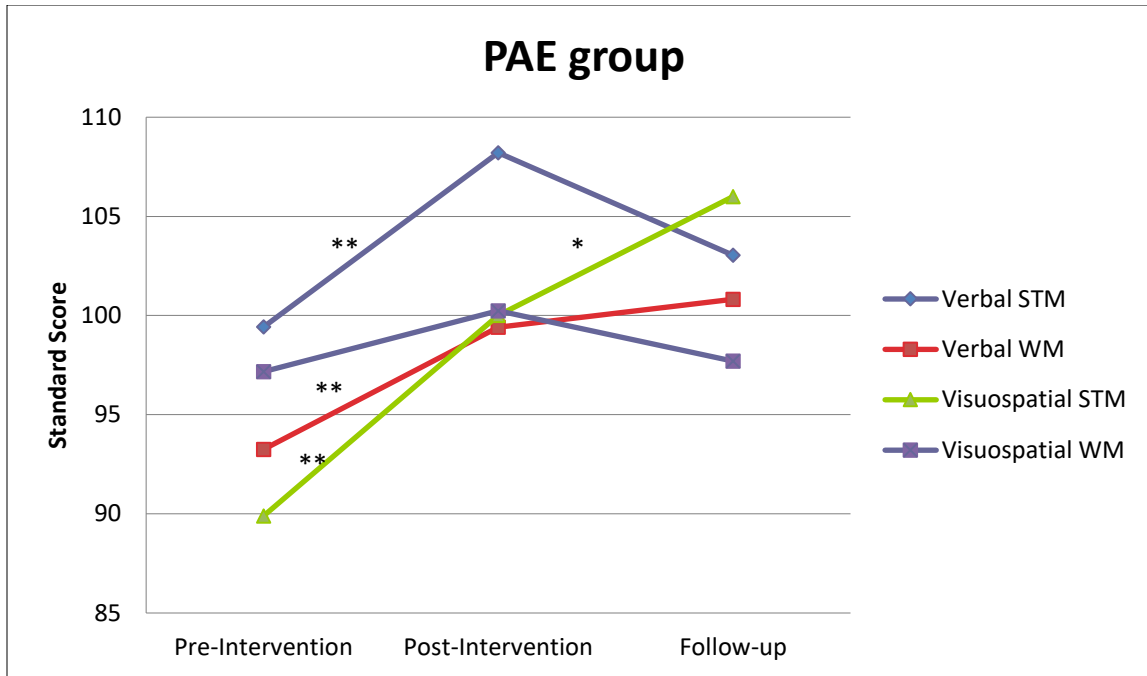


Figure 4.2. Working memory mean scores of PAE group measured by AWMA at three time points. * Statistically significant improvement at $p < .05$. ** Statistically significant improvement at $p < .01$.

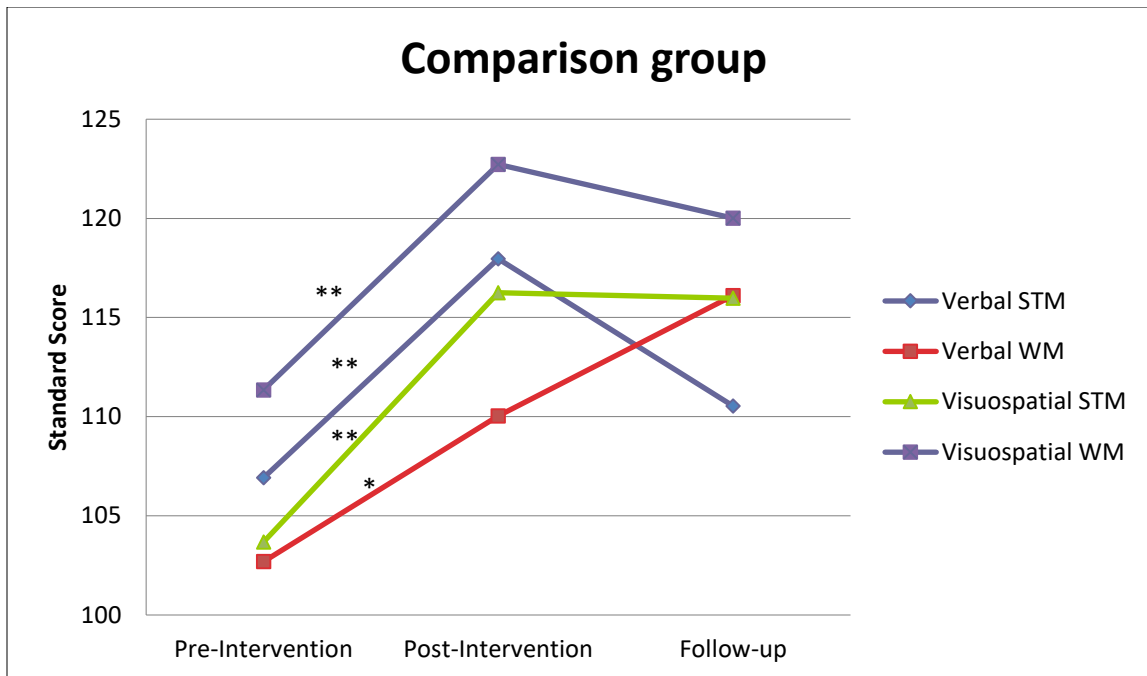


Figure 4.3. Working memory mean scores of comparison group measured by AWMA at three time points. * Statistically significant improvement at $p < .05$. ** Statistically significant improvement at $p < .01$.

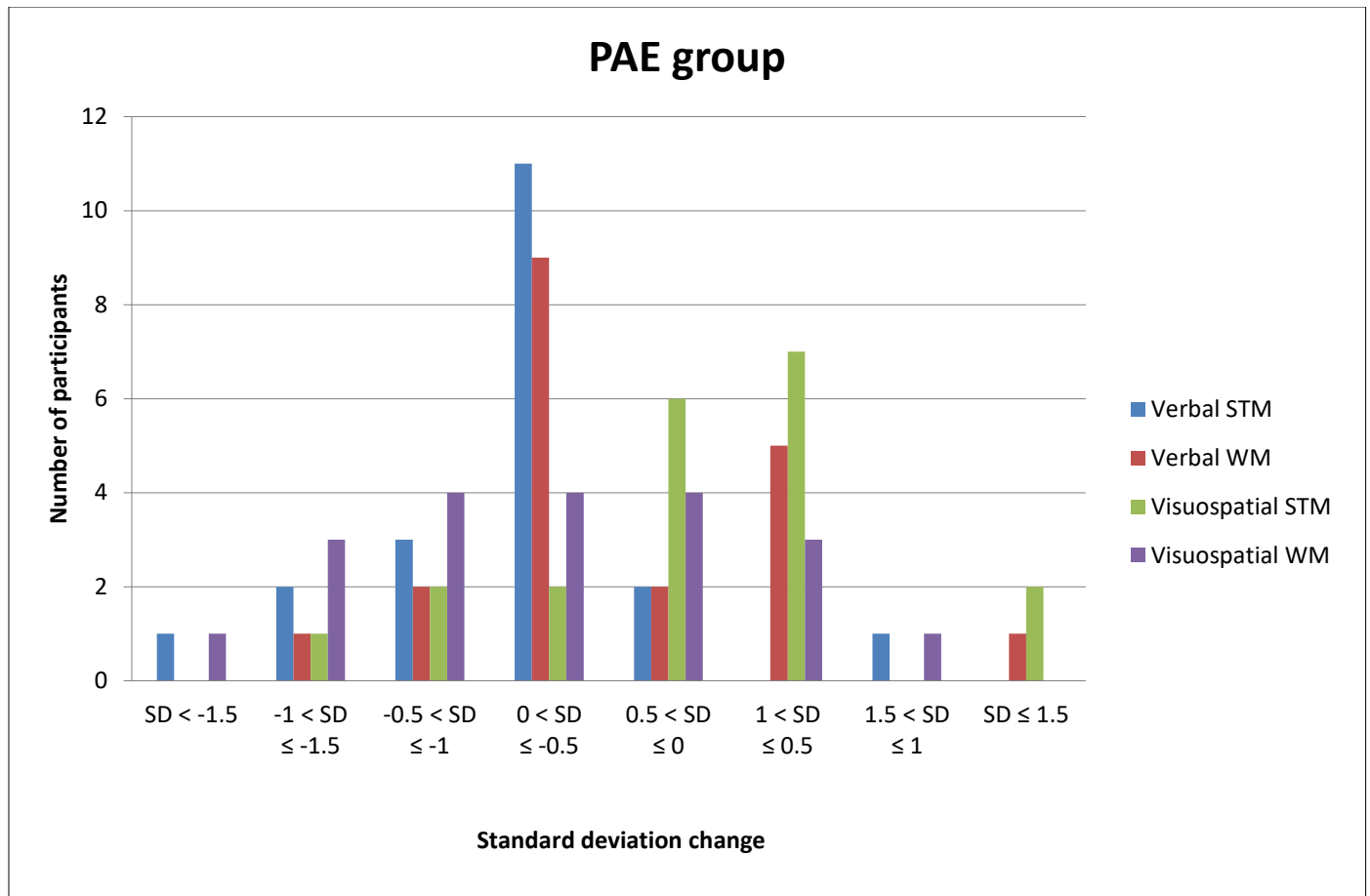


Figure 4.4. Individual improvements in PAE group measured by AWMA between pre- and post-interventions

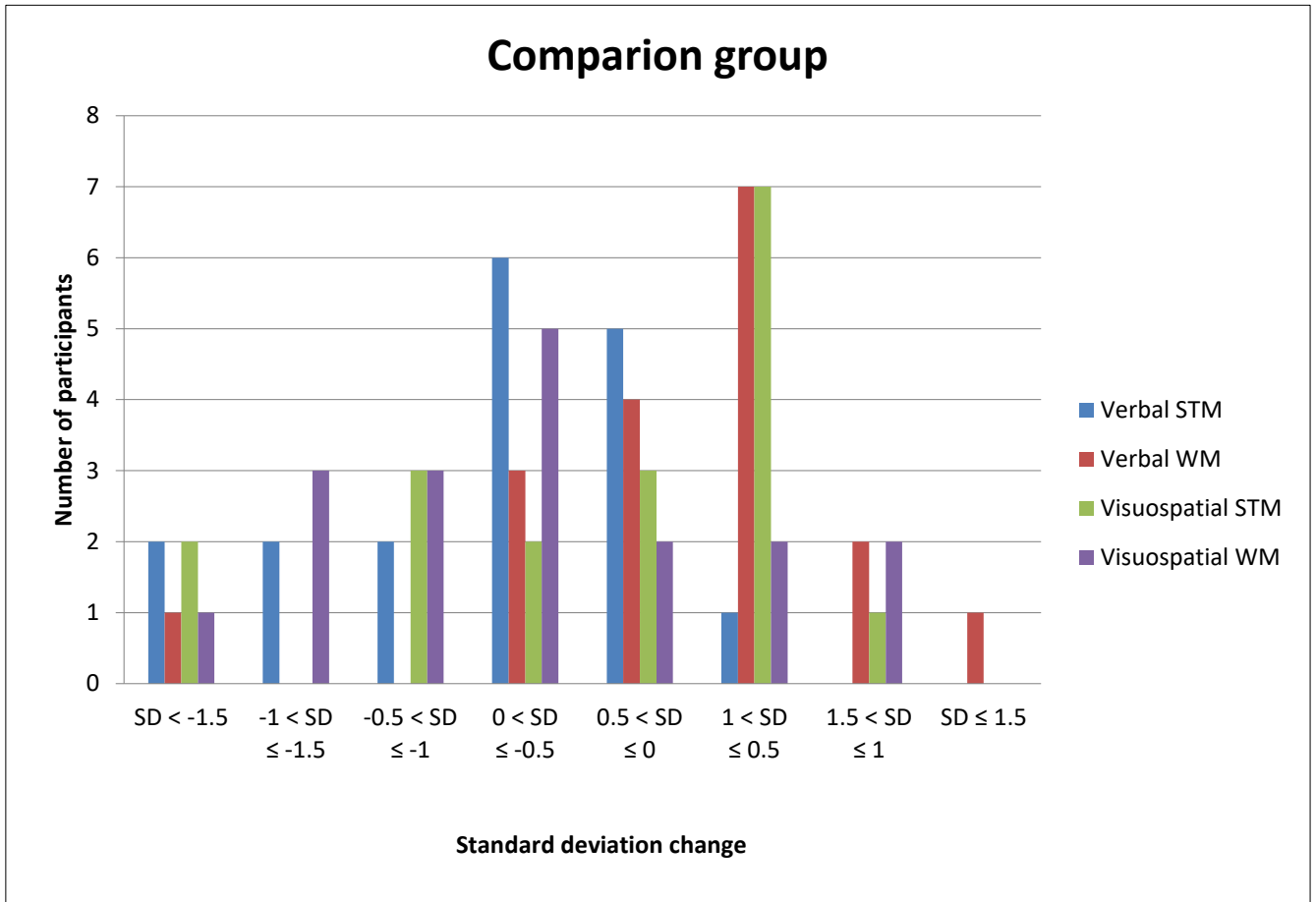


Figure 4.5. Individual improvements in comparison group measured by AWMA between pre- and post-interventions

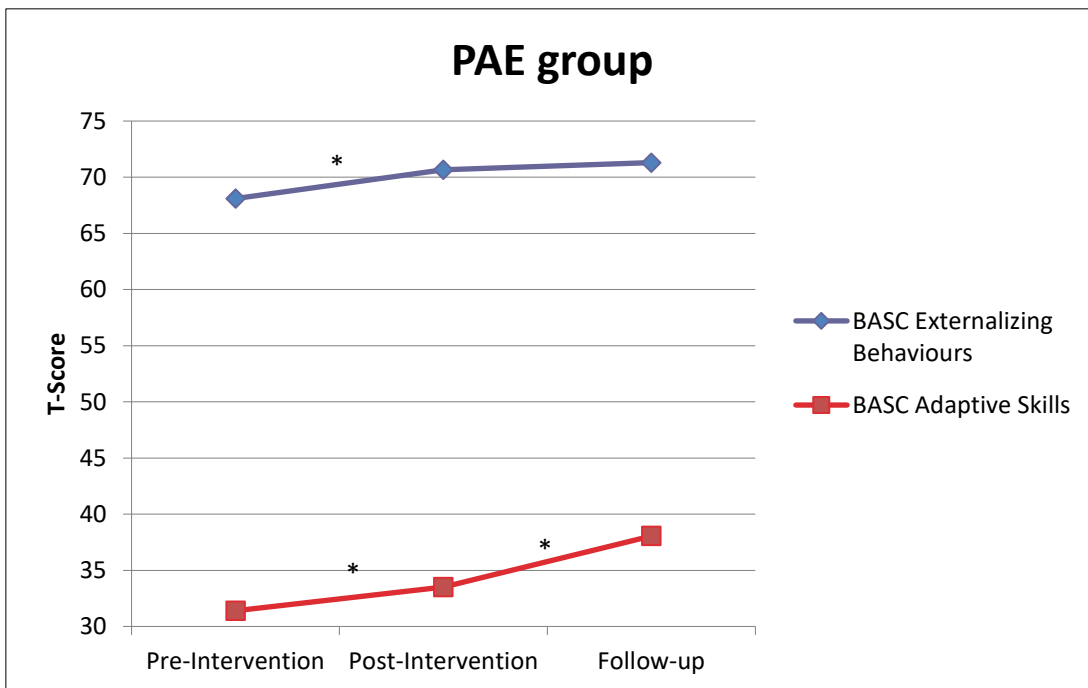
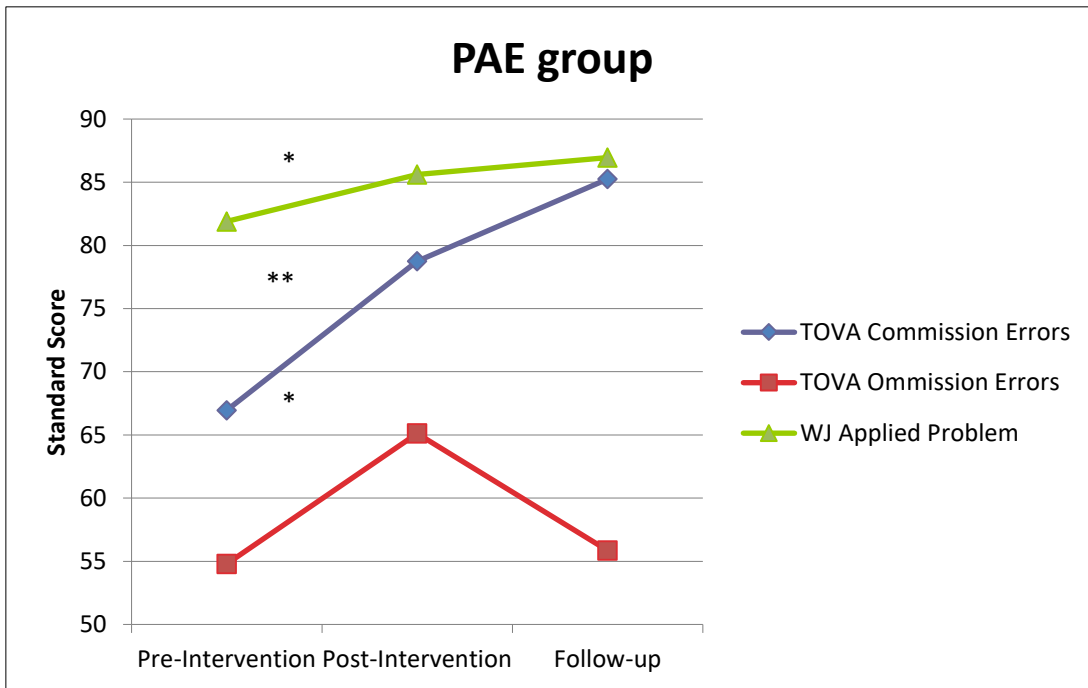


Figure 4.6. Mean scores of attentional control, externalizing behaviours, and adaptive skills measures in PAE group at three time points. * Statistically significant improvement at $p < .05$. ** Statistically significant improvement at $p < .01$.

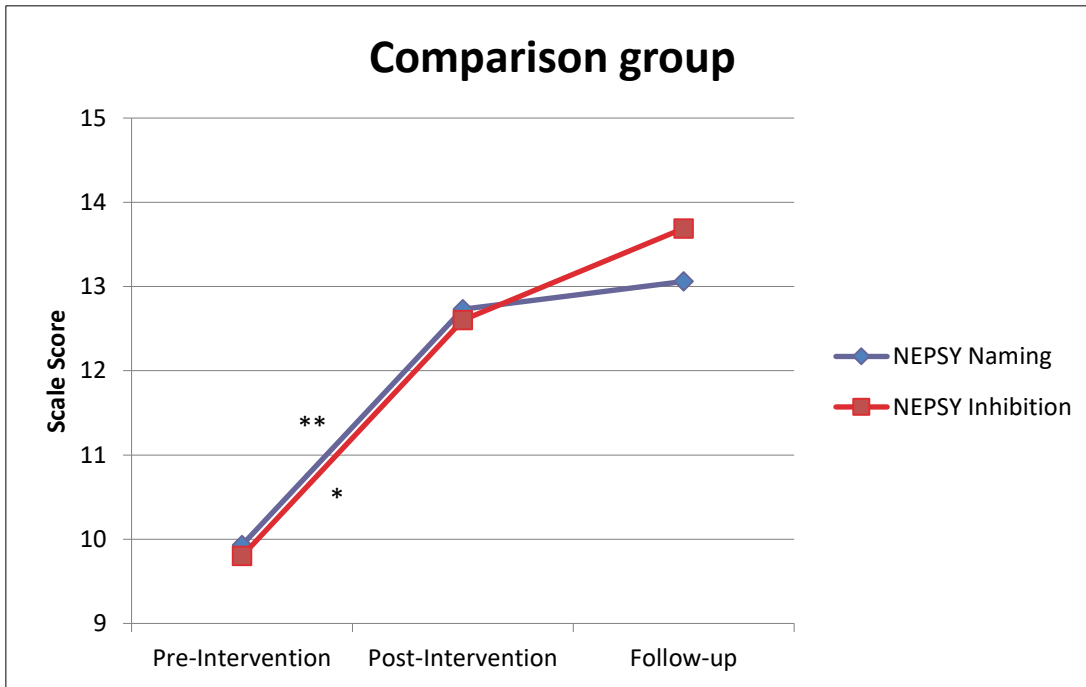
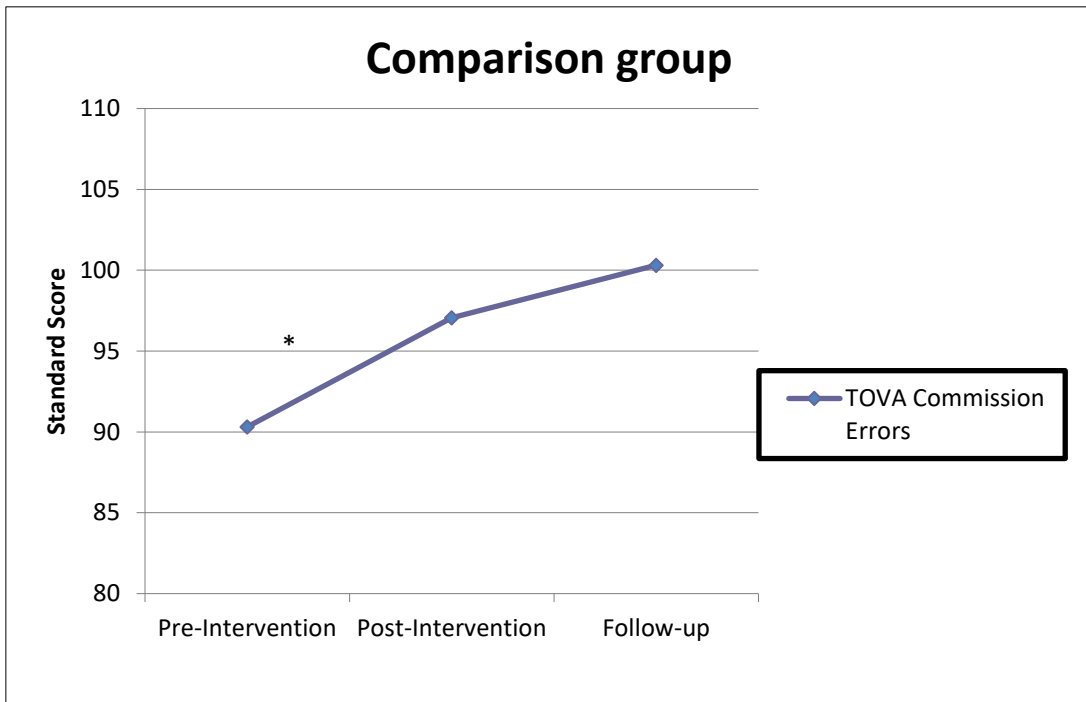


Figure 4.7. Mean scores of attentional and inhibitory controls measures in comparison group at three time points. * Statistically significant improvement at $p < .05$. ** Statistically significant improvement at $p < .01$.

Chapter 5: Conclusion

In past decades, researchers and clinicians made great efforts to gain a better understanding of and to support individuals with FASD. Recently, barriers were identified that contribute to the development of adverse outcomes. These barriers include lack of knowledge, delayed diagnosis, and availability of services (Petrenko et al., 2013). Noticing these gaps in the field, I decided to 1) examine the relative strengths and weaknesses within the neurobehavioural profile of local individuals with Prenatal Alcohol Exposure (PAE) in Alberta, Canada (Study 1) and 2) investigate the short- and long-term effects of the Cogmed[©] intervention for children with PAE (Study 2).

In Study 1, I found that children with PAE demonstrated strengths in working memory, auditory attention, and cognitive fluency, as assessed using objective measures in a clinical setting. Unfortunately, these areas of strengths were not observed by their caregivers in their daily lives. Children with PAE also experience more visual attention problems, mental health issues (e.g., inattention and hyperactivity), learning problems (e.g., with reading, writing, and math), and social and communication problems. Consequently, it is important to provide proper services for these children and their families. Such services include counselling or mental health workshops for the children and caregivers, academic supports in school (e.g., tutoring or strategy learning), social skills training, and speech therapy or functional communication training. The findings in this study can help service providers to understand the unique characteristics of the local population. Given the complexity of PAE, a neurobehavioural profile allows service providers to tailor strength-based interventions to the needs of affected individuals.

Study 1 also sheds light on EF assessment tools. I observed that when caregivers

reported that children with PAE had significant learning problems, those children tended to have impairments in all three of the fundamental areas of EF (e.g., inhibition, working memory, and cognitive flexibility) and lower math achievement scores. This suggests that learning problems observed in the classroom may be an indication of EF impairments. In these cases, the teacher or caregiver may consider offering effective interventions to target EF deficits toward children at an early age to reduce the risk of adverse outcomes. Moreover, I found that objective and subjective EF measures do not always align, so future research should investigate the way in which different EF measures come together, which can help to streamline the assessment tools and improve the current diagnostic process. Lastly, I found that CONNERS-3 has the potential to be an effective screener for PAE. Future research should investigate the key characteristics identified by CONNERS-3 and use that information to further develop a specific screener for individuals with FASD, which may bring us one step closer to standard assessment tools for FASD.

In Study 2, I found that the Cogmed[®] intervention is an effective working memory intervention, with promising short-term effects for improving working memory and attentional control for children with PAE and typically developing children. As the Cogmed[®] intervention effectively addresses the core EF deficits of individuals with PAE (e.g., working memory), there are potential gains in other non-targeted EF and behavioural, social and academic functioning after intervention. Moreover, there is also an indicator for the potential maintenance of gain (long-term effect). Since there are also improvements in the non-targeted measures, future neuro-imaging studies should be conducted, as these will help clarify the neurobiological causes underlying these changes and why and how the Cogmed[®] intervention is effective.

The findings indicate that the Cogmed[®] intervention can be proactive for individuals with PAE, thus minimizing possible adverse outcomes and societal costs in the long run. Future research could explore generalizing the intervention to school settings. Petrenko and Davis (2017) suggested that interventions are most effective when coordinated across different settings such as home and schools, so it is important to allow opportunities for generalization. It would be ideal for schools to incorporate this training for students with PAE to improve their ability to focus in class and meet academic demands. Moreover, the Cogmed[®] intervention can be an option for students who are home-schooled or live in rural areas with limited resources. Although this is a pilot study, the positive findings may fuel future research to further investigate the effectiveness of this intervention and its generalizing effect with a larger sample size.

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