



## **NDIA Planning Project**

**Critical Review of the Literature  
FETAL ALCOHOL SPECTRUM DISORDERS**

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This critical review was commissioned by the National Disability Insurance Agency and undertaken by the Telethon Kids Institute in 2015.

The authors wish to advise that a revision has been made to the Fetal Alcohol Spectrum Disorder diagnostic categories as detailed on page 10 of this report, to fit with the revised Australian Diagnostic Criteria, April 2016. There may still be reference made to Fetal Alcohol Spectrum Disorder (FAS), partial Fetal Alcohol Spectrum disorder (p-FAS) and Neurodevelopmental Disorder Alcohol Exposed (NDAE) throughout the report – 14 June 2016

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## Glossary of Terms

### **Academic Achievement**

The skills in core academic areas (reading, maths, and written language) including school readiness, attendance, engagement, and performance.

### **Adaptive Functioning**

The skills required to carry out daily activities necessary for personal and social self-sufficiency, or the ability to respond successfully to everyday demands. It incorporates function and participation in activities of daily living, social participation and work or school performance.

### **Attachment**

Attachment is an emotional tie which develops between one person and another. In infancy this relationship is formed with at least one primary carer and is based on a continuing relationship which is secure, nurturing and responsive to the infant's needs.

### **Attention**

The processing capacity for selective, focused, sustained, and flexible attention, as seen, for example, in the behaviours of concentration and impulsivity.

### **Cognition**

The set of all mental abilities and processes related to knowledge: attention, memory and working memory, judgment and evaluation, reasoning and "computation", problem solving and decision making, comprehension and production of language, etc. An important facet in this aspect of mental functioning is comparison of verbal and nonverbal thinking abilities.

### **Communication**

This refers to the ability to convey information from one person to another. Communication is conveyed through words and actions. It involves the ability to express one's self and understand others through words or actions.

### **Early childhood intervention**

The process of providing specialised support and services to infants and young children with developmental delays or disabilities (aged from birth through to school age) and their families in order to promote development, wellbeing and community participation. Differs from early intervention, defined below.

### **Early intervention**

Early intervention (in the context of this report) refers to a range of services and supports for different age groups across the life course, at the earliest possible time, once functional impairments are recognised or a diagnosis related to FASD is made.

### **Executive Functioning**

An umbrella term referring to a group of higher-level abilities that help guide and control behaviour, encompassing intellectual abilities, activity, attention and information processing, language, visual processing, learning and memory, number processing and social cognition.

### **Fetal Alcohol Spectrum Disorder (FASD)**

Is the term used to describe various conditions or diagnoses resulting from prenatal alcohol exposure.

### **Intellectual functioning**

Intellectual functioning is determined by many factors. However, a primary source of this capacity is mental ability or "intelligence." Intelligence refers to the ability to reason, plan, think, and communicate. These abilities allow us to solve problems, to learn, and to use good judgment. One measure of intelligence is called the intelligence quotient, or IQ.

### **Language**

Language encompasses all aspects of expressive and receptive language. It includes the ability to integrate the specific language skills such as grammar and sentence structure and the use of words to convey meaning.

### **Language disorders**

Those disorders which affect the ability to understand spoken language or the ability to express oneself through speech.

### **Memory**

Our ability to encode, store, retain and subsequently recall information and past experiences in the human brain. It can be thought of in general terms as the use of past experience to affect or influence current behaviour.

- **Explicit memory**  
Explicit or declarative memory is the conscious recall of knowledge about people, places, and things.
- **Implicit memory**  
Those tasks that do not require conscious memory but instead rely on unconscious recall of a previously performed task.

### **Motor**

Motor functioning encompasses general abilities to use and coordinate large and small muscles. Gross motor skills include walking, running, hopping and climbing. Fine motor skills include hand writing and eating. Eye hand coordination refers to the ability to coordinate vision with movement.

### **Personal independence at home or in community settings**

This refers to the ability to take care of yourself. Some examples are bathing, dressing, and eating. It also includes the ability to safely complete day-to-day tasks without guidance. There are also routine activities performed in the community such as grocery shopping.

### **School or work functioning**

This refers to the ability to conform to the social standards at work or school. It includes the ability to learn new knowledge, skills, and abilities. People must apply this information in a practical, adaptive manner; without excessive direction or guidance.

### **Secondary effects of FASD**

Difficulties that a person is not born with, but which are linked to the primary impairments of FASD. Secondary effects can be reduced and prevented through improved community and professional understanding, as well as adequate and appropriate interventions, provided as required across the life course. Secondary effects are sometimes referred to as secondary disabilities.

### **Self-Regulation**

This refers to how efficiently and effectively an individual deals with a stressor and then recovers. A lack of ability to self-regulate behaviour can result in outcomes including impulsivity and a lack of social inhibition.

### **Sensory Processing**

Relates to how information is received through the senses in order to function smoothly in daily life.

### **Sleep disorders**

There are a range of sleep disorders, but the most common ones are sleep apnoea, difficulties getting to sleep and staying asleep, sleep-walking and nightmares/night terrors. Sleep difficulties may occur as a result of self-regulation and behavioural difficulties, or because of physiological conditions.

### **Social communication**

The ability to communicate effectively and appropriately in a variety of social situations with both peers and adults, including the ability to relay verbal information coherently and cohesively.

### **Substance use disorders**

Substance use disorders happen when the recurrent use of alcohol and/or drugs causes clinically and functionally significant impairments, such as health problems, disability, and failure to meet major responsibilities at work, school, or home.

### **Zone of Proximal Development**

Is the gap between the child's developmental level, as indexed by independent problem solving, and the child's potential developmental level, as determined by problem solving under adult guidance or with capable peer collaboration.

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## Executive Summary

### *Overview:*

Fetal Alcohol Spectrum Disorders (FASD) is a diagnostic term relating to permanent brain damage and functional impairment resulting from prenatal alcohol exposure (PAE). Revised Australian diagnostic entity changes identify two sub-categories within FASD: FASD with three sentinel facial features, similar to the previous diagnostic category of Fetal Alcohol Syndrome (FAS); and FASD with less than 3 sentinel facial features, which encompasses the previous diagnostic categories of Partial Fetal Alcohol Syndrome (p-FAS) and Neurodevelopmental Disorder-Alcohol Exposed (NDAE)}. The unifying feature of a diagnosis of FASD is significant central nervous system (CNS) abnormality or impairment in three or more 'structural or functional domains'. Impairments and interventions in this review relate to all sub-categories of FASD diagnosis.

The National Disability Insurance Agency (NDIA) has identified FASD as an important category of disability for consideration within the National Disability Insurance Scheme (NDIS). NDIA contracted Telethon Kids Institute to conduct a critical review of the available published and unpublished literature relating to impairment and interventions for FASD. A structured review and synthesis of literature has been provided, with oversight and input from an Expert Review Panel of leaders in the field of FASD and Disability.

This review is set out in two sections:

- The first section summarises (a) the principles relating to assessment, diagnosis and planning interventions for FASD, (b) a summary of the evidence on functional domains that are impaired in people with FASD across the life course, and (c) a summary of evidence for interventions to reduce impacts of functional impairment and improve overall quality of life.
- The second section documents the methods used to identify and review the literature, detailed reporting of the findings, and appendices.

The purpose of the review is to describe the types of disability supports, services and interventions that individuals with FASD require across their life course. This information will inform an approach to assessment and documenting impairment associated with FASD that will enable support and intervention planning within the NDIS.

### *Key findings:*

#### Principles relating to planning supports and services:

Comprehensive services for individuals with FASD require the involvement of the individual, their family members/caregivers and multiple systems of care. Given the variability of functional impairments seen in FASD, assessment to determine a neurodevelopmental profile specific to the child or young person is necessary for intervention planning.

The following principles identified in the literature are relevant when planning access to services and supports:

- A multidisciplinary approach to assessment and support which may include: medical specialists, occupational therapy, speech and language therapy, behaviour therapy, psychology/neuropsychology, psychiatry, physiotherapy, social support and individualised education plans. Interventions change over time depending on the number, type and severity of FASD associated problems.

- Support for a *coordinated family + school + therapy partnership* which facilitates access to multiple interventions, especially when these are provided in different settings and involve multiple agencies.
- Family centred ‘wrap-around’ care to strengthen and build capacity for families to support children and to support the person into adult life through maximising participation in everyday life.
- Building on caregiver and child/young person strengths and provide emotional support for caregivers that reduces FASD-related stigma or ‘blame/shame’.
- Reframing FASD for caregivers and professionals involved with the child/young person, e.g. educating that behavioural difficulties are ‘brain-based’. Facilitate respite and self-care opportunities for carers, to support their own mental health, and to address substance misuse problems where relevant.
- Providing risk assessment and monitoring for children within the caregiving setting.
- Planning for the transition to adulthood for individuals with FASD, viewed as a transition to ‘interdependence’ rather than necessarily a transition to independence.
- Ensuring cultural security for Indigenous and ethnically diverse communities: embedding programs and resources in community controlled health organisations where possible and employing Indigenous and culturally diverse people in intervention teams.

#### Impairment and Interventions across the life course:

FASD needs to be understood as a permanent disability with predominantly behavioural and functional impairments. People with FASD have significant brain based differences that give rise to disability because of functional impairments in a variety of areas, including cognition, memory, language, attention and executive functioning. Because the brain disability is permanent, FASD is not ‘outgrown’, and primary impairments are not ‘cured’. An approach to management of FASD over the life course that is multifaceted, cross disciplinary and collaborative will enhance participation and quality of life for individuals, families and communities.

The profiles of functional impairment vary between individuals with FASD. There are two sub-categories within FASD: FASD with three sentinel facial features (similar to the previous diagnostic category of Fetal Alcohol Syndrome); and FASD with less than three sentinel facial features (which encompasses the previous diagnostic categories of Partial Fetal Alcohol Syndrome and Neurodevelopmental Disorder-Alcohol Exposed). A person with FASD with less than three sentinel facial features may have a similar or greater degree of impairment as a person with FASD with three sentinel facial features, and all must be considered for disability support and intervention planning. The emergence of specific domains of functional impairment across the life course are summarised in Figure 1.

Importantly for the NDIA, the utilisation of IQ scores as a determinant of eligibility for services and supports cannot fully respond to the unique needs of persons with FASD. Functional need must determine access to support for individuals with FASD.

Functional impairment may become more apparent at key transition times: during infancy, on entering formal education, when transitioning to secondary education and during adolescence and when leaving education and seeking employment. Providing intervention early, and at adequate intensity and frequency, improves neurodevelopmental and functional outcomes and decreases social and mental health problems later in life. Consistent interventions are required across the home, school, and

community setting. Determining the appropriate intervention is dependent on a coordinated approach to assessment and diagnosis. A process of referral ➤ assessment ➤ intervention ➤ review ➤ re-assessment, with ongoing case coordination is recommended.

#### Principles relating to Interventions:

The following principles identified in the literature are relevant when planning interventions:

- Coordination and case management based on a key worker model allows an interagency approach and facilitates communication while simplifying the service milieu for families.
- A collaborative partnership approach between the family and support services including school, work and community is needed to ensure consistency in the individual's environment.
- Interventions should be integrated into an existing education or therapy structure (e.g. local early childhood intervention services, school based education services, universal services and everyday environments).
- Strengths based approaches build therapy interventions around an individual's strengths as identified in the process of assessment and diagnosis.
- Early intervention that includes supporting parent/child attachment and introducing concepts of self-regulation are universally required and effective. Providing explicit interventions to improve self-regulation and attention/executive functioning are effective before and throughout the school years.
- Intervention design incorporates that individuals with FASD need to learn and build skills that will help them regulate their emotions and behaviours, as well as be provided with environmental accommodations that increase the likelihood of adaptive behaviours.
- Specific caregiver positive behaviour support training around the neuropsychological impairment that co-occurs with the challenging behaviour of children with FASD.
- Collaborative problem solving and stress management training and support for caregivers, which should be implemented concurrently with interventions for the child.
- Individualised interventions
  - Understanding the child/young person's cognitive-behavioural profile when designing and intervention program. Strategies include: careful presentation of information (multimodal and repeated) and 'hands on' learning.
  - Use strategies from the child/young person's zone of proximal development (ZPD) by providing activities that are just beyond the independent level capable of being achieved by the child/young person.
- Children/young people with FASD may require explicit instruction to acquire skills that typically developing children would learn through observation or abstraction.
- Promoting the concept of scaffolding and structure by: providing environmental accommodations (also referred to as the '*external brain*') to support executive functioning and emotional and behavioural control; and, emphasising the importance of structure through explicit teaching of functional routines to provide a scaffolding framework.
- Combining a 'domain general' approach (eg. Families Moving Forward Program or The Alert program<sup>®</sup>) with a 'domain specific' approach (eg. the MILE program) will be more effective than isolated intervention approaches.
- There is a need for mental health services, particularly for children older than three years of age with an increase in intensity of support towards adolescence and at key transition times.



- Promoting the use of informal support networks can be an intervention in itself for families raising a child with FASD.
- Considering the medical needs of children with FASD and the need for medical referrals and recommendations which may include psychiatric care, medication management, vision and/or hearing screening, neurological consultation or clinical management, ADHD evaluation or treatment, investigation of growth faltering/short stature assessment, hygiene sleep medicine, brain imaging, and genetic evaluation or consultation.

This review provides a detailed summary of current knowledge of impairments seen in FASD across the life course, and interventions and supports that have been shown, or are likely, to ameliorate the impact of these impairments. Based on the available literature and deliberation by an Expert Review Panel, guidance is provided on the level of intervention and support required for individuals with varying levels of impairment severity.

The authors expect that this review will provide an evidence base for the impairments seen in FASD to be considered within the NDIS, so that effective interventions and supports may be provided within an appropriate funding framework. As with other disabilities, outcomes for people with FASD can be improved through early diagnosis, early intervention, and a life course approach to improving function and participation.

The immediate policy implication for NDIA is for FASD to be embedded in the disability sphere as a permanent and lifelong disability that is amenable to intervention and supports to improve life outcomes for affected individuals.

## Section One

### 1. Introduction

#### 1.1 Aim of the critical review

This is a critical review of the available information, published and unpublished, on the known functional impairments and related interventions/supports across the life course for people living with Fetal Alcohol Spectrum Disorders (FASD). The review aims to inform the development of best practice guidelines for the NDIA.

#### 1.2 Background

The National Disability Insurance Agency (NDIA) was established by the Australian Government to implement the National Disability Insurance Scheme (NDIS). The NDIS is a universal funding mechanism to support people with a permanent and significant disability that affects their ability to take part in everyday activities.

The NDIA is a Commonwealth authority working with state and territory governments to deliver the Scheme to ensure improved support for people with disability, their family and carers. The NDIA identifies the services and supports that people with disabilities need to live their lives. These may help achieve goals in many aspects of a person's life, including independence, involvement in the community, education, employment and health and wellbeing. The NDIS gives people living with a disability more choice and control over how, when and where supports are provided and give certainty that people will receive the support they need over their lifetime. (In relation to children, the NDIS focuses on early intervention where getting early supports can improve functional outcomes and reduce the impact of disability on families).

#### 1.3 Fetal Alcohol Spectrum Disorders

Fetal Alcohol Spectrum Disorders (FASD) is a diagnostic term with features of significant functional impairment across multiple CNS domains. Please note the following changes have been made to the Australian diagnostic entities since the original publication of this report. A revised Canadian guideline on the diagnosis of FASD was published<sup>1</sup>, just as the Australian instrument was being finalised, and so the Australian FASD Diagnostic Instrument was reviewed and modifications made in April 2016<sup>2</sup>. Specifically, the Australian diagnostic instrument has adopted the concept that Fetal Alcohol Spectrum Disorder be used as a diagnostic term. For a diagnosis of FASD, an individual must have prenatal alcohol exposure and severe neurodevelopmental impairment in at least three of ten specified domains of central nervous system structure or function. The overarching diagnostic term of FASD simplifies the terminology and emphasises the primary importance of the severe neurodevelopmental impairment that results from an acquired brain injury caused by alcohol exposure before birth. The new revised changes identify two sub-categories within FASD: FASD with three sentinel facial features (similar to the previous diagnostic category of Fetal Alcohol Syndrome); and FASD with less than 3 sentinel facial features (which encompasses the previous diagnostic categories of Partial Fetal Alcohol Syndrome and Neurodevelopmental Disorder-Alcohol Exposed).

Previously the Australian FASD diagnostic guidelines recognised three specific diagnoses on the FASD spectrum: Fetal Alcohol Syndrome (FAS), partial FAS (pFAS), and Neurodevelopmental Disorder-Alcohol Exposed (ND-AE)<sup>3</sup>. FAS and pFAS require the presence of characteristic facial features and all three diagnoses require significant structural and/or functional CNS abnormalities in three or more domains. A person with FASD with less than 3 sentinel facial features, may have a similar or greater degree of

impairment as a person with three sentinel facial features, and sub-categories on the FASD spectrum must be considered for disability support and intervention planning. Severity of disability in FASD depends on which domains are affected and to what degree, not upon the sub-category within the FASD spectrum. FASD are permanent and lifelong.

FASD is the leading preventable cause of intellectual disability in the western world<sup>4</sup>. Children prenatally exposed to alcohol often have impaired intellectual functioning<sup>5 6 7</sup> and significant impairment in a range of other CNS domains. Studies from North America demonstrate a link between impaired cognitive and adaptive skills among individuals living with FASD with subsequent adverse life outcomes or secondary effects. These include poor educational outcomes, contact with the criminal justice system, mental illness, and drug and alcohol problems. There is growing concern about the impact of FASD on individuals, families and communities in Australia, and, the need for services and supports to improve quality of life for individuals living with the disorder.

#### **1.4 Diagnosis and Assessment**

Diagnosis of FASD is difficult, complex and important. Diagnosis involves excluding or considering differential diagnoses that may be genetic, environmental or traumatic in origin. Hence the diagnostic process is one of exclusion and requires extensive history taking to exclude other exposures and risk factors, and may include a range of investigations including blood tests and medical imaging. Early life trauma or neglect can lead to or compound behavioural and cognitive impairments, and acquired brain injury from trauma, infection or metabolic conditions may lead to a similar neurodevelopmental profile to that seen in FASD. Some diagnoses may co-occur with FASD such as Attention Deficit Hyperactivity Disorder (ADHD) or Autism or other mental health disorders. In infancy, diagnosis may involve a paediatrician or child development specialist documenting global developmental delay. Where global developmental delay is not documented, there can be subtle delays that require careful screening of prenatal alcohol exposure, physical signs and development. In the school years, diagnosis ideally involves a multidisciplinary team including a paediatrician, psychologist/neuropsychologist, occupational therapist, speech pathologist and physiotherapist. In adulthood, there is an increasing role for mental health providers and general practitioners to be involved in the diagnostic assessment and investigation process. Parents/carers and teachers also provide crucial information during the diagnostic process. Neurodevelopmental profiles of individuals may change over time or become more pronounced with transitions through school and into adolescence.

## **2. Prevalence**

### **2.1 FASD prevalence**

Internationally the estimated prevalence of FASD is between 2-5%<sup>8</sup>. There are pockets of higher prevalence in populations at risk for higher rates of alcohol use, including for children in foster care, youth in the justice system, and some remote disadvantaged communities<sup>9 10</sup>. In Australia most estimates of FASD prevalence underestimate the problem due to limited FASD screening or diagnosis, with under-ascertainment being compounded by limited knowledge about FASD among health professionals. The only national study of the birth prevalence of FAS in Australia, utilising monthly reporting by paediatricians, reports a birth prevalence of 0.06 per 1000 live births, with significantly higher rates in the Indigenous population<sup>11</sup>. A second national surveillance study is being conducted in 2015. The only population-based FASD prevalence data from Australia are from a community-led active case ascertainment study in remote communities of Western Australia. A prevalence of FAS/pFAS of

120.4 per 1000 has been published<sup>12</sup>. Unpublished data from this study indicate that in this population approximately one in five children have FASD.

# MAP OF FASD IMPAIRMENTS & INDICATIVE ASSESSMENTS ACROSS THE LIFE COURSE

Emerging impairments affect ability to participate in activities - activity limitations lead to participation restriction (WHO - ICF)

Each individual affected by FASD has a unique neurocognitive profile determining their degree of executive and adaptive functioning impairment; requiring assessment to determine the types of interventions required to improve activity participation



Figure 1: The FASD Map

### 3. Conceptual Overview

The CNS functional impairments most commonly associated with FASD, the ages at which these are likely to emerge during the life course and the key assessments used to establish how severely an individual may be affected are detailed in the critical review. These are summarised in Figure 1 - The Map of FASD Impairments across the life course (Figure 1 – The FASD Map).

Key transitional times are evident for individuals when they will potentially require more intensive interventions. These are when children first enter formal education through to grade three, then when moving into secondary education; during adolescence and again when leaving education and seeking employment.

As neurodevelopmental profiles vary between individuals with FASD, each affected individual requires a range of assessments at transitional times to determine the extent of their functional cognitive and behavioural impairments, ideally leading to referral to appropriate interventions.

Support may be consistently required to assist with daily living, or an individual may need support to enable them to address their functional impairments by learning skills that help them with daily living. The challenge of living with brain-based functional deficits remains across life. With improved understanding of the impacts of FASD, interventions continue to be identified, implemented and evaluated.

To demonstrate the complexity of assessing and supporting an individual affected by FASD to achieve their potential through access to age appropriate services and supports, three Case Profiles are provided. These case profiles consider a young child, a school aged child and an adolescent.

#### Case Profiles

Each case profile is structured to include: (1) information on the family/caregiver context, (2) history of prenatal alcohol exposure, (3) the process of referral for assessment, (4) the diagnostic process and specific impairments identified, and (4) the approach to therapy/family support.

#### Case Profile 1 – Young child

Charles: 22 months

**History:** Charles' parents are busy professionals, both working long hours with a moderate degree of tension in their relationship. His father uses recreational drugs to maintain a high work output and long hours, and his mother self-medicates feelings of anxiety by drinking alcohol on most days. Her usual consumption is half a bottle of wine in the evenings after work, with higher quantities on weekends. Their pregnancy with Charles was unplanned, and pregnancy was not recognised until 11 weeks gestation. His mother cut back her heavier weekend drinking once realising she was pregnant, however continued to drink two to three glasses of wine on occasion throughout pregnancy.

Charles was born at term with no birth complications. He passed his neonatal screening testing. His neonatal period was remarkable for him being irritable with a poor sleep/wake cycle, and slow to establish breastfeeding.

**Screening and primary care:** Compared with other infants, Charles was noted by his mother to be slow to reach his milestones at the 12-18 month period. His mother raised this with a child health nurse at an immunisation clinic. After measuring Charles' growth and head circumference (all within normal limits) the nurse completed an Ages and Stages Questionnaire and found that Charles scored 'below cut-off' for communication, fine motor, and problem solving, 'close to cut off' in personal-social, and 'above the cut off' in gross motor. The child health nurse explained these screening results to Charles' mother, indicating that these findings would likely need referral to allied health specialists and also to a developmental paediatrician. The child health nurse referred Charles to a local GP with experience in child health and development. The GP took a history that included ascertaining that alcohol was used throughout pregnancy, conducted a child health check, and referred on to a speech language pathologist (SLP) and occupational therapist (OT), and to a private developmental paediatrician at the mother's request.

**Diagnostic assessment:** Charles' developmental assessment process was targeted and streamlined by the screening ASQ test. The child health nurse informed the SLP and OT that a developmental paediatric appointment was scheduled, and it was agreed among clinicians that a full assessment using the Griffiths Mental Developmental Scale (Extended Revised) or Bayley Scales of Infant development would be completed by the Paediatrician, SLP and OT together in a multidisciplinary clinic. This approach prevented the family receiving duplication of clinic appointments. Blood tests including genetic testing was requested by the paediatrician.

Assessment with the Griffiths Mental Developmental Scale (Extended Revised) showed that Charles was below the 3<sup>rd</sup> percentile of function for Language, Eye Hand Coordination, and Performance subscales. His physical examination revealed two of the three FAS facial features, however no growth impairment and a head circumference within normal limits. It was also evident that there were some difficulties with child-mother attachment. Charles' father had not been present during any clinical assessments, and Charles' mother indicated significant personal stress and anxiety. Blood test results were normal. A diagnosis of partial FAS was made and communicated to Charles' mother and father in a follow up clinic visit.

**Therapy approach:**

Support for maternal mental health and family functioning: The GP followed up with the parents, and arranged for individualised support for maternal mental health, and family counselling emphasising the need for a stable and structured home environment in light of Charles' developmental challenges.

A social worker with the local Child Development Service (CDS) agreed to coordinate the therapy services, and communicated between the Child Health Nurse, GP, CDS Allied Health team and private developmental paediatrician. The Child Health Nurse had received training in infant mental health intervention specialised for young children with prenatal alcohol exposure, and carried out this individualized intervention.

Specific therapy provided by SLP and OT: The SLP and OT were the mainstay of Charles' developmental therapy team and implemented a structured goal-focused program around language development, fine motor activities and behavioural management strategies including sleep hygiene.

**Developmental surveillance and review:** 6 monthly reviews with the developmental paediatrician were scheduled, and screening tests using the ASQ used to monitor developmental progress over time. A multidisciplinary case conference was coordinated at 12 month intervals and a repeat Griffiths assessment at 4 years of age (2 years after diagnosis) was planned.

## Case Profile 2 – School aged child

Ray: 7 years

**History:** Ray lives with his maternal grandmother in a regional mining town. Ray's mother was subject to domestic violence and significant emotional stress while pregnant with Ray, and was self-medicating with alcohol and marijuana during her pregnancy with him. She drank 10 or more standard drinks, 2-3 times per week throughout all trimesters of her pregnancy, and smoked marijuana at least once per week as well as cigarettes daily. Ray was born at 39 weeks gestation. His birth weight was 15<sup>th</sup> percentile, birth length 20<sup>th</sup> percentile, and birth head circumference 15<sup>th</sup> percentile. He required no significant resuscitation. He experienced neonatal abstinence syndrome and had a poor suck, requiring feeding support for his first 2 weeks of life. Maternal attachment, breastfeeding and weight gain was difficult and Ray required weekly follow up by community health nurses for his first months of life. He had a poorly developed sleep pattern. The family unit was very unstable and Ray was taken into care at 18 months of age, and has since lived with his maternal grandmother who cares for five other children. His father works interstate in mining, and been estranged from the family and has no contact with Ray. His mother has an itinerant lifestyle and longstanding mental health and drug and alcohol problems and has infrequent contact with Ray.

**Screening and primary care:** Ray's grandmother noticed that he was slower to start speaking than other children his age, however, he did not receive developmental screening at the time of receiving immunisations. Difficulties with staffing of child health nurse positions and infrequent visiting paediatric or allied health services meant that Ray 'fell through the gaps' of adequate primary care screening or developmental assessment. He was seen by a child health nurse for treatment of middle ear infections and on one occasion had audiometry testing indicating mild conductive hearing loss in both ears. On entry into the school system his teacher applied the Australian Early Development Index and he was found to be developmentally vulnerable in the domains of language and cognitive skills, communication skills and general knowledge.

His teacher referred him to visiting speech pathology and occupational therapy services who conducted standardised assessments that indicated significant impairment in expressive and receptive language (in Standard Australian English, the only language spoken at home), and fine motor control. General therapy strategies for language development and handwriting were provided for Ray in the school and home environment and he was reviewed for 1-2 hours each school term. He was not referred for assessment by a paediatrician until he was 6 years of age.

**Diagnostic assessment:** A visiting multidisciplinary team provided a comprehensive behavioural and neurodevelopmental assessment for Ray at age 7, almost one year after receiving a referral from the school that highlighted language and fine motor impairment, and behavioural problems including inattention and being fidgety. This assessment was conducted over two days, involved the grandparent and child, and included use of the Conners-3 Child Behaviour Rating Scales, the latest version of the Adaptive Behaviour Assessment System, the Behaviour Rating Inventory of Executive Function (BRIEF), the Short Sensory Profile, Lifeskills checklist, a cognitive assessment using the latest version of the Wechsler Intelligence Scale for Children-IV, additional tests of attention, memory and executive function, occupational therapy assessment using the BOT-2 test of motor proficiency, and language assessment using the CELF-4 screener and the latest version of the Children's Communication Checklist. A full paediatric examination was conducted. Ray's weight had been persistently on the 5<sup>th</sup> percentile since 5 years of age despite adequate nutritional intake. His height was on the 15<sup>th</sup> percentile and his head circumference on the 10<sup>th</sup> percentile. He had three out of three facial features of Fetal Alcohol Syndrome (Palpebral fissure length -2.3 Standard deviations



below the mean using the Hall charts, lip and philtrum rank 4 using the University of Washington Lip-Philtrum guide).

Ray was found to have significant impairment in the following domains: expressive and receptive language and higher language function; executive functioning; attention deficit/hyperactivity (inattentive/impulsive); sensory processing; fine motor control. His cognitive function and memory were in the low average range.

Blood tests were conducted for genetic and endocrine (hormonal) conditions that might cause Ray's developmental delay, and all were normal.

Ray was diagnosed with Fetal Alcohol Syndrome and a parent-centred therapy report was provided to the family, school and Department for Child Protection.

***Therapy approach:***

Support for maternal mental health and family functioning: The Department for Child Protection were informed of the diagnosis and took a proactive role in coordinating carer support and respite care for Ray's grandmother. His mother was also informed and her own case-worker provided education about FASD and family planning, with the intention to prevent further pregnancies being alcohol exposed. The social worker had received training in the Families Moving Forward Program and introduced this intervention to Ray's grandmother during regular sessions with the family. The social worker also talked with Ray's mother when appropriate. Ray's grandmother found therapy very helpful, learning how to manage Ray's behaviour, meet her own needs, feel more effective as a carer and to better advocate for Ray.

Specific therapy provided by SLP and OT and school: The formal documentation of Ray's neurodevelopmental delay/disability enabled his carer to access NDIS funding for additional speech language therapy, and occupational therapy input. This involved intensive speech/language interventions with a consistent message across the home and school environment. At school an Individual Education Plan was developed and implemented.

An occupational therapist provided information about sensory processing and explicit training of Ray, and his teacher and carer in The Alert program® was provided. This provided intervention strategies to improve Ray's self-regulation and ability to attend and learn.

***Developmental surveillance and review:*** Ray was reviewed regularly by the visiting paediatric service and for monitoring of his ADHD symptoms. These were found to improve with carer support and participation in therapy programs. He was seen regularly by visiting allied health services, documenting modest improvements in his language function and attention. School psychology assessment was conducted regularly including at the time of his transition to high school to inform and update his individual education plan.

### **Case Profile 3 – Adolescent**

Serena: 16 years

***History:*** Serena is 16 years old. Her mother drank two to three standard drinks most nights during her pregnancy, predominantly wine and some spirits, a habit that had developed when she moved from overseas to work in hospitality in Australia. Serena's parents divorced when Serena was 4 years old, after the birth of her younger brother, Shannon. Serena's mother moved back to her home country leaving her father as the sole parent, although his sister (Serena's paternal aunt) became a 'de-facto'

mother. Serena has contact with her mother every few years. Her mother has attended alcohol rehabilitation programs several times and has abstained from drinking for two years.

Serena's father has many concerns about his teenage daughter. Most of all, he is worried about the fact that she is involved in online interactions with older men; including provocative text and photo messaging, which the parent of one of Serena's school friends alerted him to.

Serena is currently living with her paternal aunt due to arguments with her father.

Stealing from the clothes stores at the local shopping centre has been a regular occurrence, with Serena now facing her second court appearance. On one occasion, she was accused of assaulting a store security officer. Serena appears to have little understanding of the consequences of her behaviour, frustrating both her probation officer and her father, despite their repeated advice.

Serena's school attendance has been increasingly erratic, because of a suspension for smoking marijuana in the school grounds at the start of last term. Her teachers reported she was 'talkative', seemed 'bright' but was highly oppositional, frequently untruthful and disorganised, rarely completing homework. She had agreed to psychology assessment by the school counsellor. This had been suggested by her year-level coordinator due to Serena's consistent performance well below grade expectation for two years.

**Screening and primary care:** Serena was a healthy child whose development progressed well apart from difficulties socialising with her peers in her early primary school years. In later primary school, her teachers had raised concerns about her ability to focus in class. On some occasions, she had 'emotional outbursts' towards teachers which they found difficult to handle and hard to understand. Serena had difficulties adjusting to the learning expectations of middle school, but managed to develop a peer group and maintained her grades at a sufficient (though low) level so that she can progress to high school. Her father's main concern was Serena's 'obsession' with boys and her many boyfriends from year 7 onwards.

**Diagnostic assessment:** Serena's father read about FASD in the newspaper and arranged a referral from his GP to a paediatrician and adolescent medicine physician. Serena's aunt agreed that 'something had to be done'.

Birth records obtained from the local hospital documented 'maternal alcohol abuse', normal range birth weight, length and head circumference, and a normal neonatal check with no dysmorphism noted.

Serena's growth parameters were within the healthy range, as were available birth and postnatal growth measurements. She had no diagnostic facial features. Serena's school counsellor had recently completed a cognitive assessment that indicated her cognitive skills were in the low average range in all areas apart from processing speed, which was in the borderline range. Her maths capabilities on the WIAT II were overall at the 3<sup>rd</sup> percentile, numerical operations were at the 2<sup>nd</sup> percentile and maths reasoning at the 3<sup>rd</sup> percentile.

The adolescent medicine physician completed a HEADDSS adolescent risk and protective factors assessment, and diagnosed Generalised Anxiety Disorder, Conduct Disorder and marijuana misuse. She referred Serena for further psychological testing with a neuropsychologist. Her lawyer arranged assessment promptly due to her upcoming court appearance. Testing included Serena completing the NEPSY-II over several hours with scores below the 3<sup>rd</sup> percentile in several components of the Executive Function and Attention and Memory and Learning domains. Her aunty completed the Behaviour Rating Inventory of Executive Function and Vineland Adaptive Behaviour Scales, the latter

showing significant deficits in Socialization Skills (Interpersonal Relationships and Coping Skills) and moderate impairment in aspects of Daily Living.

Serena was found to have significant impairment in the following domains: academic achievement; executive functioning; memory; attention deficit/hyperactivity and behaviour problems: and adaptive behaviour/social skills.

The adolescent physician and neuropsychologist discussed Serena's assessment results. Her high level marijuana use was considered a significant potential contributing factor to her weaknesses on testing. However the effects of her confirmed high-risk prenatal alcohol exposure were considered to be evident in her neurobehavioural and cognitive impairment in multiple domains. *A diagnosis of Neurodevelopmental Disorder-Alcohol Exposed* was made in addition to her other diagnoses.

Blood tests including iron studies, TSH, fragile X and microarray were normal. Formal optometry and audiology tests were normal, both needing to be repeated due to Serena's flippant approach to testing the first time around.

**Therapy approach:** Serena's parents and lawyers notified the school and courts of her diagnosis on the FASD spectrum. This was taken into consideration at her second court appearance, leading to mental health diversion rather than having the matter finalised in the Children's Court.

A multidisciplinary strength and resilience based approach was required to address Serena's range of physical, emotional and behavioural issues, especially her unsafe sexual behaviour and recurrent criminal behaviour. This included:

- Arranging a sexual health check with adolescent paediatrician, discussion of contraception options and ongoing monitoring and support regarding sexual health by a female GP.
- Arranging her own Medicare card, so Serena could access healthcare independently.
- Referral to youth mental health service, for:
  - Regular counselling/therapy (re anxiety disorder, emotional regulation, marijuana use and sexual/social relationships) with Serena's caregivers involved as appropriate.
  - Psychiatry assessment (particularly regarding medication options).
- Case coordination/case manager through the juvenile justice system or family support organisation.
- Caregiver support and education, with Serena's father and aunt educated about FASD and connected to a parent support group.
- Developing an individualised educational plan:
  - Emphasis on a life skills curriculum and keeping Serena in school, including recognition of her Specific Learning Disorder with impairment in mathematics.
  - Vocational support, developing strengths and interest area (with school involvement) in hairdressing.
- Case conference arranged and planned 6 monthly.

**Developmental surveillance and review:** an ongoing plan of review for Serena would involve:

- Psychologist (weekly/fortnightly).
- GP (3 monthly).
- Adolescent physician (6 monthly).
- Psychiatrist (yearly/as required).

## 4. Key Findings

This section sets out:

- Key findings of the review of FASD across the life course, to assist with planning access to interventions, services and supports for people living with FASD.
- A framework of principles and best practice indicators to guide NDIA planning processes for disability support which assists individuals achieve their best possible life outcomes.
- Guidance relating to the level of support for individuals with FASD impairments of varying severity.

### 4.1 FASD over the life course

Lack of awareness of FASD is one of the overarching constructs currently disrupting the capacity of many individuals living with FASD to access the services and supports they require to fully participate in society<sup>13</sup>. FASD presents as a range of **lifelong primary disabilities** with strikingly high rates of secondary effects on daily function and participation<sup>14</sup>. **Primary disabilities** are those disabilities caused directly by prenatal alcohol exposure. Secondary effects include mental health problems, drug and alcohol dependency, and contact with the criminal justice system. **Secondary effects** are disabilities that an individual is not born with. Preventable secondary effects may be described as the constant misunderstanding of primary disabilities and a poor fit between the individual's needs, developmental age and the environment<sup>15</sup>. This may be the result of the primary FASD disabilities not being appropriately addressed.

Accumulating evidence since the late 1980s, primarily from the USA and Canada, describes impairments attributable to FASD and associated interventions<sup>16 17 18 19 20</sup>. Despite the increasing evidence health, education and social support systems have continued to struggle with adequately supporting individuals living with FASD.

The burden of FASD on individuals and their families is increased with this lack of awareness<sup>21</sup>. In a number of studies caregivers describe treatment barriers, unmet needs, and high levels of parenting stress<sup>22</sup>. While early childhood intervention is known to mitigate impact, FASD is often not recognised until a child enters formal education. For example, in a very recent study with foster and adopted children; 86 per cent of youth who met criteria for a diagnosis related to FASD had never been previously diagnosed or had been misdiagnosed<sup>23</sup>. Even if FASD is considered, other barriers may delay understanding the full extent of an individual's neurodevelopmental impairments and what this means in their daily life. Among health and other professionals, for example in education and justice, understanding of FASD is only more recently gaining awareness with significant gaps existing in the knowledge base of professionals<sup>24 25 26 27</sup>.

Over the lifespan, caregivers and individuals with FASD interact with a wide range of service systems, but frequently encounter barriers. Caregivers raising children with FASD have many unmet important needs<sup>28</sup>. Access to appropriate services and supports across all systems of care is a clearly stated need and a high priority among caregivers raising children with FASD and community professionals supporting them<sup>29 30</sup>. These caregivers also show significant parenting stress<sup>31</sup> and at levels higher than those raising children with autism<sup>32</sup>.

While there are barriers for individuals living with FASD, there are also enablers. These include the presence of known protective factors, such as a stable and caring environment and assisting individuals to have easy access to the services and supports they require across their life. When combined, the evidence suggests that integrated support and coordinated service delivery at key

transitional points maximise the lived experience of each person with FASD. What are lacking are system based approaches, such as case management models, to support integration and coordination of services and supports. An approach to management of FASD over the life course that is multilateral, cross disciplinary and collaborative is required to enhance the quality of life for individuals, families and communities and can assist to mediate against the vulnerabilities present with FASD.

#### **4.2 FASD and functional impairment**

Functional impairment in FASD falls into two broad areas: 1] underlying brain functioning; and, 2] the behavioural consequences of impaired brain functioning. Underlying brain functioning is referred to throughout the review as the **brain domains of functional impairment**. Interventions described in this review are those targeted at modifying behaviours resulting from functional impairment.

The range of domains of functional impairment is explained in detail in Section Two. The domains correlate to one or more brain region/s responsible for controlling human functioning and behaviour, creating a unique but variable neurodevelopmental profile. For each individual with FASD, while there is an obvious overlap between underlying brain functioning and behaviour, it is evident that recent research is emphasising the primacy of identifying which neurodevelopmental domains are impaired *prior* to considering how to modify resulting behaviours through targeted intervention.

Ideally, a diagnosis of functional impairment achieved through comprehensive, multidisciplinary assessment is required to set in motion the most relevant interventions and supports for the individual. A specific diagnosis (FAS/pFAS/ND-AE) on the FASD spectrum does not automatically connect to a 'matched set' of impairments, as each individual manifests a unique profile of neurocognitive impairments that express differently over their lifetime<sup>33 34</sup>. Assessment of impaired brain domains is therefore not a one-time process and is not reliant on diagnosis. Access to services requires more than a diagnostic label and must be based on **functional need**. As tasks associated with daily functioning become more complex across the individual's life course, **assessment/re-assessment** is required to evaluate the severity of functional impairment at known transitional times.

The categories of neurodevelopmental impairment commonly identified in FASD, for which **evidence-based interventions** have been identified, include:

- Adaptive functioning (everyday skills required to function and meet environmental demands; includes motor skills, language and social communication).
- Behaviour and self-regulation (attention, hyperactivity, impulsivity, sensory processing, impulse control).
- Cognition and executive functioning (academic achievement, memory, judgement, organisation, attention, visual motor integration).

#### **4.3 FASD and intervention**

While early diagnosis leading to early intervention may occur for children with distinctive FAS (as demonstrated through cardinal facial characteristics and growth impairment); other children on the spectrum, without these features, may miss out on crucial early childhood intervention because of a delayed or inaccurate diagnosis<sup>35</sup>. For example, infants and young children with FASD may initially be diagnosed with Global Developmental Delay<sup>36</sup> (or be missed altogether if their delays are subtle) when knowledge of prenatal exposures, including alcohol, is missing. With limited FASD awareness and diagnostic capacity in Australia, diagnosis may be delayed until 4-5 years of age or later<sup>37</sup> or may not occur at all. These circumstances delay the implementation of a range of interventions essential to improving the functioning of children with FASD<sup>38</sup>. Implementation of routine screening for alcohol

use in pregnancy and subsequent developmental surveillance of infants known to be at high-risk for FASD may lower the age of diagnosis and enable early childhood intervention to be instigated in the future.

At a policy level, taking a life course approach to diagnosis, screening, referral, and assessment/reassessment related to FASD will direct children and families into relevant supports, services and interventions at the earliest opportunity. This approach requires clear articulation by the NDIA to ensure that each individual affected by FASD, at which ever age this is identified, can enter and exit a life course trajectory of support as required and at transitional times to enhance their capacity to participate in education, employment and social activities. There are clear benefits to individuals in this approach, not least because the economic impacts of the chronic and lifelong conditions associated with FASD can be reduced through timely and appropriate intervention and support<sup>39</sup>.

### ***Key findings summary***

It is established that FASD is a lifelong condition characterised by a range of significant domains of functional impairment, often requiring behavioural modification and environmental accommodation interventions.

FASD is characterised by primary disabilities which are the result of prenatal alcohol exposure. Secondary effects are those disabilities that an individual is not born with but are caused by a lack of appropriate services and supports to address the primary disabilities. Individuals affected by FASD will have varied neurocognitive profiles and the extent of individual impairment may not become apparent until there is formal engagement with education. There is a growing body of evidence on effective interventions to address different aspects of cognitive and behavioural issues associated with FASD, which benefit from exposure to services and supports designed to address these.

While early diagnosis or assessment is optimal to enable early childhood intervention to take place, there are key transitional points along the life course when individuals are likely to benefit from assessment/re-assessment and referral to age appropriate interventions designed to improve daily functioning and participation in education, employment and social opportunities.

## 5. Domains of Functional Impairment

The domains of functional impairment associated with FASD diagnosis are significant and distinct to each affected individual. Understanding the extent of individual impairments and the points at which these are able to be identified is crucial to establishing a pathway to appropriate interventions. A detailed presentation of the analysis of studies directly related to central nervous system (CNS) functional impairments indicated in FASD is included in Section Two. The most relevant outcomes from analysis of the evidence are summarised here.

Overall, the evidence demonstrates that deficits in the executive functioning skills of individuals are a hallmark of FASD<sup>40</sup>. The complexities of executive functioning in daily life, and how this might interrupt individual capacity, is discussed in detail in Section Two. Other affected brain domains likely to be impaired are: attention/activity level/sensory processing, academic achievement, adaptive behaviour/social skills/social communication, cognition (or global development if < 5 years of age), language (expressive and receptive), memory, fine or gross motor function, affect regulation (e.g. DSM-5 criteria for major depressive disorder, social anxiety disorder etc.) and structural abnormality/microcephaly/hard neurological signs/seizure disorder. Assessment related to the functioning of these brain domains are critical to successful FASD identification and management<sup>41</sup>. An integrated approach to management must then be considered within the context of the everyday environments of the individual with FASD<sup>42</sup>.

Executive functioning encompasses intellectual abilities, activity, attention and information processing, self-regulation, language, visual processing, learning and memory, number processing and social cognition<sup>43 44</sup>. Adaptive functioning incorporates function and participation in activities of daily living, social participation and work or school performance<sup>45</sup>, while sensory processing relates to how information is received through the senses enabling an individual to function smoothly in their daily life<sup>46</sup>. Self-regulation refers to how efficiently and effectively a person deals with a stressor and then recovers<sup>47 48 49</sup>. With FASD, as brain injury occurs prenatally, this interrupts a child's ability to self-regulate behaviour with outcomes including impulsivity, a lack of social inhibition and impaired language development<sup>50</sup>.

An important note for the NDIA is the cautious use of IQ scores as a determinant of eligibility for services and supports as IQ is not always indicative of impaired functioning for persons with FASD. For example, children who have been prenatally exposed to alcohol have been found to have lower overall intellectual functioning, however their intelligence scores have been found to vary widely within this population. IQ scores in those with FASD can range from as low as 40 to as high as 114<sup>51</sup>. There can also be a significant discrepancy between IQ and adaptive functioning, in that a child may have an average IQ but be unable to apply it in an 'adaptive' manner.

Further, cognitive functioning is affected even in children who do not show the facial features that are characteristic of FAS. In the absence of facial features, a measure of IQ may be used but the average IQ estimate for individuals with FASD is 80<sup>52</sup>. Therefore, **overall** functional need must determine access to support in the presence of FASD<sup>53</sup>, and using an IQ cut-off is not recommended.

It is worth noting that there is a striking similarity between CNS abnormalities and the behavioural phenotypes of traumatised children and children with FASD. This apparent overlap has not yet been well addressed in the medical or behavioural science literature<sup>54</sup>. For the purposes of this review, we note that childhood trauma occurring independently of FASD may contribute to exacerbation of FASD impact.

As children with FASD get older, they lag further behind their peers in self-regulatory and social communication abilities<sup>55 56</sup>. Children with FASD have a predisposition and vulnerability to mental health disorders and are at risk for abuse and neglect over their life course. They have increased rates of externalising and internalising behavioural problems which are commonly reported by both parents and teachers<sup>57 58</sup>. Externalising problems include: ADHD and impulsivity with significant deficits in social behaviour including aggression<sup>59</sup>. Rates of oppositional defiant disorder and conduct disorder are also elevated in alcohol-exposed children<sup>60</sup> further exacerbating deficits in mood and behavioral regulation. Internalising symptoms such as low self-esteem, social isolation, and mood disorders such as major depressive disorder and suicide<sup>61</sup> are frequently reported with higher rates of negative affect<sup>62</sup>. When an early diagnosis is absent, or early intervention is not implemented, secondary effects are more likely. These include disrupted school experiences, trouble with the law, inappropriate sexual behaviour, and alcohol/drug problems<sup>63</sup>.

There is no well-ordered age and stage categorisation of domains of functional impairment related to FASD in the literature. Instead, the available evidence has been synthesised below to highlight the likely age related progress of functional impairments. These are described for: infancy/early childhood; childhood (4 to 12 years); adolescence (13 to 18 years); and, adulthood.

### **5.1 Infancy/early childhood**

More evidence is needed investigating the impact of alcohol in the 0-3 age range. However, in the neonatal period it is possible to assess habituation<sup>64</sup> and other infant behaviours. For example, abnormal rates of habituation have been related to later delays in intellectual development<sup>65</sup>. Further, neonates displaying difficulty in maintaining calm/alert sleep states suggests that self-regulatory problems are evident from the time of birth. Other neonatal characteristics correlated with prenatal exposure to alcohol include: increased hand to mouth activity, a strong MORO (startle) reflex, hypotonia (reduced muscle tone) and a delayed stepping reflex threshold. Early non-specific indicators of developmental vulnerability include feeding difficulties, poor visual and auditory habituation, sleep disturbances, poor sleep/wake cycle, and delays in reaching motor milestones such as sitting, crawling and walking<sup>66</sup>.

Insecure attachment and parenting difficulties and irritability (negative affectivity) of the infant are often reported with suspected or confirmed prenatal exposure to alcohol. O'Connor and Paley (2009) provide a comprehensive review of psychiatric conditions associated with prenatal alcohol exposure as these relate to the life course<sup>67</sup>. Overall, insecure attachment may lead to impaired or atypical social-emotional development, poor school readiness, behavioural problems, psychiatric disorders, and impaired long term mental health outcomes<sup>68 69</sup>.

In early childhood, global developmental delay in the context of known prenatal alcohol exposure may be a common indication of FASD, although more subtle deficits are also common. Indeed, just over half of children aged birth to 3 years with prenatal alcohol exposure (or with FAS/pFAS) show marked developmental delay<sup>70</sup>. This includes delays in gross and fine motor skills, speech and language, cognitive and social and emotional development. This means that many young children who later have diagnoses on the FASD spectrum (including FAS) are not identified by common methods of ascertaining early, global developmental delay<sup>71</sup>.

Infants and young children prenatally exposed to alcohol may also have a variety of associated medical problems such as hearing and vision impairment and failure to thrive. Birth defects of the heart, kidneys, and musculoskeletal system, and structural CNS abnormalities have been commonly



described in FASD and must be considered during medical assessment. Ongoing management of these important medical issues sits alongside neurocognitive and behavioural interventions in FASD.

## **5.2 Childhood (4 to 12 years)**

By the time a child reaches four years of age, evidence shows the main difficulties starting to emerge are attention<sup>72 73 74</sup> (a component of executive functioning) and behavioural problems<sup>75 76</sup> within the self-regulation and social communication domains of functioning, gross<sup>77</sup> and fine motor skills<sup>78 79</sup> sensory processing and adaptive functioning<sup>80 81</sup>. A wider variety of neurobehavioural assessments can be administered in this age range (see Table 8, Appendices). In addition to attentional difficulties and slower reaction times, children around this age have also been noted as acting young for their age, with hyperactivity reported, and impulsivity and restlessness or irritability also becoming evident<sup>82</sup>. It is important to note that the inattention and hyperactivity associated with prenatal alcohol exposure may denote a particular clinical subtype with an earlier onset, a different clinical and neuropsychological presentation, and probably a differential medication response than idiopathic attention deficit hyperactivity disorder (ADHD)<sup>83 84</sup> which is a common comorbidity alongside FASD.

Children in the 5-8 years age range have been shown to have sensory processing dysfunction using the Short Sensory Profile (SSP) and the sensorimotor core domain on the NEPSY-SM (a neuropsychological assessment tool) than typically developing children, as well as more problems with sensory modulation and sensory motor performance. They also have more significant difficulties in the spelling and arithmetic sections in the Wide Range Achievement Test (WRAT-3) with significant correlations between the SSP and the maths and spelling sections of the WRAT-3, suggesting a link between sensory processing dysfunction and the academic function of some children with FASD<sup>85</sup>.

It is noteworthy that children with pFAS or ND-AE may perform worse with their sensory processing scores and adaptive behaviours than those diagnosed with FAS, but perform better on measures of intellectual functioning<sup>86</sup>. These results support the theory that deficits in adaptive behaviours may be independent of lower IQ<sup>87 88</sup>. Lower levels of adaptive functioning can be assessed using specific tools such as the Scales of Independent Behaviour-Revised (SIB-R) or the Vineland Adaptive Behaviour Scales (VABS) to determine the degree of impairment and its impact for the individual<sup>89 90</sup>.

Generally, self-care, self-management and social functioning are impaired and may change during childhood as the complexities of the tasks and activities increase. For example, in younger children toileting, dressing and mealtime delays may be evident, while in middle childhood problems such as understanding money/value will start to emerge. Children on the FASD spectrum have also been found to exhibit disruptive, uncooperative and socially inappropriate behaviours compared with typically developing children; requiring extensive or frequent support and supervision to manage daily adaptive skills and behaviour compared with typically developing children only needing intermittent support<sup>91 92</sup>. Mental health and behavioural disorders may also manifest from this age<sup>93</sup>.

Sleep disturbances have an impact on behavioural outcomes, particularly self-regulation<sup>94</sup> but also on memory, learning, attention and cognitive flexibility. These have also been identified in other neurodevelopmental disorders such as autism or ADHD with studies reporting very high rates of sleep disturbances in affected children<sup>95 96</sup>.

All these indicators support the need for referral to diagnostic and/or multidisciplinary assessment services if FASD has not previously been identified as the potential cause of emerging atypical behaviours.

### 5.3 Adolescence (13 to 18 years)

Diagnosis and assessment of FASD in adolescence, and adults, creates special challenges due to a change in or lack of physical features as well as additional environmental factors that add to the individual's history such as alcohol and drug abuse and mental health problems<sup>97</sup>. A recent cross sectional study considered age related differences across a broad range of neuropsychological variables in three age groups: 5-8, 9-12 and 13-17 years<sup>98</sup>. This study is an important attempt at understanding how neuropsychological impairments manifest across the life course of children and adolescents with FASD. Academic achievement, language, memory, visual motor coordination and executive functioning were all areas of impairment identified in children with FASD. In terms of age related differences, the brain domains where older subjects performed worse than younger subjects were: executive functioning, arithmetic, learning and memory, visual motor integration and motor abilities. There were no significant differences in IQ between the three age groups in this study with mean IQ scores of 85 across the three age groups.

With regard to academic achievement, arithmetic is the greatest area of impairment consistent across studies of FASD<sup>99</sup>. Impaired receptive language as measured on the Peabody Picture Vocabulary Test-revised (PPVT-R) is also consistent with other studies<sup>100</sup>. Both children and adolescents with FASD have been shown to have significant difficulty when tested on learning and recall of verbal information<sup>101</sup>, although it has been found that while explicit memory may be impaired, implicit memory may be intact<sup>102</sup>.

Attention, spatial learning deficits and tasks requiring the complex manipulation of information in 14 year olds has been reported<sup>103</sup>. Other studies have reported difficulties with problem solving, verbal and spatial learning and flexibility of thinking, which were identified using the card sorting tests of the Delis Kaplan Executive Function System (D-KEFS)<sup>104</sup>.

Secondary effects that have developed as a reaction to, and as a way of coping with, the primary impairments of FASD include: increased anti-social behaviours; earlier use of alcohol and other drugs; increased self-perception of difference; and, the self-conceptualisation of being a 'poor student'. Stress is identified as a trigger for more difficulties with organising materials and processes and with a high impulsivity rating on the Child Behaviour Checklist (CBCL). Adolescents have personally identified their school experiences as difficult as they report finding it "hard to learn", although they may not identify that FASD was the reason behind their learning difficulties<sup>105</sup>.

In addition to the tasks of adolescent development and living with FASD, some adolescents may also have children of their own to raise who may or may not be residing in their care. Parenting when affected by FASD represents many challenges and is an important opportunity to begin interventions that can minimise secondary effects and promote coping with the brain based primary impairments<sup>106</sup>.

In adolescents and young adults, mental health concerns are often expressed in alcohol misuse/disorders<sup>107 108</sup>, depression<sup>109 110</sup> and suicide<sup>111</sup> and a variety of other mental health disorders<sup>112</sup>. A general lack of engagement with the community and contact with the justice system are other common characteristics identified in adolescence. Although not specific to adolescence, the impact of FASD is also mediated by environmental factors such as living with an alcoholic parent or being subjected to child abuse or neglect, or other adverse life conditions such as poverty which often lead to child welfare involvement and a child's admission to care<sup>113</sup>.

## 5.4 Adult

For over three decades pioneering FASD researcher Ann Streissguth has documented risk factors for secondary disabling effects or adverse life outcomes associated with a FAS diagnosis. For young adults mental health problems, contact with the justice system, disrupted education, and alcohol dependence/substance abuse issues were significant at 21 years of age whilst less than 10 per cent of subjects with FASD were able to live or work independently. Motor deficits were also evident, particularly speed of information processing<sup>114</sup>. Frequent adverse life outcomes included inappropriate sexual behaviours (40-50 per cent increasing with age) and mental health problems at higher than 90 per cent<sup>115</sup>. Co-occurring risk factors included being the victim of domestic abuse or violence. In addition the incidence of independence in daily living activities amongst 21 year olds with FASD was very low for activities such as managing money and making decisions, with 80 per cent of individuals requiring help or supervision<sup>116</sup>.

Other evidence from a long term German study<sup>117</sup> demonstrated that characteristics such as growth retardation, microcephaly, developmental delay and hyperactivity generally persisted into adulthood. While FAS facial features present earlier in life were generally not apparent in adulthood, the continued prevalence of microcephaly at follow up was strongly correlated with those subjects who were intellectually disabled at the initial assessment. In that study, there was a high persistence from childhood to adulthood of aggressive, externalising and delinquent behavioural problems. In addition, a number of secondary life outcomes including poor school achievement (49 per cent) and receiving special education past primary school (38 per cent) were evident. In that group only 13 per cent had completed secondary school and held an ordinary job, 27 per cent lived in institutions, 35 per cent were in a dependent living situation with assistance from others, with only 14 per cent able to live independently alone.

In a further study, sixteen young people (age range 16-27) with FASD and a range of intellectual functioning levels (low average to above average IQ) manifested clear deficits on neuropsychological measures sensitive to complex attention, verbal learning and executive function. Both the frequency and severity of cognitive impairment demonstrated in both FAS groups were greater than would have been predicted on the basis of IQ alone<sup>118</sup>.

### ***Domains of Impairment Summary***

There are a range of neurodevelopmental domains, including neurocognitive, adaptive functioning and behavioural and self-regulation, which are impaired in individuals with FASD. Each affected individual has an individual neurocognitive profile resulting in functional impairments to any of the brain domains and which may not become apparent until formal engagement with the education system occurs. Attachment difficulties in infancy and both global and subtle developmental delays, in the context of prenatal exposure to alcohol, may be useful indicators highlighting the need for multidisciplinary diagnosis and/or assessment for FASD to assist with early childhood intervention.

The domains of functional impairment considered for FASD diagnosis are:

Attention/activity level/sensory processing

Academic achievement

Adaptive behaviour/social skills/social communication

Cognition (or global development if < 5 years of age)

Executive functioning

Language (expressive and receptive)

Memory

Fine or gross motor function

Affect regulation (e.g. DSM-5 criteria for major depressive disorder, social anxiety disorder etc.)

Structural abnormality/microcephaly/hard neurological signs/seizure disorder

Assessments relevant to measuring impairments in the brain domains are critical to successful FASD identification and management. This needs to be followed by an integrated approach to management within the context of the environment of the individual with FASD.

Functional impairments requiring substantial or very substantial support are reliant on an individual profile being determined for each individual, with reassessment at transitional times across the life course to determine which types of support is required relevant to the impairment. Reassessment is vital to continue to provide appropriate support for the individual with FASD as task demands increase with age and with the increasing incidence of comorbidities such as mental health disorders that significantly impact on daily functionality. Functional need must determine access to support in FASD, irrespective of the intellectual ability of the individual.

## 6. Interventions

Treatment and management approaches addressing primary disabilities and secondary effects arising from FASD are evolving.

These are based on increased understanding of brain structure and function and what happens when brain domains are impaired through prenatal exposure to alcohol. Currently, interventions are focussed on improving individual daily functional capacity and are therefore cornerstones to improving behaviours resulting from impaired brain functioning. Interventions may include both those directed at the affected individual and those which also support family members and caregivers.

International guidelines have recently been published by the Substance Abuse and Mental Health Services Administration (SAMHSA) in the USA with a Treatment Improvement Protocol (TIP 58) for Addressing Fetal Alcohol Spectrum Disorders<sup>119</sup>. The TIP 58 provides information and concrete resources, strategies and example policies to help guide the provision of substance abuse and mental health care services which are FASD informed.

Interventions specific to education or justice programs, or which solely utilise pharmacological interventions for individuals with particular FASD neurocognitive profiles under medical management have been excluded from this review. Rather, interventions which have been developed, trialled and evaluated for participants with FASD or similar conditions and which are integrated across settings or are primarily community based have been included. Collaborative interventions which involve family/caregivers, teachers or community support workers, and/or therapists in an integrated response to an individual's needs have been identified as particularly relevant to NDIA.

As the evidence base for interventions is emerging, reference is made to both higher and lower level evidence (i.e. Level 1 – sufficient rigour associated with study design and outcomes; and Level 2 – limitations in study design, but of sufficient quality as emerging evidence). Twenty one quantitative studies of sufficient quality are included in the critical review with the intervention programs summarised in Table 1 and set out in detail in Section Two. Emerging interventions are addressed in this section with further detail of these in the Appendices. An additional nine intervention studies in progress or recently completed are also described (Table 11, Appendices). A number of these are funded by the USA Centres for Disease Control and Prevention (CDC) and appear to be high quality RCTs with adequate sample size. Five qualitative studies of sufficient rigour (Level 1 evidence) have also contributed to the evidence base included in this review.

The review of interventions has also enabled identification of notable principles currently informing intervention practice, and which therefore constitute current best practice guidance. This information has been extracted as planning principles and best practice indicators, and is described further in the Discussion.

### 6.1 Overview

Recent advances in cognitive neuroscience and understanding of the neuropsychology of FASD is maximising the success of interventions designed for children prenatally affected by alcohol<sup>120 121 122</sup>. A neurodevelopmental framework based on cognitive neuroscience principles therefore guides development of interventions for children with FASD. Within this framework, targeted, domain-specific interventions (e.g. targeting executive attention and self-regulation) are hypothesised to lead to generalised improvements in cognitive function and behaviour<sup>123</sup>.

From the outset it is important to consider the child, young person or adult with FASD within the context of his/her everyday environment, enabling pathways to the sharing of resources between

health, education and other involved services – in effect a **coordinated family + school + therapy partnership approach**. This partnership approach requires a **coordinating or lead agency** capable of appointing case managers. In the case of young adults who have left the education system, a partnership approach is still relevant, but will potentially involve different agencies or services in place of school. It is vital that the family remain central to all aspects of decision making with the intervention team. How coordination is undertaken will depend on the location, the available agencies (for example, the Disability Services Commission, Western Australia) working with a family to determine what is required to reach identified goals and facilitating access to the available support services.

To achieve functional goals requires a holistic approach, paying attention to familial, community and cultural considerations relevant to the individual. Given the heterogeneity of CNS effects, a neurodevelopmental profile specific to the child or adult is needed for intervention planning. Such profiling gives vital background for planning which resources are required for the individual, taking account of the range of neuropsychological variables that may be affected<sup>124</sup>.

In a review of interventions now available specifically for FASD, factors noted as associated with success included:

- Parent education or training alongside interventions for a child with FASD.
- Explicitly teaching children with FASD skills that typically developing children may learn through observation or abstraction.
- Integration of individualised interventions into an existing system of treatment (e.g. local early childhood intervention services, school based education services, universal services and everyday environments)<sup>125</sup>.

It has also been established that as standard practice most individuals with FASD benefit from multiple types of intervention services<sup>126</sup>. These include occupational therapy, speech and language therapy, psychological therapy, physiotherapy, social support and individualised education plans. Interventions will change over time depending on the number, type and severity of FASD associated problems. This further emphasises the principle of a lead agency supporting a coordinated family + school + therapy partnership which facilitates access to multiple interventions, especially when these are provided in different settings and involve multiple agencies.

Intervention recommendations will vary according to age of the individual with FASD; with evidence supporting recommendations for family support resources and social service-child welfare interventions made to children aged birth to two years. Mental health recommendations and referrals to community based programs for recreation or leisure activities are highest among children aged 3 years and older. Specific behavioural or environmental accommodations may increase with age, with a high proportion of this type of intervention recommendation among children aged 12 to 18 years<sup>127</sup>.

Principles for inclusion in a general framework providing guidance on best practice specific to FASD include:

- Capacity building for families and wrapping services around the child;
- Reducing stigma and shame for parents whose children may be diagnosed with FASD (e.g., by acknowledging their concern for the child's well-being and their importance as a member of the care team).
- Acknowledging the challenges of parenting a child with FASD and encouraging parents and caregivers to pay attention to their own needs; including helping families to access respite care to reduce family stress and improve family function.

- Providing resources (e.g., parent management training and parent counselling, parent support groups).
- Assessing family members for mental health problems, substance use and addiction and helping them obtain treatment.
- Determining the risk for child abuse, neglect, and violence in the family and providing referrals as indicated.
- Helping parents and caregivers to identify available intervention services (e.g., through the school or community).
- Working with parents and caregivers to ensure access to appropriate services, including vocational training and skills of daily living (e.g., personal hygiene, money management)<sup>128</sup>.

While these general principles are a starting point, there are other considerations relevant to the Australian context which NDIA would need to take account of to formulate best practice guidelines.

## **6.2 Cultural security**

Planning FASD services and supports when these are intended for Indigenous and other culturally and linguistically diverse (CALD) people requires consideration of culturally specific factors which may affect access and service delivery. Currently, there are no evaluations of Australian programs aimed at alleviating the effects of FASD in Indigenous communities<sup>129</sup> or indeed in CALD populations.

Instead, evidence from USA and Canadian studies identifies programs shown to be effective when used in contexts that are not specifically Indigenous, but which could be interpreted and adapted for use in the Australian Indigenous or CALD context. These are: the Families Moving Forward Program; Parent-Child Assistance Program (PCAP) Children's Friendship Training; Neurocognitive Habilitation Therapy; The Alert program<sup>®</sup>, and, computerised attention training (CPAT). Other programs in use in the Australian context, such as Triple P, might also be useful.

While The Alert program<sup>®</sup> is in use in Australia for autism and other neurodevelopmental disabilities it has only very recently (2015) been introduced in remote communities of the Fitzroy Valley, Western Australia, in the context of a randomised controlled trial, adapted to suit the specific cultural and language context in a largely Indigenous population with an identified high prevalence of FASD. For example, assessments used to determine impairment in executive functioning and self-regulation have been translated into local languages and local Aboriginal researchers employed to administer these assessments.

There are also some general principles drawn from the broader health service delivery sphere which are relevant to providing disability interventions, services and supports in Indigenous contexts.

For example, cross cultural issues and language differences, and logistical challenges of delivering services to regional and remote Indigenous communities all need to be considered when planning referrals in all Australian states and territories. Further, understanding the social determinants, lived histories and the impact of intergenerational trauma on Indigenous people in Australia<sup>130</sup> are important considerations in a planning process. Finally, consulting closely with and drawing on the strengths of local communities is likely to improve the capacity to engage affected individuals and their families with available services when these are provided in a culturally secure way.

A strengths based approach has been found to achieve positive outcomes in supporting and enhancing good family functioning among Indigenous families<sup>131</sup>. Two Australian case studies that provide guidance with an objective to simultaneously strengthen Indigenous family functioning and address the risk factors associated with negative family functioning are relevant. These are the Learning

Together (Literacy) project; a literacy and early learning program delivered through facilitated playgroups for Indigenous and non-Indigenous families with children under 4 years; and, the Jalaris Aboriginal Corporation working with marginalised Indigenous families to improve health, child development and family functioning<sup>132</sup>.

Drawing on the Canadian experience indicates other potential approaches that may provide guidance in the Australian Indigenous, and potentially also the CALD, contexts. For example, virtual collaborative processes, involving researchers, service providers, health system planners and Aboriginal health advocates in small virtual communities, are being used to address the needs of Indigenous women using substances in Canada<sup>133</sup>. This approach is enabling widely dispersed geographical areas to come together and share information, while maintaining cultural sensitivities. A holistic approach that addresses vocational, mental health, and physical health needs as well as culture and tradition can improve interest, engagement and relevance of treatment for families<sup>134</sup>. There is potential for this virtual collaborative process in the Australian context using existing telehealth services.

Similarly with the Canadian experience<sup>135</sup> Australian Aboriginal and Torres Strait Islander children are more likely to come into welfare care more frequently. This is often as a result of the presence of multiple risk factors, including early life trauma. Understanding is required that this most likely stems from the negative impacts of racism, poverty and intergenerational trauma. These issues have arisen from a combination of policy and societal impacts experienced by Aboriginal and Torres Strait Islanders peoples (p.8)<sup>136</sup>. These circumstances provoke other variables which lead to childhood trauma, including alcohol use by parents to cope with adverse life conditions, with substance use during pregnancy the primary risk factor for the occurrence of FASD<sup>137</sup>.

To address family functioning, mentoring programs based on the Parent-Child Assistance Program (PCAP) model to support women after childbirth, and advocacy/mentor type programs for women with FASD themselves are showing promise within Indigenous populations<sup>138</sup>. New efforts are underway to integrate these programs with other elements, such as incorporating infant mental health interventions with the PCAP model, to further enhance their effectiveness. These approaches assist access to services and supports when these systems are integrated and culturally secure.

It is evident there is a substantial gap in evidence related to effective services and supports for both Indigenous and CALD populations. In a highly ethnically diverse population as exists in Australia, this is an area requiring attention.

### **6.3 Family centred care**

Family members, foster carers and the broader community play a crucial role in supporting individuals affected by FASD to achieve their full potential. This is particularly true as the capacity to live and work independently is often limited. Family members may not be the biological parent/s, and may include relatives, foster carers and other non-related caregivers. Family 'type' is significant, because the diversity of families includes birth, foster, and adoptive families and they all respond differently to the presence of a child with FASD<sup>139</sup>.

Estimates of the percentage of children with FASD raised by people other than their biological parents range from 60 to 80 per cent across Australia, Canada and North America<sup>140 141 142</sup>. Foster carers may not receive adequate information and support from professionals<sup>143</sup> and have highlighted the need for respite care, information and training about disabilities, and access to support services<sup>144</sup>. There is also an identified need for improved knowledge of FASD among the health care professionals who foster carers rely on for support and advocacy<sup>145 146</sup>.



Regardless, whoever constitutes the family of a child, adolescent, or young adult with FASD ideally plays an integral part in the educational and services and supports planning process, with the individual and family involved in goal setting. Further, the family also requires services and supports to assist them in caring for those in their care. The diversity of ‘families’ needs to be recognised and the views of family considered central to planning processes.

Family-centred care means that service providers join in partnership with the individual and their family to identify goals for intervention and to plan services and supports. This approach also requires that case coordinators engage in building the family’s capacity to understand systems of care, to make informed choices and to advocate for the person with a disability. This approach is congruent with the specific principles identified in the NDIS Operational Guideline-Planning and Assessment- Overview (v2.0)<sup>147</sup> in addition to the general principles of the NDIS Act 2013.

Canadian research has used the social determinants of health as a framework for intervention planning and evaluation of FASD prevention and support programs<sup>148</sup>. Potential indicators of a FASD-informed approach to planning include programs and service providers having: training in FASD; using person first language (“child with FASD”, not “FASD child”); employing a relational and strengths based approach; gearing practice to developmental age; making accommodations for communication, program format and physical environment; using an individualised care plan and one to one support; having ongoing FASD focused supervision; and, agencies being resourced to enable smaller caseloads<sup>149</sup>.

Some FASD specific family centred care programs that have been evaluated include The Step by Step program<sup>150</sup>, the Families Moving Forward Program<sup>151</sup>, the Parent-Child Assistance Program<sup>152</sup> Parent-Child Interaction Therapy<sup>153</sup>, and the Key Worker program<sup>154</sup> all of which are described in this review.

#### **6.4 FASD and trauma**

Children with FASD may have other associated life trauma, independent of brain trauma caused by prenatal alcohol exposure as the primary disability. The impact of life trauma on children’s development will differ dependent on when they have been exposed and on their temperament and existing resilience factors. Chronic childhood trauma interferes with the capacity to integrate sensory, emotional and cognitive information into a cohesive whole. Children who have suffered chronic abuse or neglect often experience developmental delays across a broad spectrum, including cognitive, language, motor and socialisation skills<sup>155</sup>.

One of the key messages to emerge in recent times is that trauma affects the whole person: their mind, brain, body, spirit and relationships with others. Evidence shows that the earlier intervention is applied, the greater the chance of recovery. Children who are neglected and abused in infancy stand the greatest chance of recovery if intervention occurs in the first year of life. The older the child, the longer they have been exposed to trauma, the more difficult it is for them to recover. However, the presence of other caring adults in the child’s life will build resilience, maintain hope, and provide a different template of possibility<sup>156</sup>. Resources such as *Calmer Classrooms* commissioned by the Child Safety Commissioner in Victoria are useful as these address the needs of children who have been traumatised by abuse and neglect. These children may be involved in the child protection and family support systems and may not be able to remain in the care of their families, instead living in foster care or other forms of state care<sup>157</sup>.

## 6.6 Early childhood intervention

The positive effect of early childhood intervention has been established in FASD like conditions, such as autism, and is optimal in addressing any brain based impairments. When there is known prenatal exposure to alcohol and indications of functional impairments early childhood intervention takes advantage of the developing brain's plasticity<sup>158</sup>.

Early diagnosis or awareness of the child's prenatal exposure to alcohol along with an understanding of their neuropsychological impairments occurring before the age of 6 years is one of the most important protective factors associated with positive long term outcomes for individuals with FASD<sup>159</sup>. Early childhood intervention has been shown to be effective in other groups of children who show developmental disabilities<sup>160</sup> including autism<sup>161</sup>, traumatic brain injury<sup>162</sup> and polydrug exposure<sup>163</sup>.

Standard best practice in early childhood intervention programs for children with other types of neurodevelopmental disabilities can be adapted to meet the specific needs of children and families affected by FASD<sup>164</sup>. Adaptations of evidence based early childhood intervention approaches for children with FASD and/or their caregivers, include:

- Offering services over a long period of time;
- Expecting slower progress in the intervention, which should include repeat, review and practice concepts;
- Using examples, modalities and treatment goals that are appropriate to the population;
- Using visual strategies;
- Considering children's sensory sensitivities and behaviour regulation problems when identifying interventions; and,
- Building on caregiver and child strengths and providing emotional support for caregivers<sup>154</sup>.

Interventions designed for children with autism spectrum disorders also provide some assistance in identifying principles relevant to planning for interventions for those affected by FASD. These include:

- Entry into treatment immediately after diagnosis/assessment;
- Creation of natural environments such as school and home, that build on strengths and compensate for deficits (from a neurodevelopmental viewpoint this means providing environmental and other accommodations for children at home and school); and
- Strong need for education and support for caregivers raising children with FASD<sup>165</sup>.

Some of the strongest evidence in early childhood intervention for children with autism spectrum disorders includes adapted behavioural techniques such as intensive, individualised behavioural treatment to improve cognitive skills<sup>166</sup> and these are likely to be useful in FASD interventions. Other approaches that have shown promise include the Denver Early Start Model<sup>167</sup>. The focus on teaching parents general principles to understand how their child learns as well as recognising the deficit areas is likely to also be applicable to FASD<sup>168</sup>. Literature on traumatic brain injury can also be used to assist with thinking about FASD and how to help parents handle the often unpredictable or challenging behaviour of children with FASD. Emphasis on positive caregiver coping skills, good psychological wellbeing, strong family relationships and a positive caregiver belief system are all needed<sup>169</sup>.

Further, established parenting and stress management interventions for children with neurodevelopmental disabilities can be used as a guide for families raising a child with FASD. Research in the broader neurodevelopmental field has revealed that multiple component interventions that

address both parental wellbeing and behavioural training together are more effective than parent training or cognitive behavioural training alone<sup>170</sup>.

In addition, FASD intervention development that focuses on helping caregivers raising a child with prenatal alcohol exposure to 'reframe' and take a 'neurodevelopmental viewpoint' is useful<sup>171</sup>. Reframing is discussed further along in this section of the review.

### **6.7 Intervention along the life course**

While early childhood intervention is optimal, interventions to improve functioning for individuals with FASD are relevant across the life course. As explained, recognition of FASD may be delayed. Additionally, individuals affected by FASD have changing needs as they grow older. For those without the identifiable FAS facial features, there are potential barriers to accessing interventions and supports because of a lack of awareness of the implications of a FASD diagnosis<sup>172 173</sup>. Further, age appropriate services are required at different phases of development, with particular emphasis on key transitional points along the life course. Interventions may be required or desirable at these key times, while not excluding interventions at other times. There are concerns shared by parents/carers and educators of children and young people with FASD about their social and emotional vulnerabilities as they move through and then away from the education system and into adult life<sup>174</sup>.

As there is currently a lack of coordinated approaches at a systems level in Australia and elsewhere to addressing the impacts of FASD, this has prompted sharing of anecdotal information and strategies through websites, online forums and other social media. These sources of information have contributed to researchers being more fully exposed to the impacts on daily functioning that FASD has for those affected as they age. It has also enriched partnerships between researchers and community members in Australia (and elsewhere) to address the problems and create a more cohesive response to diagnosis, assessment, treatment and prevention initiatives.

Good quality caregiving and stability of the home environment and environmental supports in middle childhood have been shown to reduce the likelihood by two to four times the risk of adverse life outcomes or secondary effects<sup>175 176 177</sup>. The quality and stability of the caregiving environment is regarded as a strong protective factor. Other positive characteristics that can be scaffolded for intervention, and have been identified by caregivers of children with FASD, include a willingness to participate in family activities and household routines, a strong engagement with their families and a willingness to seek and receive help<sup>178</sup>.

Further, several qualitative studies of adults with FASD have identified self-reported themes of accomplishment including: raising children and/or getting them back from foster care, graduating from school, maintaining a job, dealing with anger issues, and quitting or reducing substance use<sup>179</sup>.

Often though, children affected by FASD are raised in unstructured home environments with disrupted parental attachment. For example, it was found that for most children prenatally exposed to alcohol there were other prenatal substance exposures as well as postnatal environmental adversity<sup>180</sup>. This combination is most likely to exacerbate secondary effects associated with FASD.

### **6.8 Vulnerability at transition**

There are two aspects – cognitive and environmental – associated with transitional vulnerabilities.

Firstly, at a cognitive level, difficulties with transition refer to task and activity based transitions (moving between tasks/activities) requiring cognitive flexibility. Cognitive flexibility is in the domain of executive functioning<sup>181</sup> and is very difficult for an individual with FASD. This is reflected in their concrete thinking style and is sometimes expressed through an inability to generalise rules as well as

creating difficulties with social interactions<sup>182</sup>. Children with FASD frequently “melt down” or “shut down” before, during or after a necessary cognitive transition<sup>183</sup>. This transitional vulnerability is directly linked to the impairments associated with FASD such as self-regulation and sensory processing. It is during these daily transitions that neuropsychological impairments of FASD are most likely to show.

Secondly, transitional points are evident along the life course, for example, at the onset of puberty, and present particular vulnerabilities and difficulties for individuals with FASD and their families. Parents of children with FASD often describe the teen years as living life on a rollercoaster<sup>184</sup>. For these children the teenage years can be dominated by increased failure and less satisfaction in academic pursuits, more social isolation, and low self-esteem and depression. Planning for the transition to adulthood including the provision of social skills assistance is an important aspect in securing a pathway to employment and independent or supported living arrangements.

Further, as they transition through the school years, children and young people living with FASD are vulnerable to bullying and may encounter problems with making and keeping friends due to their social and communication difficulties and the sometimes compounding nature of their disabilities. These challenges can also be affected by co-existing disorders, such as ADHD and disruptive behaviour disorders.

The overall implications for learning is the need for extrinsic motivation to learn new skills or complete tasks such as life skills, hygiene routines, and school based tasks, particularly in secondary school-aged children. These in turn require repetitive reminders and external refocusing from supervising adults to keep children and young people on task<sup>185</sup>.

The transition to adulthood for individuals with FASD should be viewed as a transition to “interdependence” rather than necessarily a transition to independence<sup>186</sup>. Many individuals with FASD may not be prepared for independence and there are often deleterious outcomes, such as difficulties obtaining meaningful employment and being incarcerated, discussed by families<sup>187</sup>. The prevalence of FASD is considerably higher for certain subgroups, and is alarmingly high for at risk groups, such as juvenile offenders<sup>188</sup>. Surveys have found that between 15 and 20 per cent of juvenile and adult offenders have FASD, but this is generally considered to be an underestimate. Furthermore, one consistent finding is that the social incompetence of young people with FASD becomes even more marked and disabling as they enter adolescence and adulthood<sup>189</sup>. Another important point to consider in transition planning is related to youth with FASD when they transition out-of-care, a particularly vulnerable and difficult time for these individuals and their families<sup>190</sup>.

Recommendations for transition planning should include attention to the planned living arrangements for the future, eligibility for an extension of supportive care, an assessment of independent living skills, referral to appropriate resources to develop life skills, and plans for education and vocational training<sup>191</sup>.

Useful transition strategies for caregivers and health professionals working with children and youth with FASD have been developed for the Canadian context<sup>192</sup>. For example, programs such as the Cold Lake project have developed transition plans for young people moving from youth to adult services and produced tools for employment support agencies to work with people with FASD<sup>193</sup>.

## **6.9 Destigmatising and cognitive ‘reframing’**

As noted, lack of awareness of FASD is an overarching construct interrupting systems responses for those affected<sup>194</sup>. Another aspect of FASD is the stigmatisation which often accompanies diagnosis or assessment of FASD as the primary disability. This includes addressing the impact of prenatal alcohol

consumption where there is long term alcohol abuse and supporting the family to address this issue alongside identifying the services and supports required to support the individual with FASD.

Raising a child with FASD is very different when compared to other developmental disabilities. FASD carries an emotional overlay, because it is a birth defect that could have been prevented and the disruptive influence of parental substance abuse will always be part of the family history<sup>195</sup>. Stigmatisation associated with FASD may prevent families from accessing vital services if care is not taken with how this is managed by medical practitioners, other health professionals and support workers.

A basic intervention for caregivers and professionals involved with the child with FASD has been described as 'reframing'<sup>196 197 198</sup>, and is central to family-centred FASD intervention. This process of cognitive (and emotional) reframing is important for caregivers and professionals involved with the child to address the behavioural difficulties of their child by understanding that the behaviours are brain-based. Reframing is especially vital when children or young people with FASD have externalising behavioural difficulties. One program in which reframing is specifically addressed is the Families Moving Forward Program<sup>199</sup>, and this program is especially useful for the high needs group of families raising preschool and school-aged children with prenatal alcohol exposure and externalising behaviour. The process of reframing can assist parents to understand and accept the child's difficulties, improve the parent-child relationship and provide more effective outcomes from intervention.

#### **6.10 Environmental Accommodations**

In designing interventions, children with FASD need opportunities to learn and build skills that will help them regulate their emotions and behaviours as well as environmental accommodations that increase the likelihood of adaptive behaviours. It has been suggested that an individual with FASD may need an '*external brain*' to assist with staying on track due to the nature of their executive dysfunction<sup>200</sup>. The external brain term, while perhaps not ideal, does highlight that caregivers need to provide ongoing support in the areas of executive functioning and emotional and behavioural control. This support may come in the form of environmental modifications, making abstract rules more concrete, teaching individuals to ask for help and providing ongoing supervision and prompts for self-monitoring. An individual's environment has also been referred to as an '*external nervous system*' to emphasise the importance of structure and explicitly teaching functional routines to provide a scaffolding framework<sup>201</sup>. This can be achieved by utilising visual structures, which compensate for executive functioning deficits in set shifting, working memory and attention. The use of detailed visual schedules and a detailed visual representation of rules that cue appropriate behaviour are recommended.

As children with FASD demonstrate deficits in their social functioning it may be important for a child to learn explicit social interaction steps, through rehearsal, modelling, and coached practice with feedback based on the basic rules of social behaviour<sup>202</sup>. These skills can represent an important target for intervention, because poor peer relationships are predictive of early withdrawal from school as well the development of later mental health disorders and comorbidities<sup>203 204</sup>.

Children with executive functioning difficulties specifically in working memory may be unable to follow directions, retain information that has been presented or generalise information from one situation to another<sup>205</sup>. Organising and keeping track of personal belongings at school, or performing a task through to completion requires understanding that a task has a beginning, middle and end. Visual and external structure strategies are particularly useful compensatory tools to aid deficit areas. Visual or

picture schedules have been shown to be effective with children with autism<sup>206</sup> as in the form of a calendar or daily planner which may help alleviate anxiety during daily points of transition between tasks and give a concrete reference to the structure of the day. Planning frameworks can reduce stress during transition times which have been identified as a difficulty in individuals with FASD<sup>207</sup>.

In addition, sensory processing difficulties (SPDs) often experienced by children/young people with FASD can lead to inattention, hyperactivity and distractibility. These challenging behaviours triggered by sensory processing difficulties can be assisted by strategies that include: subdued lighting, minimising noise and distraction, the consideration of textures and smells, and where a child/young person is seated in the room. Colour coding items and trays related to specific tasks assist with maintaining attention on the task as well as minimising distraction. Use of earphones, calming music and masks can be incorporated into a calm space where children can visit when they become overwhelmed. An example is the 'Marulu room', a quiet space with minimal distractions in the Fitzroy Valley District High School<sup>208</sup>.

Reinforcement and reminders of routines and activities of daily living are helpful<sup>209</sup>. Likewise executive functioning deficits are directly linked to challenging behaviours which are particularly evident in the school setting and negatively affect a child's ability to learn and function successfully in the school environment<sup>210</sup>. It is important that the behaviours which may have been labelled as "defiant", "lazy" or "manipulative" are recognised as symptoms of the underlying brain based impairments<sup>211</sup> and are reframed by all those involved with a child or young person with FASD.

Further, children may need more domain specific cognitive problem solving strategies as task complexities increase<sup>212</sup>. The difficulties with increases in task complexity are further evident in the learning of sequences such as in arithmetic which are a hallmark difficulty for those with FASD<sup>213 214 215</sup>. Arithmetic involves an understanding of the relationship between the order and function of numbers and the steps required to solve a problem, a more complex cognitive function involving working memory. An example of a functional skill involving arithmetic is the management of money with which individuals with FASD have significant difficulty.

A future direction for interventions may involve combining caregiver support with targeted interventions that relate to specific neurobiological constructs. For example, an approach that integrates the home and school environment may include family focused interventions (e.g. the Families Moving Forward program) combined with self-regulation programs (e.g. The Alert® program)<sup>216 217</sup>.

### **6.11 General recommendations**

Finally, there are general recommendations useful to families with children with FASD that aid in behaviour management and the promotion of cognitive and social skills<sup>218 219 220 221</sup>.

For example, *8 Magic Keys: Developing Successful Interventions for Students with FAS* is referred to frequently in the literature and was developed from the work of Evensen and Lutke<sup>222</sup>. As part of the cognitive reframing of FASD and its implications, 8 Magic Keys provide succinct explanations of eight areas of daily functioning. These are outlined in Box 1.

Key factors have been identified as relevant to the development of interventions which address individual needs, use a neurodevelopmental framework and provide a starting point for a 'best practice' framework<sup>223</sup>.

- *Pay attention to the child's overall cognitive-behavioural profile when designing an intervention program for him or her.* Cognitive dysfunction has been extensively studied

within FASD, with several patterns and areas of dysfunction found to exist. The more complex the task the more difficulty the child with FASD has when two or more domains of cognitive functioning are required to complete that task. It is important to incorporate strategies such as presenting the information at a slower rate than normal, using multimodal and repetition of information. Hands on experiences rather than verbal instruction facilitates encoding of information, such as demonstrated in utilisation of computer games to teach materials<sup>224</sup>.

- *Use strategies from child's zone of proximal development (ZPD).* Managing a child/young person at their developmental age/level rather than what is expected of them based on their chronological age.
- *Provide attention and self-regulation training as early intervention.* Attention training has been shown to be generalizable across other domains of learning. Self-regulatory skills which emerge during the preschool years are particularly indicative of long term outcomes. Specific self-regulatory skills are strongly associated with academic outcomes. Attentional skills allow children to encode information efficiently as they are able to sustain attention effectively in the presence of interference and then hold more information in their working memory<sup>225 226</sup>. Targeting attention and self-regulation (domain general) may prove to be more effective than utilising domain specific interventions such as literacy or math training.
- *Provide enriched input in a guided fashion.* An enriched environmental input enhances cognitive functioning in humans and animals and preliminary evidence to support this idea in FASD has been found<sup>227 228</sup>.
- *Suggestion that evidence based behavioural and pharmacological intervention is most effective when used in combination.*
- *Family in partnership approach.* The family + school + therapy partnership approach is needed to ensure consistency in the child's environment. This can be achieved by working with the family and school teams, observing the child's skills and capacity in maintain attention, independence, social interactions, functional language, and their strengths, interest and behaviour across multiple observations.

Other useful strategies such as cognitive behavioural therapy and interventions utilised for children with ADHD can assist with intervention planning for FASD as these strategies specifically address executive functioning deficits such as those seen in children prenatally exposed to alcohol. Intervention planning recommending the use of visual cues and schedules, teaching of self-directed speech and problem solving, social skills training, role play, cognitive modelling and coaching to support executive functioning difficulties in the areas of nonverbal memory, verbal working memory, self-regulation of mood, motivation and level of arousal and problem solving have been proposed for interventions with FASD<sup>229</sup>.

Further, to support children with FASD to function effectively in the classroom, intervention planning should consider strategies and ideas such as evident in Positive Behaviour Support (PBS), a framework that is well supported and utilised in disability organisations in Australia. The PBS model for intervention is a collaborative team approach that is focused on understanding the function or communicative intent behind the problem behaviours, as well as the contributing environmental events and then teaching more appropriate and functional behaviours<sup>230</sup>.

### ***Summary of Interventions***

The evidence base for effective interventions is growing in the field of FASD. Targeted interventions addressing specific strengths and needs of children with FASD such as attention, social communication and behaviour will significantly enhance the evidence base available to inform management of FASD. Both domain specific and domain general approaches have been identified as relevant when considering appropriate interventions. These should be integrated across the individual's environments.

Effective interventions must consider the interplay between behavioural symptoms and the neuropsychological effects of prenatal alcohol exposure. Executive functioning deficits are directly linked to challenging behaviours, and are highly likely to be present in an individual with FASD. Together with the whole neurodevelopmental profile, these deficits in daily functioning may require multiple interventions, or a variety of interventions provided at different time points.

It is vital that interventions using a variety of strategies to teach the child new skills are integrated with the use of extensive environmental accommodations to support the use of new skills.

Multiple component interventions addressing parental wellbeing and behavioural parent training are thought to be more effective with the complex set of issues found in FASD.

Emerging intervention programs (addressed in the following section) have been identified as showing promise with other developmental disabilities, Indigenous communities and in other countries with similar geographical diversity. This emerging evidence base requires ongoing review of available interventions to ensure that innovation in the field is incorporated into disability planning processes in the future.



## Box 1: 8 Magic Keys

The 8 Magic Keys are strategies which underpin successful development of interventions for students with FASD. These are simple, functional strategies to use with young people with FASD and can be used by caregivers, teachers and health professionals. They were developed for use by the FASD Centre for Excellence, Substance Abuse and Mental Health Services Administration (SAMSHA.GOV) in the USA.

- 1. Concrete Terms** *Children living with FASD do well when parents/carers and educators talk in concrete terms. Refrain from using words with double meanings, idioms, etc. The social emotional understanding of children living with FASD is often below their chronological age, therefore it helps to 'think younger' when providing assistance, giving instructions, etc. It is also important not to make deficit judgements*
- 2. Consistency** *Due to the difficulty that children with FASD experience in generalising learning from one situation to another, they do best in an environment with few changes. This includes consistency of language and routines. Educators and parents/carers should coordinate with each other to use the same words and or gestures for key phrases. Communication books are effective ways of sharing what's happening and advising on language use and behaviours in classrooms and homes.*
- 3. Repetition** *Children with FASD have chronic, short-term memory problems. They forget things they want to remember, as well as information that has been learned and retained for a period of time. In order for them to commit something to long-term memory, it often needs to be repetitively re-taught.*
- 4. Routine** *Stable routines and consistent visual cues that do not change from day to day make it easier for children with FASD to know what to expect next, and decrease their anxiety, enabling them to learn*
- 5. Simplicity** *Remember to keep input short and sweet. Children with FASD are easily over stimulated, leading to 'shutdown', at which point they can take in no more information. Break down tasks and always communicate the task in the positive: 'we walk inside' instead of 'don't run!'*
- 6. Specific Language** *Say exactly what you mean. Remember that children with FASD have difficulty with abstractions, generalisations and 'filling in the blanks' when given an instruction. Tell them step-by-step what to do. This will help them develop appropriate habit-forming patterns. Keep instructions concise and broken into achievable chunks.*
- 7. Structure** *Is the 'glue' that enables a child with FASD to make sense of the world. If this glue is taken away, things fall apart. A child with FASD achieves and is successful because his or her world provides appropriate structure as a permanent foundation for learning.*
- 8. Supervision** *Due to their cognitive challenges, children with FASD bring a naivety to daily life situations. They need constant supervision, as with much younger children, to develop habit patterns of appropriate behaviour and ensure safety and wellbeing at all times.*

**Table 1: Intervention Programs**

Intervention	Domain/s targeted	Target group	Who delivers	Delivery mode	Time frame	Where
<b>DOMAIN SPECIFIC STRATEGIES</b>						
<b>MILE program</b> Kable, Coles & Taddeo 2007 <sup>231</sup> Coles, Kable & Taddeo 2009 <sup>232</sup> Kable et al, 2015 <sup>233</sup> Kable et al, 2012 <sup>234</sup>	Learning strategies to compensate neurodevelopmental deficits (Maths) Case management and caregiver education	Children 3-10 year olds Parents	teachers or psychologists or occupational therapists or speech pathologists and parents	Individual (child) Group (Parent) or Parent component Mixed face to face and web	6 weeks Follow up: 6 months  15 weeks	clinic or community  or web based delivery parent education
<b>Language and literacy training</b> Adnams et al, 2007 <sup>235</sup> Timler et al, 2005 <sup>236</sup>	Language (pre literacy, reading and spelling)	9 to 10 year olds	speech pathologist	Mixed: individual and group face to face	60 min duration over 9 months Follow up: 9 months	school or clinic
<b>Cognitive control therapy (CCT)</b> Adnams et al, 2003 <sup>237</sup>	Attention, information processing also body position, movement and awareness	8 year olds	psychologist or occupational therapist	Group Face to face	60 mins per week for 10 school months	school
<b>Working memory strategies</b> Loomes et al, 2007 <sup>238</sup>	Working memory rehearsal (executive functioning)	Children 4 to 11 year olds	psychologist or occupational therapist	Group face to face	Rehearsal training (follow up: Range 6-21 days)	clinic or school or community
<b>Computerised attention training (CPAT program)</b> Kerns et al, 2010 <sup>239</sup>	Attention Working memory	6 to 15 year olds	psychologist or occupational therapist	Individual Face to face	9 weeks 16 hours (30 min duration) (average 30.5 sessions)	school or clinic
<b>DOMAIN GENERAL APPROACH</b>						
<b>The Alert program® (Neurocognitive habilitation)</b> Wells et al, 2012 <sup>240</sup> Nash et al, 2015 <sup>241</sup>	Executive functioning through self-awareness of self-regulatory skills	Children 5-12 year olds and parents	occupational therapists, social workers, psychologists	Group Face to face	12 weeks 75 minute duration Follow up: 6 months	school or clinic

**Table 1: Intervention Programs (cont.)**

Intervention	Domain/s targeted	Target group	Who delivers	Delivery mode	Time frame	Where
<b>SOCIAL SKILLS INTERVENTION</b>						
<b>Children’s Friendship Training (CFT)</b> O’Connor et al, 2006 <sup>242</sup> O’Connor et al, 2012 <sup>243</sup>	Social skills (peer social interactions)	Children 6-12 year olds	Speech pathologist, occupational therapists, psychologists, Parents as facilitators	Group Face to face	12 weeks Weekly 90 min duration	clinic or school or community Follow up: 3 months
	And parent sessions	Parents				
<b>CFT with psychopharmacological management</b> Frankel et al, 2006 <sup>244</sup>	Variety of grouped medication with CFT vs no medication and CFT	5 to 11 years	Clinical psychologists			
<b>PARENT &amp; CHILD/FAMILY INTERVENTION</b>						
<b>Family support and management</b> <b>Coaching families program</b> Leenaars et al, 2012 <sup>245</sup>	Stress reduction, advocacy, referrals to families of children with FASD	<1 to 23 year olds	Case manager	Individual	Varied depending on needs of family. Months to years	community
			Coordinates	face to face		
<b>Parent child interaction therapy (PCIT) and parenting support and management</b> Bertrand, 2009, (Study 4) <sup>246</sup>	Parent-child relationship (reduce parenting stress and target child behaviour )	3 to 7 year olds	Case Manager delivers and coordinates	Group face to face	12 weeks 90 min duration 2 weeks Children/ parents separate sessions	community
<b>Families Moving Forward Program</b> Bertrand, 2009 (Study 5) <sup>247</sup>	Family intervention (meet family needs, reduce child problem behaviours)	5 to 11 year olds	Case Manager coordinates	Family Face to face	9-11 months 16 fortnightly sessions 90 min duration	community

**Table 1: Intervention Programs (cont.)**

<b>Intervention</b>	<b>Domain/s targeted</b>	<b>Target group</b>	<b>Who delivers</b>	<b>Delivery mode</b>	<b>Time frame</b>	<b>Where</b>
<b>Parent-Child Assistance Program (PCAP) Remodelled</b> Grant et al, 2004 <sup>248</sup>	<b>Modify PCAP for clients with FASD</b> <b>Educating community service providers</b>	<b>Young women (mothers with a child with FASD)</b>	<b>Case manager delivers</b>	<b>Individual</b> <b>Face to face</b>	<b>12 months (begin during pregnancy or early post partum)</b>	<b>community</b>
<b>Step by Step program</b> Denys et al, 2011 <sup>249</sup>	Assist parents to access supports in community Parenting and life skills	Parenting adults (majority Aboriginal/female)	Case manager delivers	Individual Face to face	~ 3 years 2 to 4 goals set for 6 months	community
<b>BEHAVIOUR BASED EDUCATIONAL STRATEGIES</b>						
<b>Virtual reality intervention</b> Padgett et al, 2006 <sup>250</sup>	Fire safety (behaviour based) Generalise to real world	Children 4 to 7 year olds	Psychologists, occupational therapists	Group face to face	2 visits follow up: 1 week	clinic
<b>Computer game intervention</b> Coles et al, 2007 <sup>251</sup>	Attention Fire and Street safety (behaviour based) Generalise to real world	Children 4 to 10 year olds	Psychologists, occupational therapists	Group face to face	Virtual reality computer game with follow up: 1 week	clinic

## 7. Emerging Evidence

### 7.1 Emerging family interventions

As awareness of FASD improves and research is focussed on developing interventions to address the significant areas of functional impairment, programs are being developed and/or refined and evaluated for effectiveness. As such, this critical review has included information relevant to programs with emerging evidence for those with FASD, especially those related to supporting families. These are summarised here, with further details provided in Table 11, Appendices.

**7.1.1 The Strongest Families Research Program** has been developed by the Nova Scotia Centre for Research in Family Health at the IWK Health Centre, Canada. The program uses input from key stakeholders to determine the need for services and supports and develop and evaluate a distance training program for parents/caregivers. This program eliminates the need for families to travel to an intervention site on a regular basis, as the intervention can be completed in families own homes and participants can be recruited from across the country. The Strongest Families program has shown success with children with behaviour and anxiety disorders as well as night time bedwetting with results from randomised trials indicating effectiveness one year later targeting mild and moderate conditions<sup>252</sup> and will be adapted for families with FASD in a randomised control trial to be conducted in Canada<sup>253</sup> (see Appendices for further details).

**7.1.2 Triple P Stepping Stones Parenting** in Australia has shown promise in reducing child behaviour problems, improving parenting styles and decreasing parenting stress with parents of children with developmental disabilities<sup>254 255</sup>. The Triple P Parenting program is designed for parents of children aged 2 to 7 years of age. It has also been tested with families raising children aged 2 to 9 years with developmental disabilities<sup>256</sup> and autism spectrum disorders with behavioural gains maintained at 6 month follow up<sup>257</sup>.

Additionally, Triple P has been tested and shown to be effective in many culturally and ethnically diverse populations around the world<sup>258 259 260</sup> including with Indigenous Australians<sup>261</sup>.

**7.1.3 The Circle of Security**<sup>262</sup> is a group mental health intervention that is designed to help caregivers provide a 'safe base' or safe haven for their child. It is focussed on the parent-child relationship. It provides psycho-education and psychotherapy for families at high risk, and based on attachment theory. It has been empirically evaluated with toddlers and their parents, with results reporting improvements in attachment behaviour<sup>263</sup>. Further studies have been completed in a jail diversion program<sup>264</sup> and with a home visitation program<sup>265</sup> for infants and their mothers deemed at high risk.

There is an attachment related clinical trial currently underway utilising the Circle of Security Intervention with preschool children aged 2-5 years with FASD, or who are at risk for FASD, and their caregivers in Manitoba, Canada<sup>266</sup> (see Appendices for further details).

#### **7.1.4 Multidimensional treatment for preschool children in foster care**

The Early Intervention Foster Care (EIFC) for pre-schoolers, originally developed by Fisher and colleagues (1999)<sup>267</sup>, is a model currently being applied with children in foster care, although has not been explicitly used with children with FASD. It is expected to improve short and long term outcomes for children in foster care, and predict school behaviour, long term placement stability, and mental health status. The project targets behaviour problems, physiological dysregulation and developmental delays. A team approach is utilised with multiple layers of intervention with caregivers receiving specific training and consultation from professionals, alongside interventions for the children.

Preliminary research has indicated that when compared to regular foster care, attachment behaviours were found to improve, with increased secure behaviours and a reduction in avoidant behaviour<sup>268</sup><sup>269</sup>. Potentially this is a promising program that could have a place with foster families of children with FASD.

The rate of FAS has been reported to be 10 to 15 times higher for children in foster care than in the general population<sup>270</sup>. The high percentage of children with FASD in foster care represents another challenge when designing and planning interventions as the children often experience placement instability and disruptions to the caregiving environment<sup>271</sup>.

**7.1.5 The Lakeland Centre for FASD** in Alberta, Canada, is a good example of communities supporting individuals with FASD. It is a unique, not-for-profit organization based on the principles of interdependence, lifelong support, harm reduction, prevention of problems, and respect. The Centre provides mobile, multidisciplinary diagnostic and assessment services for children and adults and functions as a bridge to local community services. Every person diagnosed in the clinic is linked to a lifelong outreach worker and to a local system that follows through on the diagnostic team's recommendations and addresses any other issues that arise. Mothers who are in a cycle of drinking and having babies are supported with a mentor in the evidence based Parent-Child Assistance Program (PCAP) who works long-term and intensively in an outreach capacity to support women to be alcohol free during pregnancy and beyond. In addition, the centre provides training and education, organizes numerous awareness campaigns and information-sharing activities and has participated in community development in FASD around Canada<sup>272</sup>.

## **7.2 Emerging cognitive and behavioural interventions**

### **7.2.1 GoFAR - Intervention for Affective and Metacognitive Control in FASD**

The GoFar program focuses on a core area of impairment identified in FASD; which is attention and arousal. It is designed to improve children's self-regulation and adaptive living skills (i.e. social function, daily living skills, communication). GoFAR is developing methods to address these problems in young children aged 5-10 years of age, by bringing together computer game technology and behavioural techniques for affective and cognitive control<sup>273</sup>. The program aims to teach children and their parents strategies to improve children's self-regulation while learning basic everyday life skills.

Two projects funded by the USA Centre for Disease Control from 2009 included:

#### **7.2.2 Project Step Up**

This research was aimed at 13-18 year olds and was designed to provide parents with tools to facilitate their teens' difficult transition to adulthood and to better understand the neurocognitive deficits associated with prenatal alcohol exposure and alcohol misuse in adolescents. The aims were to decrease in alcohol risk scores and alcohol related negative behaviours in the intervention compared to the control group. Outcomes reported that adolescents in the intervention group had lower levels of alcohol use and fewer high risk behaviours. However, intervention affects were not maintained at the 3 month follow up<sup>274</sup>. Further details are provided in Table 10.

#### **7.2.3 Partners for Success**

The project aimed to reduce maladaptive behaviours among adolescents and young adults (16 through 25 years of age) with FASD. The therapy anticipated to reduce behaviours for participating youth with FASD, reduce stress among their families, and improve the quality of life for all participants. The intervention used a team of clinicians, therapists, and mentors. It was anticipated that Partners for Success would provide an evidence-based intervention to increase parenting skills and reduce the occurrence and severity of maladaptive behaviours for young adults with FASD and improve the quality of life for them and their families<sup>275</sup>. This is still listed as a current project and no outcomes have been reported. Further details are provided in Table 11.

With increased access to technology, gaming platforms have increasingly emerged in the FASD interventions field.

#### **7.2.4 Computer game interventions for FASD**

Gaming platforms have been explored by several researchers as a novel approach to managing and improving the symptoms associated with FASD<sup>276</sup>. Some of these have already been included in Table 1, as evidence based examples. Others include Cognitive Carnival<sup>277</sup> (further details in Table 11) and Caribbean Quest<sup>278</sup> which involve research partnerships with NeurodevNet, a Canadian Network of Centres of Excellence. These interventions hold significant promise for addressing many of the complex neurological challenges associated with FASD. They can be easily customised to meet the needs of each individual and can adjust for changes in brain function that may occur across the lifespan.

Finally, other new approaches to intervention research in FASD include mindfulness, neurofeedback<sup>279</sup> and aerobic exercise<sup>280</sup>.

### **7.3 Promoting Integration**

When planning interventions, integrated responses to the delivery of services across multiple agencies and professionals are required. Integrated care models allow more effective coordination to manage the complex medical and behavioural health care needs of children and adults with FASD<sup>281</sup>. An integrated case management model is important for ensuring that children and young people with FASD, and their families/caregivers, have access to an ongoing suite of interventions and supports. One example, the Key Worker model, has been provided to indicate the principles involved in an integrated approach to managing life trajectory planning for FASD services and supports.

#### **7.3.1 Key Worker and Parent Support program**

The BC Key Worker and Parent Support program was implemented in British Columbia, Canada in 2005. Its overall goal is to maintain and enhance the stability of families with children or youth with FASD and/or similar complex developmental conditions. The program is based on research and practice evidence from three areas: research in FASD such as that conducted by Streissguth and colleagues<sup>282 283</sup>, Olson<sup>284</sup>, and Malbin<sup>285</sup>; research in the disabilities field; and, community based, FASD parent support projects that were in operation. At the heart of the program is an understanding that FASD is a brain based physical disability with behavioural symptoms.

Key components and characteristics of the program include:

- Regional service delivery model that allows for regional variation and modifications to meet unique local needs and family circumstances.

- Availability of an expert in FASD to provide consultation to regional offices and contracted agencies, in order to help facilitate learning and strategies related to use of appropriate environmental accommodations.
- The Key Worker is a facilitator who assists parents, family members, caregivers and service providers in the child's environment to come to a common understanding of the child's/youth's needs and to develop supportive environmental accommodations accordingly.
- Key Worker is supervised by a qualified professional.

The Key Worker and Parent Support program is based on a case management model that provides consistency and a central contact point. As interventions, supports and services are likely to include multiple agencies, a coordinated approach organised by a lead agency is needed in the Australian context.



## 8. Severity Guidance

The critical review has aimed to determine from the literature and advice from the Expert Review Panel, levels of severity associated with FASD and how this may link to an intervention severity index. However, no clear evidence has emerged to inform a severity index. Rather, international and Australian diagnostic criteria indicate a picture of impairments that are complex and unique to each individual. As such, a severity guidance framework has been developed to guide NDIA approaches to determining service and support needs for individuals affected by FASD.

### FASD and Disability Support

Confirmed diagnosis on the FASD spectrum\* based on current criteria.

(\*using Australian FASD diagnostic guidelines)

Documentation of impairments in the absence of diagnosis (with or without confirmed PAE) by qualified professionals using standard validated instruments#.

(#instruments appropriate to age, literacy and culture)

#### SEVERITY GUIDANCE

The range of brain based impairments, and the degree of impairment severity, varies greatly for individuals with FASD. Impairment is established using standardised assessments (where feasible) within a multidisciplinary team, and defined as 1.5-2SD (with clinical concern), or  $\leq 2$ SD from the mean. Impairments will likely alter over the life course, requiring reassessment at key transitional times. In infancy, a lower threshold for disability support (e.g. impairment in 2 of the brain domains) is recommended as early intervention is crucial in improving later life outcomes.

##### Indicators for disability support planning:

Impairment in a minimum of 3 or more of the following brain domains:

Attention/activity level/sensory processing  
Academic achievement  
Adaptive behaviour/social skills/social communication  
Cognition (or global development if < 5 years of age)  
Executive functioning  
Language (expressive and receptive)  
Memory  
Fine or gross motor function  
Affect regulation (e.g. DSM-5 criteria for major depressive disorder, social anxiety disorder etc)  
Structural abnormality/microcephaly/hard neurological signs/seizure disorder

OR impairment in 2 or more of the functional domain categories proposed in the DSM-5 diagnostic criteria:

Functional domain categories:

- 1) Neurocognitive functioning, includes -  
*a] cognition; b] executive functioning; c] academic achievement; d] memory;*
- 2) Behaviour and Self-regulation functioning, includes -  
*a] attention/hyperactivity/sensory processing/impulse control; b] affect and behavioural regulation*
- 3) Adaptive functioning, includes -  
*a] language; b] social communication & interaction; c] daily living skills; d] motor skills.*

##### Functional severity indicators\*\*:

- **Moderate (requiring substantial support)** – impairments falling into two of the three functional domain categories.
- **Severe (requiring very substantial support)** – impairments falling into three of the three functional domain categories.
- **Profound (requiring constant supervision and support)** – impairments falling into three of the three functional domain categories including intellectual disability (IQ<70) and severe adaptive functioning impairment.

\*\*This is a guide only and should be interpreted with clinical consideration based on multidisciplinary assessment. For example, it is feasible that assessment may demonstrate 3+ impairments as severe while falling into only two functional domain categories.

## 9. Discussion

This critical review of the literature has highlighted the complexities associated with implementing a systematic disability planning approach to assist individuals affected by FASD access the services and supports they require.

The reviewed literature has included published journal articles, unpublished reports, information obtained through websites and other material provided by members of an Expert Review Panel and a Consultative Group to present a comprehensive picture of FASD relevant to the Australian context.

As such, the review has focussed on: the domains of functional impairment; interventions designed to address impaired functionality in these domains; and models of care designed to provide individuals and their families/caregivers with integrated and coordinated supports and services that improve daily functionality.

As a permanent and lifelong disability, planning supports for children, adolescents and adults with FASD requires a clearly articulated approach to enable entry to and exit from supports and interventions as required and at key transitional times along the life course trajectory. Such an approach enhances individual capacity to participate in education, employment and social activities.

The relatively recent recognition of FASD as a primarily brain based disability of varying severity, combined with issues associated with achieving a diagnosis and the need for multiple assessments at multiple time points across the life course to identify required interventions, have all been taken into account in the formulation of best practice indicators. These will provide the basis for the formulation of best practice guidelines to guide future disability planning processes.

To begin, each individual with FASD requires consideration of their unique needs. Ideally this starts with a multidisciplinary diagnostic or assessment process. The diagnostic pathway will most likely be implemented when cardinal FASD indicators (such as facial characteristics) are present, ideally leading to early childhood intervention as an optimal response. Alternatively an assessment pathway may occur when a child or young person deviates from a typical developmental trajectory. The point at which FASD is first recognised requires age and culturally appropriate assessments to be used. In this context, other factors may also be relevant. These include cultural considerations and the location of residence, both of which may limit access to services and supports. Innovative responses might therefore be required to maximise the opportunities for people with FASD to participate in their local community and the broader society.

Neither a specific diagnosis (FAS/pFAS/ND-AE) on the FASD spectrum, nor assessment that leads to recognition of FASD as the primary cause of a person's disability, necessarily indicates a clearly 'matched set' of functional impairments. Rather, each person affected by FASD will have an individual neurocognitive profile arising from the gestational timing and 'dose' of their prenatal alcohol exposure and the types of other risk factors which may be present, such as childhood trauma or neglect.

Relevant assessments of the domains of neurodevelopmental impairment associated with FASD are not reliant on a diagnostic entry point. Other factors may lead to a range of assessments being used to identify each individual's primary disabilities arising from FASD. In the current circumstances, with a broad lack of awareness of FASD across the whole society including health professionals, a child with FASD may first be identified when they begin formal education or during their primary school years.

Once conducted, assessment must then lead to referrals to relevant interventions, services and supports to facilitate optimal daily functioning. This potentially includes the use of technologies, such

as virtual systems, to promote access and create equity. In addition it is essential that the NDIA consider the variety of family types that care for children with FASD, and particularly the needs of foster families, as it is common for children with FASD to be placed with foster carers.

A comprehensive profile of functioning determines the specific needs of the individual's level of functioning in academic, cognitive and behavioural learning areas. Multidisciplinary approaches leading to goal setting and individualised plans which take into account various universal settings and everyday environments, and the skills needed to manage these, is required. The caregiving environment (or what the child constitutes as their family) is central in guiding the support system for the child with FASD. A *coordinated family + school + therapy partnership* approach, or community agencies for those who have left the school system, supported through a case management or key worker model is needed to ensure consistency in the individual's lived environment.

From the review of the literature, principles which support planning approaches have been identified. These are set out below.

## **9.1 Planning Principles**

Principles identified from the critical review are set out as these are relevant to: general planning principles; and, those principles which underpin access to supports and services or assist in identifying interventions specific to FASD (or draw on best practice for FASD like conditions). These principles are followed by best practice indicators to inform the future development of best practice guidelines.

### **9.1.1. General planning principles**

Comprehensive services for individuals with FASD require the involvement of the individual, family members, and multiple systems of care<sup>286</sup>. A learning profile specific to the child or adult is particularly useful for intervention planning given the heterogeneity of CNS effects.

- A focus on promoting participation and highlighting the whole child within the context of development in the home and community, rather than on attempting to address children's deficits or impairments<sup>287</sup>.
- As standard practice most individuals with FASD benefit from multiple types of intervention services including occupational therapy, speech and language therapy, behaviour therapy, physiotherapy, individualised education plans etc., with interventions changing over time depending on the number, type and severity of FASD associated functional impairments.
- Capacity building for families and wrapping services around the child's needs:
  - Considering the current Early Childhood Intervention best practice principles<sup>288</sup>.
  - Helping parents and caregivers to identify available intervention services (e.g., through the school or community).
  - Working with parents and caregivers to ensure access to appropriate services, including skills of daily living and vocational training (e.g., personal hygiene, money management).
  - Informing parents about the education laws pertaining to children with FASD, and assisting families to negotiate individualized education programs.
  - Offering services over a long period of time.
  - Expecting slower intervention progress – repeat, review and practice concepts are a key feature of interventions.
- Build on caregiver and child strengths, and, provide emotional support for caregivers.

- Reduce stigma and shame for parents whose children may be diagnosed with FASD.
- Acknowledge the challenges of parenting a child with FASD and encourage parents and caregivers to pay attention to their own needs.
  - Help families access respite care to reduce family stress and improve family functioning;
  - Provide resources (e.g.; parent management training and parent counselling, parent support groups); and
  - ‘Reframe’ FASD for caregivers and professionals involved with the child, e.g. educating how behavioural difficulties are ‘brain-based’.
- Assess family members for mental health problems, substance use and addiction and help them obtain treatment.
- Determine the risk for child abuse, neglect, and violence in the family and provide referrals as indicated.

### **9.1.2 Transition planning principles**

Transition to adulthood for individuals with FASD should be viewed as a transition to ‘interdependence’ rather than necessarily a transition to independence<sup>289</sup>, and includes:

- Young people moving from youth to adult services require tools to enable employment support agencies to work with people with FASD.
- Attention to the planned living arrangements for the future.
- Eligibility for an extension of care.
- An assessment of independent living skills.
- Referral to appropriate resources to develop life skills.
- Plans for education and vocational training<sup>290</sup>.

### **9.1.3. Cultural Security principles**

There are a range of factors relevant to planning process for services and supports in Indigenous contexts. These include: embedding programs and resources in community controlled health organisations and Indigenous people being directly employed in delivery of interventions. Other principles from the broader health service delivery sphere are relevant here including: consultation with local communities; being sensitive to local conditions, in particular the need to avoid blame and shame; and ensuring that local knowledge is used to develop plans which include respect for cultural protocols. Further, the consideration of the social determinants of health and the lived histories and impact of intergenerational trauma need to be taken into account. Strengths based approaches are likely to be very relevant to intervention planning.

There is a need to consider access to services and supports in remote communities in particular. An integrated approach, inclusive of all relevant services (e.g. local health, education and community support services) provides collaborative training and awareness raising opportunities. Further, existing distance health services such as telehealth should be considered as well as virtual collaborative processes, such as those currently being used in the Canadian context.

### **9.1.4. Intervention principles**

Earliest possible diagnosis or assessment leading to multidisciplinary assessment with referral to relevant interventions is foundational to addressing functional impairments<sup>291</sup>.

The following principles underpin successful interventions:

- Provide **attention** and **self-regulation training** as early intervention. Attention training has been shown to be generalizable across other domains of learning. Self-regulatory skills which emerge during the preschool years are particularly indicative of long term outcomes. Specific self-regulatory skills are strongly associated with academic outcomes.
- Family in partnership approach. The *family + school + therapy* partnership approach is needed to ensure consistency in the child/young person's environment. This can be achieved by working with the family and school teams, observing individual's skills and capacity in maintaining attention, independence, social interactions, functional language, and their strengths, interests and behaviour across multiple observations.
- Parent education or training alongside interventions for the child/young person.
- Consider parental cognitions/attitudes and problem focused management as a useful adjunct to helping parents learn skills through behavioural parent training.
- Explicitly teaching children skills that typically developing children may learn through observation or abstraction.
- Integration of individualised interventions into an existing system of treatment (e.g. local early childhood intervention services, school based education services, universal services and everyday environments).
- Promoting environmental accommodations (also referred to as the 'external brain') to support executive functioning and emotional and behavioural control
  - Teaching individuals to ask for help and ongoing supervision and prompts to improve self-monitoring such as the use of external devices e.g. mobile phone calendars and reminders.
- Emphasising the importance of structure and explicitly teaching functional routines to provide a scaffolding framework.
  - Utilising visual aids which compensate for executive functioning deficits in set shifting, working memory and attention. It is important to work with caregivers and teachers to design and then follow a plan for providing and withdrawing appropriate amounts of assistance at appropriate times.
- Individualised interventions
  - Understanding the child/young person's cognitive-behavioural profile when designing and intervention program. Strategies include: careful presentation of information (multimodal and repeated); and, hands on experiences.
  - Use strategies from the child's zone of proximal development (ZPD). A distinction between a child's actual developmental level and their potential developmental level<sup>292</sup>. A caregiver or teacher can use hints, prompts and cues which can then later be removed.
  - Scaffolding can be used as it involves orchestrating social contexts known to support children's learning, such as make-believe play or specifically designed group activities. Scaffolding may also involve introducing children to special tools (such as an alphabet chart) and behaviours (such as private speech or self-talk) that children can use to self-assist while mastering a new skill or concept.

## 9.2 Best practice indicators for planning services and supports

1. Comprehensive services for individuals with FASD with the involvement of the individual, family members, and multiple systems of care.
2. Family centred planning, with the family's needs comprehensively assessed and decision making undertaken as a partnership between the family/caregivers, case/key workers, educators, therapists, etc.
3. A strengths based approach – often used in educational settings and relevant to intervention types which promote building on an individual's strengths as determined through development of an individual neurodevelopment profile.
4. Case management based on a key worker model (for example, Local Area Coordinators, Disability Services Commission) to promote good coordination between medical, educational and community services and other professionals and individualised approaches to plan and support individual needs. The role of the key worker is to educate family, service providers, teachers and other individuals in a child's life, provide advocacy and linkage to all required services. In early childhood intervention the 'transdisciplinary key worker' model has been identified by the NDIS as a teamwork model as key practice in successful early childhood intervention<sup>293</sup>. Case management and advocacy needs to be lifelong and continuous.
5. Partnership model (family + school + therapy) supported by case management as optimal in promoting consistent interventions able to be replicated in family and other environmental exposures. From the outset it is important to consider the child or adult with FASD within the context of his/her community, allowing the sharing of resources between health, education and other involved services – in effect a *coordinated family + school + therapy partnership approach*. This partnership approach requires a coordinating body capable of appointing case managers. It is vital that the family is involved in all aspects of decision making with the intervention team. Parents and caregivers need continual support and training for each stage of development.
6. Earliest possible diagnosis or assessment, incorporating age and culturally appropriate assessments with referral to interventions relevant to the range of impairments identified. Comprehensive developmental or educational assessments as well as additional (where necessary) neuropsychological-behavioural assessments to guide individualised intervention efforts. Increased supervision and one-on-one supports to be incorporated with planning to navigate the years from 12 to 21. Recognition that, developmentally, the challenges of FASD grow as the child grows.
7. Practising harm reduction. This means having professional multidisciplinary teams that attend to the medical issues related to FASD and making available periodic screening for hearing, vision, speech, and language problems throughout the individual's development. It also means regular screening for substance use, mental health problems, and reproductive health issues; provision of assistance to cope with relationships, parenting, and family life; and specialized intake and interventions for substance use in the mental health and justice systems.

8. The prevention of secondary effects in individuals with FASD requires programs that are:
  - Available to individuals across the lifespan,
  - Have a prevention focus,
  - Are individualised,
  - Are comprehensive, and
  - Are coordinated across systems and developmental stages<sup>294</sup>.
9. Use of scientifically validated interventions appropriate to the population is vital. These include:
  - Family-focused interventions across the lifespan.
  - Age-appropriate interventions to promote self-regulation.
  - Domain-specific interventions to build skills in deficits areas.
10. Building agency capacity and competence in FASD informed care is an important first step to adopting evidence based interventions for people with FASD. Guidance can be taken from international guidelines and resources, for example, SAMHSA who have recently published a Treatment Improvement Protocol (TIP 58) for Addressing Fetal Alcohol Spectrum Disorders<sup>295</sup>.

### 9.3 Gaps in the evidence

There are a number of gaps in evidence which have emerged from this critical review of the literature and which are potentially relevant to the NDIA. These include:

1. How lack of awareness of FASD in the broader community and among health, education, justice and other professionals has prompted parents and caregivers of children and adolescents with FASD to promote and share anecdotal information through websites, online forums and other social media. These sources of information have contributed to researchers being more fully exposed to the daily functioning impacts that FASD creates for those affected and has led to partnerships between researchers and community members in Australia (and elsewhere) to address the problems and create a more comprehensive response to diagnosis, assessment, treatment and prevention initiatives. This has also revealed the need to promote community awareness of FASD. There has been no formal review of the impact or role of these technologies in education and awareness raising.
2. There is a need for research which specifically addresses the developmental trajectory for FASD as well as the neurobehavioural sequelae and health risks associated with FASD in adults. More understanding related to the relative strengths and weaknesses across the course of development could ultimately improve diagnosis and lead to appropriate intervention strategies.
  - Recognize that for people with FASD, adulthood cannot be framed by traditional life trajectories. Life trajectories need to be established along with the analysis of the costs and benefits of extending care.
3. Evidence which addresses targeted interventions for Aboriginal and Torres Strait Islander people with FASD, including:
  - An absence of literature at the time this critical review was written;
  - The comorbidity and complexity of disability which accompanies FASD amongst Indigenous Australians particularly in rural and remote areas;

- Issues around assessment for Indigenous Australians particularly in rural and remote areas, including: culturally appropriate assessment procedures including IQ testing (and the sensitivities that surround such testing), time frames for conducting these, the language of disability, and broader issues such as trust, expertise and human capital;
  - Challenges in establishing family and community support systems where there is intergenerational neurocognitive disability and extremely limited resources.
4. A lack of evidence considering the CALD population in the Australian context.
    - The provision of CALD appropriate assessment tools.
  5. More focused strengths based research action in FASD is particularly needed as its clinical utility with Indigenous and CALD groups has some evidence which can be further examined.
  6. As more individuals are recognised with the effects of prenatal alcohol exposure, there will be more opportunity for focused interventions with this group. Further research will be needed to examine the effects of combining interventions such as those that are 'domain specific' and those that are 'domain general'. As interventions with FASD are still very much in their infancy, further research is necessary to determine dose and effect outcomes and how this can be applied in a variety of settings in the community, particularly with Indigenous and CALD groups.
  7. Access to services to provide comprehensive assessment in rural and remote settings is a challenge requiring a policy response across health, education, justice, disability and social services.
  8. Future research should specifically examine both the positive characteristics of individuals with FASD and the benefits of parenting individuals with FASD and how these characteristics and benefits can be enhanced.
  9. More attention and research is required into infant mental health interventions for children with prenatal alcohol exposure and FASD, as these have been found useful among other biologically and ecologically vulnerable children.
  10. Literature addressing the issue of 'choice and control' for young people and adults with FASD, given the centrality of this concept to the NDIS and the complexity this will entail for people with impaired executive functioning and other functional domains.
  11. Future directions for research to include sleep disorders and sleep interventions for individuals with FASD. Further investigation is needed to better describe the sleep disturbances associated with FASD and their impact on overall health and daytime neurobehavioral problems in this clinical population.
  12. The inclusion of Neurobehavioural Disorder Associated with Prenatal Alcohol Exposure (ND-AE) in the proposed DSM-5 has implications for NDIA. The DSM-5 addition provides a proposed method for recognising conditions in the category of FASD in the field of mental health. Individuals impacted by PAE are significantly over represented, yet under recognised in the outpatient and inpatient mental health and psychiatric settings. This is also true in the child welfare system, juvenile detention system and in correctional facilities.
  13. Evaluation of the impact of provider training, informal parent support, respite care, parent education, and specialised parenting interventions on caregiving, family quality of life and individual outcomes is needed using quality research design.
  14. International parenting intervention models already shown to be of quality should be considered for replication in the Australian context.



#### **9.4 Implications of the critical review for policy, practice and research**

As a foundational document, this critical review has drawn together the available evidence to inform NDIA planners of relevant background for developing best practice guidelines for use in planning frameworks that support children, young people and adults with FASD to access the services they require to address their needs.

It is a dynamic document requiring future updates as more research is conducted in the field. Better and more knowledge will lead to improvements in interventions with new interventions developed as knowledge increases. This will lead to improved practice in a range of systems (e.g. therapy, educational, justice).

The immediate policy implication for NDIA is for FASD to be embedded in the disability sphere as a permanent and lifelong disability that with appropriate intervention and support can be addressed through a range of means towards improved and positive life outcomes for affected individuals.

A new model is required to meet the needs of children and youth in care: life trajectory planning that recognises that these individuals need support in the community to improve their capacity to contribute. It is important to consider the long term cost effectiveness of community based supportive services that are accessible and that are not contingent on restrictive eligibility criteria.

The practice implication is for professionals working with individuals affected by FASD to be exposed to up to date information to enhance their capacity to effectively respond and for them to be linked to the people they interact with professionally to provide a collaborative and integrated approach. This includes developing continuing professional education packages to keep clinicians, therapists, teachers, and others fully informed of advances in assessment, diagnosis, treatment and management.

The research implication is for researchers to work closely with all stakeholders (including parents/carers) towards the ongoing development of evidence based, targeted and comprehensive interventions.

## Section Two

The significant findings from the critical review have been summarised in Section One to bring focus to the key evidence relevant to informing NDIA in relation to FASD and disability services.

This section includes a description of the methods used to identify and select literature for the critical review and the detailed findings from the literature retained for analysis and reporting. It includes more detailed descriptions of interventions than included in Section One and is provided as a comprehensive background to the synthesised evidence presented in that section.

Some cautionary notes are relevant.

Firstly, the review process has not been a systematic review. It is a critical review of the literature identified through a search of relevant databases and by contacting known experts in the field of FASD. Secondly, the detailed findings in Section Two have been chosen for their relevance to describing the domains of impairment associated with FASD and the interventions designed, implemented, evaluated and reported as having a positive impact on these impairments. Thirdly, this is a first attempt at synthesising literature to present a detailed picture of the implications of FASD for the disability sector. As such, it will require updating as further evidence accumulates.

The critical review of the literature has avoided using evidence from programs of work or research specifically aimed at education and justice contexts, as these generally are separated from disability supports planning. So too, interventions which are solely the remit of medical management, such as pharmacological treatment with or without behavioural management, have been excluded for the same reasons.

Rather, interventions focussed on individuals and their families/caregivers and which may take place in home, school and/or therapy (clinical) settings, or may be integrated across these settings; are designed to address neurocognitive and neurobehavioural impairments; and, fall under the disability umbrella, have been included.

Diagnosis related to FASD is a complex process, and the relatively recent recognition of FASD as encompassing variable disabilities, each with varying severity, has meant the critical review needed to draw on published journal articles, unpublished reports, information obtained through websites and other relevant material as provided by experts, to present as detailed account as possible.

Even though Fetal Alcohol Syndrome (FAS) was first identified 40 years ago, and then revealed through ongoing research, FASD is only now beginning to be more broadly understood as a spectrum disorder, with evidence emerging about what works in addressing the significant primary disabilities and secondary effects encountered by people with FASD. The impact of FASD on individuals and in relation to their experiences of and interaction with education, criminal/justice and mental health services is a precipitating factor behind the need for a cohesive systems approach to both acknowledge and address disability associated with FASD. As with all brain based impairments, the implementation of interventions early, frequently and over sufficient periods of time will assist in improving quality of life outcomes.

## Critical Review Methodology

### Defining the topic

A critical review of the literature was conducted to identify the domains of impairment and how these impact on daily functioning, and, the services and supports required to ensure those with FASD are afforded opportunities to participate in education, employment and social activities.

Studies and programs relevant to addressing the impacts of FASD on individuals, as identified through the critical review and informed by experts, were assessed using the following criteria as a standard of inclusion:

1. A program has been evaluated using a valid evaluation framework.
2. The evaluation demonstrates effectiveness for people with FASD for an outcome related to living independently and/or participation in daily life.
3. The evaluation was assessed to be of sufficient quality, using a validated assessment checklist<sup>296</sup>.

### Methods used and justification

Programs were assessed and allocated to either best practice or promising practice programs, defined as:

- *Best practice programs* as those that meet criteria regarding research design quality, impacts, specificity and replicability, such as:
  - randomised controlled trial methodology (with at least 20 participants in both the test and control groups); or,
  - quasi-experimental methodology (with at least 20 participants in both the test and control groups); or,
  - excellent qualitative research (interviews conducted with appropriate percentage of program participants; sampling strategy appropriate to reflect diversity of views; findings provide a rich description of 'lived experiences' of participants); or,
  - pre- and post-test methodology (with control group and with at least 20 participants in both the test and control groups); or
  - mixed methods (outcome indicators are evaluated for at least 20 participants and there is an excellent qualitative research component as defined above).

Additionally, a program must have demonstrated positive impacts on desired outcomes (must be statistically significant or change of at least 20 per cent or very strong qualitative support) and the program designers/authors should not report any negative or harmful effects. Finally, there were clear statements in the available information about what the program involves, whom it is for and why it is important (specificity).

- *Emerging programs* are those that have some evidence to support effectiveness in terms of content or delivery. The criteria were more flexible than for the best practice programs to account for the emerging evidence in the field.

## Identification and Selection of the Literature

### Identification of literature for review

A desktop literature search of academic and general databases was conducted using a broad selection of terms to identify published peer-reviewed and grey literature. Personal communications were also sent to experts in the FASD and Disabilities fields with a request for copies of unpublished evaluations and program descriptions. Table 2 sets out the databases searched, together with the parameters and search terms used. Table 3 and 4 lists the organisational websites and networks referred to. Table 5 names the report authors. Tables 6 and 7 names the experts who were consulted and/or who participated on the Expert Review Panel. Expert Review Panel members also received report drafts for comment and feedback.

### Method of inclusion

The selected literature was reviewed by a team of four researchers (the report authors).

Firstly, the Kmet et al (2004) quantitative and qualitative checklists were used (see Tables 12 and 13, Appendices). These comprise a 14 point quantitative and 10 point qualitative list of questions which require a Yes/Partial/No assessment to determine a total score (maximum = 28 quantitative; 20 qualitative). For some articles, some checklist questions were not relevant and the maximum score was adjusted accordingly. Individual article scores were then converted to a percentage. Articles which scored at 75 per cent and above were regarded as Level 1 evidence. That is, the studies had sufficient rigour associated with design and outcomes. Articles which scored between 50-74 per cent were regarded as Level 2 evidence. That is, while there were limitations in study design, emerging evidence reported is relevant to creating an evidence base.

Secondly, to assess the impairments evidence, a scoring system was devised to rank the literature referring to either neurocognitive behavioural impairments, impairment assessment and diagnosis, and whether recognised or standardised assessments/criteria were used.

Neurocognitive Impairments	Behaviour	Impairment Assessment	FASD diagnosis
Score=2: The study identified neurocognitive impairments according to recognised CNS domains affected by FASD.	Score=2: The study identified behavioural impairments according to recognised CNS domains affected by FASD.	Score=2: The study identified standardised assessments of impairment Score=1: The study identified assessments of impairment that were not clearly identified as standardised.	Score=2: The study participants had a formal diagnosis on the FASD spectrum using recognised criteria. Score=1: The FASD diagnosis of participants in the study was not clearly defined using recognised criteria. Score=0: The FASD diagnosis of participants in the study was not described at all.

**Table 2: Literature Search Parameters**

Academic Databases	General Databases	Search terms	Search parameters
Scopus, CINAHL, MEDLINE, Pubmed, Informahealthcare, PsycInfo, Proquest, Global health, Sciencedirect, Embase, AUSThealth	Google, Google Scholar, Web of science	Fetal alcohol spectrum disorders, FASD, prenatal alcohol exposure, Intervention studies, Intervention*, rehabilitation, management, case management, community, services, supports, participation, function, evaluation, impairment, transition, attachment, mental health, mental health disorders, substance use, parenting, indigenous, life course, life span, secondary condition, behaviour, self-regulation, sensory processing, executive functioning, adaptive functioning, neuropsychological, Neurobehavioural	1980 to present

**Table 3: Networks**

Networks Accessed
<p><i>Key organisations</i></p> <p><b>Australia</b>            National Organisation for Fetal Alcohol Spectrum Disorders            Telethon Kids Institute            Russell Family Fetal Alcohol Disorders Association            Government of Western Australia – Department of Health            Department of Communities – Western Australia            Australian Government – Australian Institute of Health and Welfare            The Foundation for Alcohol Research and Education</p> <p><b>International</b>            Canada FASD Research Network            Fetal Alcohol Syndrome Prevention Team, National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention (USA)            National Organisation of Fetal Alcohol Syndrome (USA)            FASD Centre for Excellence – USA            The Asante Centre, Canada            National Organisation for Fetal Alcohol Syndrome UK            University of Washington, USA            University of British Columbia, USA            University of Victoria, BC, Canada</p>

**Table 4: Websites**

Websites Accessed
<a href="http://www.nofasd.org">www.nofasd.org</a> <a href="http://www.rffada.org/">http://www.rffada.org/</a> <a href="http://www.aifs.gov.au">www.aifs.gov.au</a> <a href="http://www.fare.org.au">www.fare.org.au</a> <a href="http://www.cdc.gov/fasd">http://www.cdc.gov/fasd</a> <a href="http://fasdcenter.samhsa.gov">http://fasdcenter.samhsa.gov</a> <a href="http://www.cdc.gov/ncbddd/fasd/">http://www.cdc.gov/ncbddd/fasd/</a> <a href="http://www.canfasd.ca/">http://www.canfasd.ca/</a> <a href="http://www.asantecentre.org/">http://www.asantecentre.org/</a> <a href="http://www.fascets.org/">http://www.fascets.org/</a> <a href="http://www.fasdwaterlooregion.ca">www.fasdwaterlooregion.ca</a> <a href="http://www.fasdalberta.ca">www.fasdalberta.ca</a> <a href="http://www.fasdnetwork.org">http://www.fasdnetwork.org</a> <a href="http://www.clinicaltrials.gov">http://www.clinicaltrials.gov</a> <a href="http://come-over.to/">http://come-over.to/</a> <a href="https://www.ubc.ca/">https://www.ubc.ca/</a> <a href="http://www.fasdmanitoba.com">http://www.fasdmanitoba.com</a> <a href="http://www.motherisk.org/">http://www.motherisk.org/</a> <a href="http://www.nofas-uk.org/">http://www.nofas-uk.org/</a> <a href="http://www.skfasnetwork.ca">http://www.skfasnetwork.ca</a> <a href="http://www.faslink.org">http://www.faslink.org</a> <a href="http://www.fasdjustice.ca">http://www.fasdjustice.ca</a> <a href="http://jhscso.bc.ca/">http://jhscso.bc.ca/</a> <a href="http://wrapschools.ca/">http://wrapschools.ca/</a> <a href="http://kimberleyfasdresource.com.au/pdf/FASD_ResourceForTeachers.pdf">http://kimberleyfasdresource.com.au/pdf/FASD_ResourceForTeachers.pdf</a> \ <a href="http://www.neurodevnet.ca/">http://www.neurodevnet.ca/</a>

**Table 5: Report Authors**

Author	Expertise	Institutional Affiliation
Prof Carol Bower	Epidemiology and Public Health	Telethon Kids Institute
Angela Dudley	Occupational Therapy	Telethon Kids Institute
James Fitzpatrick	Paediatrician and Senior Clinical Research Fellow	Telethon Kids Institute
Tracy Reibel	Senior Research Fellow	Telethon Kids Institute

**Table 6: FASD Expert Review Panel**

Expert	Field of Expertise	Institutional Affiliation
Dr Lynne Adamson	Director, Research Supports and Services	National Disability Insurance Agency
Dr Heather Carmichael Olson	Fetal Alcohol Syndrome Diagnostic and Prevention Network, Families Moving Forward Research Program	University of Washington School of Medicine, Seattle Children's Hospital Research Institute, USA
Jennifer Cullen	Chief Executive Officer	Synapse Incorporated
Dr Catherine Elliott	Chair of Allied Health	Child and Adolescent Health Service, Health Department of Western Australia
Prof Elizabeth Elliott	Head, Federal Government FASD Technical Network	University of Sydney, New South Wales
Damian Griffis	Chief Executive Officer	First Peoples Disability Network

Sue Ham	Trial Site Manager	Tasmania and Northern Territory National Disability Insurance Agency
Marmingee Hand	Carer and educator	
Tracy Jirikowic	Occupational Therapist	University of Washington School of Medicine, Seattle Children's Hospital Research Institute, USA
Michele Johnston	Neuropsychologist	Health Department of Western Australia
Denise Luscombe	Consultant Physiotherapist	Disability Services Commission
Malcolm Mill	Director Service Delivery	Barkly Trial Site National Disability Insurance Agency
Dr Raewyn Mutch	Paediatrician and Post-doctoral Fellow	Telethon Kids Institute
Elizabeth Peadon	Paediatrician	University of Sydney, New South Wales
Dr Carmela Pestell	Clinical Psychologist & Neuropsychologist [MAPS] - Director	University of Western Australia – Robin Winkler Clinic
Vicki Russell	Consumer advocacy Chief Executive Officer	NOFASD Australia
Clare Salter	Speech pathologist	
Trevor Sanders/Barbara Shaw	GM Aboriginal Health	Anyinginyi Health Corporation
Dr Doug Shelton	Paediatrician Clinical Director	Gold Coast University Hospital
Prof Sven Silburn	Menzies School of Health Research	Charles Darwin University
Dr Rachel Skoss	Chair, WA Ministerial Advisory Committee on Disability	Telethon Kids Institute
Claire Townsend	Manager	Synapse Incorporated
Robyn Williams	PhD Candidate Health Sciences	Curtin University
Dr Amanda Wilkins	Community Paediatrician	Child Development Service, Health Department of Western Australia
Dr Marcel Zimmet	Consultant Paediatrician	FASD Clinic, Westmead Hospital

**Table 7: Disability/FASD Experts Consultative Group**

Expert	Field of Expertise	Institutional Affiliation
Dr Lynne Adamson	Director, Research Supports and Services	National Disability Insurance Agency
Scott Avery	Policy and Research Director	First Peoples Disability Network
Kerryn Begley	Social Worker	Private FASD consultation & training
Dr Heather Carmichael Olson	Fetal Alcohol Syndrome Diagnostic and Prevention Network, Families Moving Forward Research Program	University of Washington School of Medicine, Seattle Children's Hospital Research Institute
Dr Julianne Conry	Registered psychologist	The Asante Centre, Canada
Mike Cabbage	Behaviour Support Consultation Manager	Disability Services Commission, WA
Jennifer Cullen	Chief executive officer	Synapse Incorporated
Lee Davids	Director, Continuous Improvement Scheme Design	National Disability Insurance Agency
Dr Catherine Elliott	Chair of Allied Health	Child and Adolescent Health Service, Health Department of Western Australia

Prof Elizabeth Elliott	Head, Federal Government FASD Technical Network	University of Sydney, New South Wales
Fetal Alcohol Syndrome Prevention Team	National Center on Birth Defects and Developmental Disabilities	Centers for Disease Control and Prevention, USA
Sister Suzette Fisher	Director of Client Services	Double ARC Ohio, USA
Joanna Granich	Clinical Trials Coordinator – Autism	Telethon Kids Institute
Damian Griffis	Chief Executive Officer	First Peoples Disability Network
Sue Ham	Trial Site Manager	Tasmania and Northern Territory National Disability Insurance Agency
Marmingee Hand	Carer and educator	
Jacki Hollick	Sector Service Development Manager	Disability Services Commission
Mindy Horsman	Social Work Supervisor	State Child Development Centre - Department of Health WA
Tracy Jirikowic	Occupational Therapist	University of Washington School of Medicine, Seattle Children’s Hospital Research Institute, USA
Michele Johnston	Neuropsychologist	Health Department of WA
Heather Jones	Manager of FASD Projects	Telethon Kids Institute
Dr Julie Kable	Assistant Professor Psychiatry and Behavioural Sciences	Emory University School of Medicine, USA
Patricia Lingley-Pottie	President & Chief Executive Officer	Strongest Families Institute IWK Health Centre, Canada
Denise Luscombe	Consultant Physiotherapist	Disability Services Commission, WA
Malcolm Mill	Director Service Delivery	Barkly Trial Site National Disability Insurance Agency
Dr Raewyn Mutch	Paediatrician and Post-doctoral Fellow	Telethon Kids Institute
Dr Mary O’Connor	Clinical Psychologist - Director	UCLA Fetal Alcohol Spectrum Disorders Clinic, USA
Elizabeth Peadon	Paediatrician	University of Sydney
Dr Carmela Pestell	Clinical Psychologist & Neuropsychologist [MAPS] - Director	University of Western Australia – Robin Winkler Clinic
Vicki Russell	Consumer advocacy Chief Executive Officer	NOFASD Australia
Deborah Rutman	Principal, Nota Bene Consulting Group	Adjunct Associate Professor, School of Social Work University of Victoria, Canada
Clare Salter	Speech pathologist	
Trevor Sanders/Barbara Shaw	General Manager Aboriginal Health	Anyinginyi Health Corporation
Dr Doug Shelton	Paediatrician Clinical Director	Gold Coast University Hospital
Prof Sven Silburn	Menzies School of Health Research	Charles Darwin University
Dr Rachel Skoss	Chair, WA Ministerial Advisory Committee on Disability	Telethon Kids Institute
Victoria Suen	Research and Evaluation Consultant	Alberta Health Services, Canada
Claire Townsend	Manager	Synapse Incorporated
Bree Wagner	Alert Program Study Coordinator	Telethon Kids Institute
Kristen White	Program Manager Aboriginal Health Pilbara and Kimberley	Telethon Kids Institute
Robyn Williams	PhD Candidate, Health Sciences	Curtin University



Dr Amanda Wilkins	Community Paediatrician	Child Development Service
Dr Marcel Zimmet	Consultant Paediatrician	FASD Clinic, Westmead Hospital

## Critical Review - Detailed Findings

The detailed findings presented here are the result of analysis and reporting of literature identified by the process outlined in the Methodology. The detailed findings start with a broad overview of FASD and its impact on the lives of those affected from both a life course and neurodevelopmental perspective. It then sets out the details related to the domains of impairment, before moving to detailed description of the interventions included in the analysis and summarised in Section One.

### **Overview of FASD**

Commonly, the child affected by FASD first comes to the attention of health services within an educational environment, when they are identified as noticeably functioning at lower levels than their peers<sup>297</sup>. Up to this point, affected children may have masked their neurocognitive deficits in social situations by being affable and chatty. However, attention deficits will emerge as the tasks children are required to perform become more complex. Other disruptions to cognitive functions vary, but can include diminished intellectual functioning, slower speed of information processing<sup>298 299</sup>; deficient cognitive set shifting<sup>300 301</sup>; ineffective problem solving strategies<sup>302</sup>; and, non-verbal and verbal fluency difficulties<sup>303</sup>. Thus, although many of the social behaviours in children with FASD may seem acceptable at a young age, over time these behaviours become increasingly inappropriate and markedly different from peers. Further, as social interactions become more complex, requiring higher level communication skills, challenges in social communication and adaptive functioning emerge. For example, group studies in children with FAS have found deficits in speech and language functioning including: word comprehension; articulation; and, expressive and receptive language skills<sup>304</sup>.

Adaptive functioning, defined as the skills required to carry out daily activities necessary for personal and social self-sufficiency or the ability to respond successfully to everyday demands; is often impaired in FASD<sup>305 306</sup>. In a study of adaptive functioning in children aged 5-8 years with FAS, their performance was significantly below that of their peers in all but three of fourteen subscales on the SIB-R<sup>307</sup>. In addition there are a wide variety of maladaptive behavioural characteristics described. Externalising behaviour problems, namely social problems, attention problems, and aggressive behaviour, are frequently reported<sup>308</sup> with parental reports of behavioural problems noted in numerous studies<sup>309 310 311</sup>. Many individuals with FASD are ultimately unable to live or work independently<sup>312</sup>. Further, the odds of a range of mental health disorders and traits are more than doubled for adults exposed to alcohol in utero<sup>313</sup>. Early intervention before the age of 6 years is one of the most important 'protective' factors, and can reduce secondary disabling effects for individuals with FASD by 2-4 fold<sup>314</sup>. Awareness of an individual's specific strengths and needs will guide interventions that promote function and participation<sup>315</sup>.

The importance of neuropsychological testing and the requirement to assess children affected by FASD to develop a behavioural learning profile is emphasised by several researchers<sup>316 317 318 319</sup>. However due to the individual variability of alcohol's teratogenic affects and the pattern and timing of maternal drinking, as well as the impact of epigenetic risk factors the behavioural profile of neuropsychological impairments is variable<sup>320 321</sup>.

As indicated, children with FASD are often first identified at school when behavioural/learning difficulties manifest, not necessarily because they have previously been diagnosed with a FASD. On assessment, FASD affected children will sometimes fall within average intelligence and therefore may

not be eligible for educational support. As also noted previously, where there is known or suspected prenatal exposure to alcohol, children from age six onwards will benefit from a battery of tests to determine the extent of their impairments and where their strengths and weaknesses lie.

Kodituwakku<sup>322</sup> has developed a neurodevelopmental framework based on cognitive neuroscience principles which guides development of skills teaching interventions for children with FASD.

To identify children's impairments, neurobehavioral tests include those associated with: attention, verbal learning and recall, verbal memory, auditory memory, spatial memory, auditory processing and verbal processing. In addition to standard IQ, achievement (as determined by the WRAT for example) and adaptive measures are required. A distinct profile of neuropsychological deficits is evident in individuals with FASD, regardless of the presence of the facial phenotype<sup>323</sup>.

One of the primary areas affected is executive functioning, an umbrella term referring to a group of higher-level abilities that help guide and control behaviour, acting like the 'executive' of a company to direct and organise our activities. There is no one definition of executive functioning as a number of cognitive skills are implicated. These include self-regulation, problem solving, working memory, information processing and goal orientated behaviour<sup>324</sup>. Also included is the ability to initiate a task or activity on one's own, to plan, to organise tasks and materials, to sequence, to prioritise, to shift between activities and conversational subjects, and to inhibit (stop or refrain from) an action<sup>325</sup>. The executive functioning difficulties encountered by those affected with FASD can have a significant impact on adaptive and social functioning. For example, poor working memory skills can adversely affect learning, language and academic attainment. Weaker organisation, planning and problem solving also impact on activities of daily living. Poor cognitive flexibility, inhibition and self-regulation problems also contribute to social skill deficits and affect interpersonal relationships contributing to social behaviour problems<sup>326</sup>.

A core impairment within FASD is CNS dysfunction and this domain is impaired across the whole spectrum of FASD: FAS, pFAS, and ND-AE<sup>327</sup>. Numerous studies have focused on executive functioning in FAS and difficulties have been found in children even when their intelligence fell within the normal range<sup>328</sup>. More complex tasks involving both cognition and emotion were more difficult and more impaired for children prenatally exposed to alcohol<sup>329</sup>. Cognition based executive functioning limitations may be seen in a child's inability to understand and hold in memory the specific sequences required for tasks of daily living and academic processes. For example, the child may experience difficulty with following daily routine sequences, social exchange processes, and managing time.

A comprehensive profile of functioning needs to be done to determine the specific needs of the child's level of functioning with academic, cognitive and behavioural learning areas. Multidisciplinary approaches leading to goal setting and individualised plans which take into account various school settings and the skills needed to manage these, is required. The caregiving environment (or what the child constitutes as their family) must also be included in guiding the school program for the child with FASD. A *family + school + therapy partnership* approach is needed to ensure consistency in the child's environment.

This can be achieved by working with the family, school and (multidisciplinary) therapy teams, to observe a child's skills and capacity in maintaining attention, independence, social interactions, functional language, and, to establish their strengths, interests and behaviour. This needs to occur across multiple observations and in a variety of contexts.

A mismatch may exist in a child with FASD as they may have strengths in their expressive language ability, which may mask their inability to retain information in their verbal memory, hence they may appear more capable than they really are. Research supports the impairments within executive functioning that describe the difficulties when translated to function, such as taking in new verbal information and recalling auditory directions<sup>330 331</sup>.

Further, emotion-related executive functioning deficits may present as an inability to inhibit responses to situations involving others, resulting in inappropriate actions or verbal responses, or being over-active. FASD-affected individuals may have difficulty inhibiting their actions in circumstances where they would ordinarily be expected to understand the social behaviours required. Acting before considering consequences is an extremely common characteristic seen in FASD. Individuals may be socially intrusive and have difficulty understanding the steps of social interactions, for example by encroaching on the personal space of others<sup>332 333</sup>.

FASD and ADHD may be present as co-morbidities but there are key differences between the two diagnoses. Those with FASD have most difficulty shifting and encoding new information whereas children with ADHD have more difficulty focusing and sustaining attention. Where there is more difficulty in shifting attention, there is a greater potential for perseveration and difficulty with transitioning from one task to the next and from one place to another. From a functional perspective, those with FASD have more difficulty with life changes, for example, family memberships, moving schools and transitioning to the next grade<sup>334</sup>.

## **Domains of Impairment – Detailed Findings**

We identified 27 studies (see Appendices) which included recognised neurocognitive behavioural impairments, using standardised assessments of impairments combined with a formal diagnosis related to FASD and based on recognised criteria. Three studies were excluded which did not meet these requirements. Of the included studies, the majority were focussed on early childhood through to late adolescence (3-18 years). Four studies included infants, but these were incorporated into a wide age range (most often 0-adult). There were no specific adult studies identified, although one study included an age range of 6-51 years, and another young adults aged 16-25 years.

There is a lack of consistency in studies with regard to: age ranges included in study samples, the variability of impairments assessed and the variety of assessment tools used. This resulted in an inability to draw summary conclusions or distinguish an overall hierarchy of domains of impairment related to FASD, as these are unique to the individual. Some data are drawn from population-based, longitudinal and epidemiological studies of children without diagnoses on the FASD spectrum, but with known prenatal alcohol exposure and referred to as being on the social drinking range. What was clearly indicated across these clinical and epidemiological studies is the extent of impairments across the life course requiring appropriate assessments at relevant transitional time points and prior to interventions being implemented.

The importance of considering the role of self-regulation on social cognition and behavioural functioning has been investigated in children with brain injury. Self-regulation may be defined as the capacity to manage one's thoughts, actions and feelings, in adaptive and flexible ways across diverse contexts, and deficits may include poor inhibitory control and deficits in planning and organization, often implicated in the EF domain and attributed to the prefrontal cortex<sup>335</sup>.

Astley's<sup>336</sup> comprehensive study of 1,400 patients (mean age 9.9, range 7 days to 51 years with 90 per cent under 16 years) receiving diagnostic evaluations for FASD showed that 58 per cent of those with FAS/pFAS had significant impairment in executive functioning/memory, 57 per cent had motor/sensory impairment, 50 per cent had language impairment, and 51 per cent had significant delays in development. In the group 5 years and up, 82 per cent had a mental health disorder, with most prevalent being ADD/ADHD (54 per cent) followed by Post-traumatic Stress Disorder at 7 per cent across the whole FASD spectrum and Oppositional Defiant Disorder at 9 per cent.

In a longitudinal German study with follow up over 20 years (Spohr, Willms, & Steinhausen, 2007), significant impairments were seen in behavioural and emotional areas. Most notably, there was evidence of thought disorders, attentional problems, intrusive and aggressive behaviour. The study indicated that these impairments were experienced by those at varying levels on the spectrum of FASD. The limitation of this study was its small sample size (n=37) and its specificity to location.

There is no well-ordered age and stage categorisation of domains of impairment related to FASD in the literature. Instead, the available evidence has been synthesised below to highlight the likely age related progress of functional impairments. These are described for: infancy/early childhood; childhood (4 to 12 years); adolescence (13 to 18 years); and adulthood.

### **Infancy/early childhood**

More evidence is needed investigating the impact of alcohol in the 0-3 age range. In the neonatal period it is possible to assess habituation, a learned decrease in response to a biologically irrelevant stimulus after repeated presentations<sup>337</sup>. Abnormal rates of habituation have been related to later delays in intellectual development<sup>338</sup> and thus are thought to represent one of the early measurable aspects of CNS function in newborns<sup>339</sup>. Early indicators of developmental vulnerability include low tone, feeding difficulties, poor visual and auditory habituation, sleep disturbances and a poor sleep/wake cycle<sup>340</sup>.

A population based longitudinal study investigating the effects of prenatal alcohol exposure not specific to FASD, from birth to 25 years was conducted in Seattle, USA, beginning in 1974<sup>341</sup> with some follow-up beyond age 25. Utilising the Brazelton Neonatal Assessment Scale with 417 neonates, the factor scores of habituation and low arousal were particularly significant; 'low arousal' reflected the ability of the infant to maintain a calm/ alert or sleep state and frequently alternating between awake and drowsy. Neonates having difficulty maintaining calm/alert sleep states suggests that self-regulatory difficulties are evident from the time of birth. In addition, other neonatal items correlated with prenatal consumption of alcohol were: more hand to mouth activity, a weak MORO reflex, hypertonia, and delayed stepping reflex threshold.

Global developmental delay (GDD) in the context of prenatal alcohol exposure maybe a common indication of FASD in infancy Just over half of children aged birth to 3 years with prenatal alcohol exposure (or with FAS/pFAS) show marked developmental delay<sup>342</sup>. This includes delays in: gross and fine motor skills, speech and language, cognitive and social and emotional development. Findings in a longitudinal epidemiological study at eight months with 462 infants using the Bayley Scales of Infant Development reported that babies exposed to higher amounts of alcohol *in utero*, compared to those with no or lower exposure, performed more than 1 SD below the mean in their overall mental functioning<sup>343</sup>.

A meta-analysis of studies of gross motor deficits in children with FASD indicated that in one study related to infants, a gross motor impairment amongst 7 to 12 month olds was seen on the locomotion subscale of the Griffiths Mental Developmental Scales<sup>344</sup>.

Insecure attachment and parenting difficulties and irritability (“negative affectivity”) of the infant are often reported with the infant and young child with suspected or confirmed prenatal exposure to alcohol<sup>345</sup>. Insecure attachment may lead to impaired or atypical social-emotional development, poor school readiness, behavioural problems, psychiatric disorders, and impaired long term mental health outcomes<sup>346 347</sup>.

### Childhood (4 to 12 years)

By the time a child reaches four years of age, evidence shows that the main difficulties starting to emerge are attention<sup>348 349 350</sup> (a component of executive functioning) and behavioural problems<sup>351</sup> within the self-regulation and social communication domains of functioning, gross<sup>352</sup> and fine motor skills<sup>353 354</sup>, sensory processing and adaptive functioning<sup>355 356</sup>. A wider variety of neurobehavioural assessments can be administered in this age range (see Appendices). In addition to attentional difficulties and slower reaction times, children around this age have also been noted as acting young for their age, with hyperactivity reported, and impulsivity and restlessness or irritability also becoming evident<sup>357</sup>. It is important to note that the inattention and hyperactivity associated with prenatal alcohol exposure denote a particular clinical subtype with an earlier onset, a different clinical and neuropsychological presentation, and probably a differential medication response than idiopathic ADHD<sup>358 359</sup> which is a common comorbidity alongside FASD.

Children in the 5-8 years age range have been shown to have significantly lower scores on the Short Sensory Profile (SSP) and the sensorimotor core domain of the NEPSY-SM than typically developing children as well as more problems with sensory modulation and sensory motor performance. They also have more significant difficulties in the spelling and arithmetic sections in the WRAT-3 test with significant correlations between the SSP and the maths and spelling sections of the WRAT-3, suggesting a link between sensory processing dysfunction and the academic function of some children with FASD<sup>360</sup>.

In the same study, behavioural rating reports by parents and teachers showed children with FASD demonstrated lower adaptive skills evident on the SIB-R composite score with 78 per cent of children with FASD compared to 4 per cent of control children demonstrating serious behavioural difficulties.

It is noteworthy that children with pFAS or ND-AE may perform worse with their sensory processing scores and adaptive behaviours than those diagnosed with FAS, but perform better on measures of intellectual functioning. These results support the theory that deficits in adaptive behaviours may be independent of lower IQ<sup>361 362</sup>. Lower levels of adaptive functioning can be assessed using specific tools such as the SIB-R or the VABS to determine the degree of impairment and its impact for the individual<sup>363</sup>. Broad independent living skills are particularly impaired. This will change at different points along the life course. For younger children, problems with toileting and eating are common, while problems such as understanding money/value and more complex language comprehension emerge with increasing age.

A cohort of 13 children with FAS/PFAS, aged 7-9 years and living in remote Canadian Aboriginal communities had the following CNS impairments: ADHD with or without sensory dysfunction (69 per cent), academic achievement (62 per cent), communication (54 per cent), cognition (50 per cent),

memory (50 per cent), executive functioning (50 per cent) and microcephaly (69 per cent)<sup>364</sup>. In a large USA sample of 6-8 year old children with known prenatal alcohol exposure (n=486) difficulties were documented in measures of arithmetic, perceptual motor skills, attention and impulsivity, and cooperation<sup>365 366</sup>.

Children with FASD have been found to exhibit disruptive, uncooperative and socially inappropriate behaviours compared with typically developing children. Children with FASD need direct or frequent support and supervision to manage daily adaptive skills and behaviour compared with typically developing children only needing intermittent support<sup>367 368</sup>.

Sleep disturbances have an impact on behavioural outcomes, particularly self-regulation, but also on memory, learning, attention and cognitive flexibility. They have also been identified in other neurodevelopmental disorders such as autism or ADHD with studies reporting rates of sleep disturbances in the range of 75-80 per cent<sup>369</sup>.

### Adolescence (13 to 18 years)

A recent cross sectional study looking at age related differences across a broad range of neuropsychological variables in three age groups: 5-8, 9-12 and 13-17 years<sup>370</sup> appears to be a first attempt at understanding how neuropsychological impairments manifest for children and adolescents with FASD. It is particularly important when determining appropriate intervention and supports for individuals with FASD and their families and communities. Academic achievement, language, memory, visual motor coordination and executive functioning were all areas of impairment relative to children without FASD. In terms of age related differences, the domains where older subjects performed worse than younger subjects were: executive functioning, arithmetic, learning and memory, visual motor integration and motor abilities. There were no significant differences in IQ between the three age groups in this study.

With regard to academic achievement, arithmetic was the greatest area of impairment consistent with FASD<sup>371</sup>. Receptive language was impaired as measured on the PPVT-R which is also consistent with previous studies<sup>372</sup>. Both children and adolescents with FASD had significant difficulty on the learning and recall of verbal information<sup>373</sup>. A study with 21 alcohol exposed children (18 with FAS) compared to children with Down's syndrome (n=11) and normally developing controls (n=212) and specifically examining memory, showed that children with FASD do not show a global memory deficit in a test of memory impairment. Rather, results supported explicit memory being impaired whereas implicit memory was intact<sup>374</sup>.

Two studies have examined the role of the corpus callosum in children prenatally exposed to alcohol<sup>375 376</sup>. Preliminary analyses from these studies suggest that inaccurate performance on a bimanual coordination task, and likely impaired callosal functioning, may be related to the attention and problem solving impairments experienced by children with FASD. The corpus callosum plays a major role in coordinating motor activity from opposite sides of the body, and has been found to be structurally impaired in some children prenatally exposed to alcohol<sup>377 378</sup>. There may be agenesis of the corpus callosum<sup>379</sup> or spatial displacement within the brain<sup>380</sup>. In children born without a corpus callosum, associated subtle cognitive and behavioural difficulties are poor bimanual coordination, poor psychosocial functioning, impaired processing of complex information and decreased interhemispheric transfer of learning.

At 14 years of age, attention, spatial learning deficits and problems with tasks requiring the complex manipulation of information has been reported<sup>381</sup>. Behaviour problems are identified with increased anti-social behaviours, earlier use of alcohol and other drugs, increased self-perception of difference and self-concept of being a 'poor student'. Stress was identified as a trigger for more difficulties with organising materials and processes and a high impulsivity rating from examiner ratings on the Child Behaviour Checklist.

In addition to the tasks of adolescent development and living with FASD, some adolescents may also have children of their own to raise who may or may not be residing in their care. Parenting when affected by FASD represents many challenges and is an important opportunity to begin interventions that can minimise secondary effects and promote coping with the brain based primary impairments<sup>382</sup>.

In adolescents and young adults, mental health concerns are manifested in alcohol misuse/disorders<sup>383 384</sup>, depression<sup>385 386</sup> and a variety of other mental health disorders<sup>387</sup>.

A general lack of engagement with the community and contact with the justice system are other common characteristics identified in adolescence. The impact of FASD is also mediated by environmental factors such as living with an alcoholic parent or being subjected to child abuse or neglect, or other adverse life conditions such as poverty which often lead to child welfare involvement and a child's admission to care.

## Adulthood

For over three decades pioneering FASD researcher Ann Streissguth has documented risk factors for secondary disabling effects or adverse life outcomes associated with a FAS diagnosis. For young adults mental health problems, contact with the justice system, disrupted education, and alcohol dependence/substance abuse issues were significant at 21 years of age whilst less than 10 per cent of subjects with FASD were able to live or work independently. Motor deficits were also evident, particularly speed of information processing<sup>388</sup>. Frequent adverse life outcomes included inappropriate sexual behaviours (40-50 per cent increasing with age) and mental health problems at higher than 90 per cent. Co-occurring risk factors included being the victim of domestic abuse or violence. In addition the incidence of independence in daily living activities amongst 21 year olds with FASD was very low for activities such as managing money and making decisions with 80 per cent of individuals requiring help or supervision<sup>389</sup>.

In one cohort, at 21 years of age, 90 per cent of subjects had mental health problems, 60 per cent had been in contact with the justice system, 60 per cent had disrupted education, 40 per cent had substance abuse issues and fewer than 10 per cent lived or worked independently. Alcohol dependence has been reported in 83 per cent of subjects with FASD at 21 years of age<sup>390</sup>. Motor deficits were evident in this same cohort of subjects (n=433) at 25 years of age, particularly speed of information processing.

Being the victim of domestic abuse or violence is commonly reported in subjects with FASD, with 67 per cent of a sample of 415 subjects having been the victim of physical or sexual abuse or of domestic violence. The most frequent adverse life outcome from this study is reported as inappropriate sexual behaviours, increasing over the life course from 39 per cent in children, to 48 per cent in adolescents to 52 per cent in adults.

In a 20 year follow up study with 37 patients with FASD in Germany, signs and symptoms were compared from an initial examination in mid childhood (mean age ~ 6 years) and at follow up (mean age 26 years). Characteristics such as growth retardation, microcephaly, developmental delay and hyperactivity generally persisted into adulthood, while FAS facial features present earlier in life were generally not apparent in adulthood. Continued prevalence of microcephaly at follow up was strongly correlated with those subjects who were intellectually disabled at the initial assessment. There was a high persistence from childhood to adulthood of aggressive, externalising and delinquent behavioural problems. In addition, there were a number of secondary adverse life outcomes, including poor school achievement (49 per cent) and receiving special education past primary school (38 per cent). In this group only 13 per cent had completed secondary school and held an ordinary job, while one third were in dependent living situations<sup>391</sup>.

Sixteen adults (age range 16-27) with a range of intellectual functioning levels (low average to above average) with FASD manifested clear deficits on neuropsychological measures sensitive to complex attention, verbal learning and executive function. Both the frequency and severity of cognitive impairment demonstrated in both FAS groups were greater than would have been predicted on the basis of IQ alone<sup>392</sup>.

## **Intervention Programs – Detailed Findings**

Treatment and management approaches addressing primary and secondary disabling effects arising from FASD are evolving. These are based on increased understanding of brain structure and function and what happens when domains are impaired through prenatal exposure to alcohol. Currently, interventions are focussed on improving individual daily functional capacity and are cornerstones to improving behaviours resulting from impaired brain functions. Interventions may include both those directed at the affected individual and those which also support family members and caregivers.

Interventions specific to education or justice programs, or which utilise pharmacological interventions for individuals with particular FASD neurocognitive profiles under medical management have been excluded from this review. Rather, interventions which have been developed, trialled and evaluated for participants with FASD or similar conditions and which are integrated across settings or are primarily community based have been included. Collaborative interventions which involve family/caregivers, teachers or community support workers, and/or therapists in an integrated response to an individual's needs have been identified as particularly relevant to NDIA planning.

As the evidence base for interventions is emerging, reference is made to both higher and lower level evidence (i.e. Level 1 – sufficient rigour associated with study design and outcomes; and Level 2 – limitations in study design, but of sufficient quality as emerging evidence). Twenty three quantitative studies of sufficient quality are included in the critical review with the intervention programs summarised in Table 1 and set out in detail in the Appendices. Emerging interventions have been addressed earlier in Section One. There are an additional seven intervention studies in progress or recently completed (Table 10). A number of these are funded by the CDC and appear to be high quality RCTs with adequate sample size. Five qualitative studies of sufficient rigour also contribute to the evidence base included in this review.

The review of interventions has also included principles currently informing intervention practice, and which therefore constitute current best practice guidance. This information has been extracted as planning principles and best practice indicators, and is described in the discussion.



The interventions described below have been designed to address the most significant and common areas of functional impairment.

### The Alert program®

Two studies utilised The Alert program® for Self-Regulation<sup>393</sup>, an existing neurocognitive training program that has been adapted for school aged children with FASD<sup>394 395</sup>. The original Alert program® curriculum design involved a group treatment which helped children improve self-regulatory skills using a “how does your engine run” concept to identify their arousal level, and then choose an appropriate strategy to alter this.

An adaptation of The Alert program® termed *Neurocognitive rehabilitation* aims to teach children to recognise their own individual areas of impairment and to develop strategies to compensate for areas of weakness, which allows them to build on their individual strengths and skills. It is based on the premise that the daily functioning of children with FASD is limited by their impairments in self-regulation including executive functioning<sup>396 397</sup>.

The aim of the first study was to evaluate the effectiveness of neurocognitive rehabilitation using The Alert program® with a group of foster and adoptive caregivers<sup>398</sup>. The goal was to improve executive functioning and emotional regulation in home and school environments. Seventy eight children aged 6 to 11 years with a diagnosis of FAS or alcohol-related neurodevelopmental disorders (equivalent to the Australian category of ND-AE) were randomly assigned to either the intervention condition or a control condition. Children in the intervention group received group neurocognitive rehabilitation (75 minute sessions over 12 weeks) while their parents received parent education. Results reported that children in the intervention group demonstrated significant improvements in executive and emotional functioning compared with the control group. This improvement was documented using the BRIEF (parent report measure of executive functioning) and the RATC (the measure of emotional problem solving).

The study presents a significant development in the treatment of self-regulatory dysfunction and executive functioning difficulty with children with FASD. It gives children some autonomy to address their own self-regulatory needs and patterns of dysfunction and starts to use strategies to improve their self-regulation and emotional problem solving within a group setting.

Limitations of the study included the generalizability to all school aged children as the children were in foster or adoptive homes. Further research is needed to assess executive functioning in a range of settings such as home or school with children who live with their family of origin and those in other care settings.

The second study, The Alert program® was used with a group of 25 children aged 8-12 years diagnosed with FASD, and examined whether the Alert Program® for Self-Regulation would improve self-regulation and be generalizable to other aspects of executive functioning, behaviour and social skills<sup>399</sup>. As above, the BRIEF was used and selected subtests from the NEPSY-II, CANTAB, and the Test of Everyday Attention for Children (TEA-Ch) to measure attention and executive functioning. The main finding of this study was that parent rated improvements were seen in self-regulation, most significantly with aspects of emotion regulation. There were some gains seen on the child direct measures. The children appeared to improve more on simple tasks rather than more complex ones, which fits with the research that shows that children with FASD are challenged by more complex tasks requiring executive functioning.

## MILE

Three studies have examined the Math Interactive Learning Experience (MILE) which is a socio-cognitive habilitative program that focuses on improving mathematics performance and behaviour. This program targets cognition, behaviour, and academic function; all components of neurobehavioural functioning. Mathematics difficulties are a common deficit in children with FASD<sup>400</sup><sup>401</sup> <sup>402</sup> <sup>403</sup>. This intervention was developed from evidence which investigated children with brain damage (not caused through alcohol exposure) and supports the view that interventions need be contextualised to the environment of the individual<sup>404</sup>. As such, it is best to choose a functional skill that is impacted by brain damage and then construct an intervention to remediate the deficit (i.e. working memory).

The MILE program targets 3 to 10 year old children providing a comprehensive intervention across several levels of functioning, including intensive, short term, individual instruction for the child as well as training for caregivers and teachers. Underlying neurocognitive problems are accommodated by targeting mathematic skills. The design takes into account problematic caregiving histories often reported in children with FASD as well as behavioural regulation and attentional problems experienced by this group of individuals<sup>405</sup> <sup>406</sup>. Acknowledging that many children with alcohol exposure are in foster care or have families with limited social and economic resources, the intervention design attempts to incorporate the support needed for the family to allow the child to achieve 'readiness to learn' through parent education in behavioural regulation information and FAS/caregiver advocacy.

In the 2007 study, 61 children were assigned to either the mathematics intervention group (MILE) or a standard psycho-educational contrast group (SPC). Fifty six participants completed post-testing 1, with 2 dropouts in the intervention group. The average duration between pre and post testing was 149 days for both the mathematics intervention and the SPC groups. Outcomes were reported in Readiness to Learn, with significant gains in knowledge for both FAS/caregiver advocacy and behavioural regulation. Caregiver ratings of both internalising and externalising behaviour problems were reported with significantly less total problem behaviour scores (1, 50)=15.4,  $p<0.000$ ). Both groups demonstrated gains in maths knowledge, but significantly higher gains were found in the group receiving the maths component ( $p<0.04$ ).

A 6 month follow up study of the MILE intervention confirmed that both math skills and behaviour of children had improved significantly. Participants from the original 2007 study were contacted and reassessed. Math performance was assessed using standardised measures of maths achievement and behavioural problems as reported by caregivers on the CBCL. Children in the MILE group demonstrated significantly greater scores on maths outcome measures and the behaviour ratings were improved over pre-test scores in both groups<sup>407</sup>.

Very recently, the MILE program was evaluated in a community translation involving 60 participants across three treatment groups: speciality care centre, community, and parent maths instruction only. The study evaluated instructor satisfaction with the program, knowledge related to FASD/MILE, and parental satisfaction with the outcomes. Those in the MILE intervention groups demonstrated more positive gains in maths skills than the parent instruction only group. Both the community translation

and the MILE instructor training program were well received and effective in producing positive treatment outcomes<sup>408</sup>.

### Children's Friendship Training

Three studies have examined child friendship training (CFT) for children with FASD<sup>409 410 411</sup>. Social impairment is well documented amongst children with FASD due to neurobehavioural deficits in overall intellectual functioning<sup>412</sup>, inattention and hyperactivity, executive functioning and memory problems<sup>413 414</sup>. Children with FASD have problems understanding social cues, difficulty communicating in social contexts<sup>415</sup> and have fewer social skills than unexposed children, as reported by caregivers<sup>416</sup> and teachers<sup>417</sup>.

Key features of CFT were taught through: instruction of simple rules of social behaviour; modelling; rehearsal and performance feedback during treatment sessions; rehearsal at home; homework assignments; and coaching by parents during play with a peer<sup>418</sup>. They focussed on a range of critical social situations which discriminate accepted from rejected children.

The first study with a group of 100 children aged 6-12 years diagnosed with a FASD (11 per cent children with FAS, 43 per cent pFAS, 46 per cent ARND) compared CFT (n=51) versus delayed treatment control (n=49)<sup>419</sup>. Children in the CFT group showed significant evidence of improvement in their knowledge of appropriate social behaviour, and parental reports showed that CFT resulted in improved social skills and fewer problem behaviours compared with DTC. These gains were maintained at a three month follow up. The DTC group showed similar improvement after receiving their treatment. These treatments showed children benefit from CFT but the gains in social skills were not necessarily transferred to the classroom. The therapists who delivered the intervention were Psychology Interns with specific training in the delivery of the program.

This study was the first controlled treatment for improving the social functioning of children with FASD. A follow up study in 2012 replicated the study in a community setting with 85 children aged 6 to 12 years with documentation of prenatal alcohol exposure (PAE)<sup>420</sup>. Children were assigned to either the CFT or community Standard Care (SC) condition and then treated in 6 cohorts with approximately 7 children in each condition. Both conditions consisted of 12 sessions of 90 mins each delivered over the course of 12 weeks. Sixty seven children completed the 12 week study (CFT = 32; SC = 35), representing an attrition rate of 21 per cent. Children participating in the CFT group showed significantly improved knowledge of appropriate social skills, improved self-concept, and improvements in parent reported social skills compared to children in the SC condition. Results revealed that children with PAE performed as well as children without PAE. CFT children yielded greater gains than a community SC intervention for children with and without PAE.

The third study examined CFT with stimulants and neuroleptics in FASD<sup>421</sup>. Seventy seven children aged 5-11 years of age were given 12 sessions of CFT. Children were also prescribed stimulant or medication, neither, or both types of medication. Children with FASD have deficits in attention and executive functioning that are similar to those with ADHD and commonly are diagnosed with ADHD and placed on stimulant medication. However, research evidence suggests that their behavioural and attentional profiles differ<sup>422</sup> and variable effects of stimulant medication have been reported in the literature<sup>423</sup>. Neuroleptic medications may be helpful in improving behaviour in children with FASD as prenatal alcohol exposure is linked to dopamine and noradrenaline neurotransmitter disturbance<sup>424</sup>. The outcomes of this study indicated that according to parent and teacher reports on the Social Skills

Rating System (SSRS) children prescribed neuroleptic medication showed greater improvements on all outcome measures when compared to children not prescribed neuroleptics. Specifically, after 12 weeks of CFT, statistically significant improvement was revealed in children on four out of six outcome variables (self-control, assertion, problem behaviours and teacher reported self-control) who were prescribed neuroleptics. Children prescribed stimulant medication either failed to show improvement or showed poorer outcomes when compared to children not prescribed stimulants. This study has importance both for the efficacy of CFT as a treatment intervention when combined with neuroleptic medication, but also for the consideration of the diagnosis of ADHD when children may also have FASD<sup>425</sup>. Further, this study has implications for future FASD interventions which may be combined with medication prescribed for attention difficulties in FASD.

### **Mode of delivery for parent or caregiver education**

A study was conducted that explored different formats for delivering parent or caregiver education to families within the context of a randomised clinical trial with the aim to improve the functioning of children with FASD<sup>426</sup>. One arm of the trial consisted of community standard care with an information packet of basic FASD diagnostic information, neurodevelopmental consequences related to FASD impairments and how to access a variety of community services and information sources. The second arm consisted of two workshops delivered to caregivers. The first one educated parents about FASD and included methods of advocating for their child with FASD and the second workshop focussed on how to build positive behavioural regulation skills in children. The third arm was based on internet training.

The workshop group received the highest ratings on usefulness, understanding ability, overall satisfaction and willingness to recommend the information than the community group and received higher ratings than did the internet group on amount of information received and overall satisfaction. All three groups improved on their knowledge of behavioural learning principles but only the internet and workshop groups made significant gains in knowledge of FASD and advocacy. Improved behaviour was only found in the workshop and community conditions.

Future directions for web based learning may consider the use of 'webinars' where a therapist could deliver the talk, field questions, and allow for group discussions to see if behavioural outcomes were improved.

This study presents a distance learning model that investigates the efficacy of delivering FASD information to families that may live in rural areas and/or have significant barriers to obtaining services and access to community therapists.

### **Memory training**

Working memory is considered by some researchers to be a core deficit in FASD<sup>427 428 429</sup>. Rehearsal strategies are considered an effective tool to enhance working memory and in this study, verbal rehearsal strategies were used<sup>430</sup>. Thirty three children aged 4 to 11 were included with an aim to increase working memory for numbers. The children were divided into two groups. The intervention group received training and the control group did not receive training. All subjects had a FASD diagnosis. In a pre-post-test design, the children received rehearsal training and completed a digit span memory task for baseline scoring. Rehearsal training was instructions from the examiner encouraging the child to whisper what they had heard and repeat this over in their head. Post-test 1

was completed after a short break from the pre-test. Post-test 2 occurred on average 10.6 days after the first session (range 6-21). Outcomes were reported with children in the experimental group showing a significant increase in performance across intervention sessions, while the control group did not. Children in the intervention group performed significantly higher than the control group on post-test 2 but not on pre-test or post-test 1. More children in the intervention group showed behavioural evidence and self-report of rehearsal after training. Performance on the digit span tasks did improve suggesting that this intervention has some promise.

This intervention study provides some evidence that rehearsal training might enhance the working memory of children with FASD. And as the intervention focussed on improving memory of numbers it might be relevant to addressing the deficits in mathematics frequently reported children with FASD.

### **Language and literacy**

Two studies have examined language and literacy in children with FASD<sup>431 432</sup>. The first, a pilot study, examined targeted language and literacy training (LLT) with South African children. Sixty five children with either FASD (n= 40) or no prenatal exposure to alcohol (n=25) were part of a larger ongoing study that included metacognitive and family support interventions<sup>433</sup>. The aim of the study was to determine the effectiveness of a classroom language and literacy program to improve the academic skills in children exposed to high levels of alcohol. The children with FASD were assigned to either LLT (20) or FASD controls (20). This intervention began when children were between 9 to 10 years old and was administered for 30 minutes twice a week by an experienced speech and language therapist. The treatment was carried out over 9 months (a total of 38 hours). There were no significant gains by the LLT intervention group compared to control groups on the general scholastic assessment battery, however, there were significantly greater improvements in the LLT intervention group compared to the FASD control group in language and early literacy. Significant cognitive improvements were evident in this study which targeted specific areas of difficulty with a very vulnerable population in South Africa.

A case study involving one child aged 9 years, with FASD, used pre and post assessment of social communication<sup>434</sup>. The child had a complex cognitive and behavioural profile. The intervention was carried out over 6 weeks, with a mixture of individual sessions (2 weeks) and then 4 weeks of group sessions. Two peers were also enrolled in the intervention, both with diagnoses of FASD. Role play of social scripts and a checklist was used to guide the children through a routine for resolving social situations. The checklist used a verbal script to guide the children's thinking and participation while taking turns during the role play. The intervention improved the child's social communication skills. Limitations of the study included the lack of a plan to target generalisation of the intervention components at the onset. A longer term intervention may see greater improvements in social communication.

## Computerised Progressive Attention Training (CPAT)

One study aimed at improving attention abilities in children with FASD was found<sup>435</sup>. This was a small study with ten children aged 6-15 years. The time spent was an average of 16 hours over 9 weeks at school with the support of a research assistant providing metacognitive strategies and support. The intervention in the study utilised the CPAT targets proposed attention networks. It has been utilised for children with ADHD and significant improvements in reading comprehension and passage copying were seen<sup>436</sup>. Process specific attention training has benefits for children with varying causes of attentional deficits such as Traumatic Brain Injury, ADHD and cancer<sup>437</sup>. As discussed previously, ADHD is a common comorbid diagnosis in FASD<sup>438</sup> and attention difficulties are significant for children with FASD<sup>439 440</sup>. More complex aspects of attention are particularly affected in FASD, most notably the ability to shift the focus of attention and encode new information. Process specific approaches may be particularly useful to remediate underlying attention abilities by performing repetitive tasks while gradually increasing the attentional demands (cognitive load and processing speed).

Metacognitive strategies are those that teach children to monitor their own thinking in order to allocate attentional resources and use additional learnt strategies when performance is weak. Utilising pre and post intervention assessments, significant improvements were noted on several measures of attention including sustained and selective attention. Children had an increase in reaction time on the Test of Attentional Performance for Children (KiTAP) which represented an ability to be less impulsive. There was a decrease in reaction time on the Attentional Network Test adapted for Children (ANT-C) and increase in scores on the Test of Everyday Attention for Children (TEA-Ch) all of which are essential to learning in a dynamic and changing environment, at home and school. Several measures of spatial working memory, math fluency, and reading fluency also significantly increased, suggesting that the attentional gains can lead to better cognitive performance. The results of this attention training project represent a positive change in behaviours with parent reports from their home environment on tasks requiring sustained attention. It also represents a feasible program that can be delivered within the school setting and can be delivered by school educators with specific training for the intervention.

## Computer games as teaching devices

Due to cognitive impairments associated with FASD, children may find themselves in situations where they are more likely to injure themselves. In other populations demonstrating cognitive impairments, a multi modal presentation of information allows individuals to access different areas of their memory when recalling information<sup>441 442</sup>. Virtual reality (VR) computerised games allow multiple sensory inputs to be presented in a format that is interesting to children and allows individualised learning rates.

Two small studies were included that have utilised computer games as teaching devices. In the first pilot study 5 children participated in a computerised game to teach fire safety skills. Immediately post intervention and 1 week post-test the children were tested on skills learnt and generalisability to the real world. All children reached 100 per cent accuracy on the computer intervention as they were able to complete each of the safety steps at post intervention and at the 1 week follow up<sup>443</sup>.

In the second study, 32 children aged 4-10 years with FAS and pFAS learned fire and street safety through computer games that employed “virtual worlds” to teach recommended safety skills. This built on the previous study with a larger sample of children and the inclusion of street safety. Children

with attention problems may need more repetitions than typically developing children do to encode information. An approach which enables them to learn at their own rate, with repetitions as often as the child want, is likely to assist in retaining information. Children showed significantly better knowledge of the game to which they were exposed, immediately post intervention and at follow up, with the majority (72 per cent) being able to generalise all four steps within a behavioural setting<sup>444</sup>.

Conclusions drawn by the authors of the study suggest that computer games may be useful in educating children with FASD in methods for preventing unintentional injuries and can be introduced at home and in educational settings.

## **Family-focused Interventions**

### **Families Moving Forward Program**

The Families Moving Forward (FMF) Program is a tailored behavioural consultation intervention designed for families of preschool and school aged children with prenatal alcohol exposure FASD to improve behavioural outcomes, caregiver sense of self efficacy and meet the needs of the family. The FMF Program combines motivational interviewing and positive behaviour support techniques with caregiver support and education. A randomised control trial for this program involved recruiting children from a very large clinical database in the USA, through the Washington State FAS Diagnostic and Prevention Network<sup>445</sup>. A sample of 52 children aged 5-11 years and their caregivers were enrolled. The children in the study all had evidence of clinically significant externalising or attention problems, with clear neuropsychological impairment and marked adaptive function problems across multiple developmental domains. They all had IQ of >70. The intervention is a behavioural consultation intervention that is sustained, affordable and designed to be delivered in home or clinic-based settings. It lasts 9-11 months with at least 16 every other week sessions of at least 90 minutes duration. Other medication or community services are received concurrently if the child requires these.

In this study, children and caregivers received either the FMF intervention or community Standard Care (SC). Results showed a significantly improved sense of parenting self-efficacy immediately post treatment. A greater percentage of caregivers in the FMF group reported engaging in more self-care behaviours than parents in the SC group (72 per cent vs 44 per cent). Family needs were met more often as reported by the FMF group compared with the SC group (highly significant). Parents receiving the FMF were very happy with services received, scoring the intervention as (mean) 6.51 out of a possible high score of 7. Children in the FMF intervention group showed a significantly decreased number of challenging disruptive behaviour problems as measured on the Eyberg Child Behaviour Inventory (ECBI), a treatment-sensitive parent report measure.

These data indicate a positive effect on parenting attitudes and a beginning point to reducing child disruptive behaviours. Further research in the USA is currently underway. The FMF Program is feasible, can be provided with at least some sources of existing mental health funding in the USA and Canada, and is currently being disseminated.

New developments have allowed the FMF Program to be delivered in shorter sessions (60 minutes) or in 90 minutes sessions over a somewhat shorter time period (7 months). Training to teach clinicians in how to use the FMF Program is available in the USA, and clinicians from the USA and other countries have received training.

## Coaching Families Program

The impact of FASD on families has been evaluated with a Family Intervention Program<sup>446</sup>. The main aim of this study was to conduct a preliminary evaluation of the coaching families program (CF) which aids families and caregivers raising children with FASD. Mentors work with the families to educate them about FASD, access resources, and advocate on their behalf. Retrospective data from 186 families were analysed pre to post program. A crucial component of this program is in establishing and maintaining strong relationships between the mentors and families, allowing the mentors to identify strengths and look at family resources and supports and means of accessing community supports.

There was a significant decrease in needs and increase in goal attainment from pre to post program. There was also a significant decrease in caregiver stress from pre to post program. Families reported high overall satisfaction with the CF program. The program goes beyond simple education about FASD and demonstrated improvements in other important family aspects such as housing and transportation, community involvement and parenting.

## Parent Child Interaction Therapy and Parenting Support and Management

Forty six children aged 3-7 years of age with FASD and their caregivers participated in this randomised control trial that aimed to reduce behaviour problems among children with FASD and decrease parenting stress among caregivers<sup>447</sup>. Behavioural difficulties are significant primary disabilities among young children with prenatal exposure to alcohol<sup>448</sup>. Traditional parenting methods may not be responsive to the behavioural problems experienced by caregivers of children exposed prenatally to alcohol<sup>449</sup>. Significant parenting stress has been reported by caregivers of children exposed to alcohol when compared to those not exposed to alcohol<sup>450</sup>.

This study evaluated two evidence based treatments, Parent-Child Interaction Therapy (PCIT)<sup>451</sup> which provides both parents and children with coached practice of parenting skills and a parent only Parenting Support and Management (PSM) program combining components from other behavioural programs<sup>452 453</sup>. Both treatment conditions were carried out over 14 weeks, with weekly sessions of 90 minutes. A high attrition rate was reported in this study (approximately 46 per cent completed the entire 14 week treatment program, although this was reported as within norms for children referred for externalising behaviour problems)<sup>454</sup>. There were no statistically significant differences in improvement between the two intervention conditions, although improvement over time across outcome measures was noted. It would be necessary to carry out this intervention in comparison with a control group or no treatment condition to assess it appropriately.

Parental attitudes towards the program were positive with 80 per cent of the PSM group reporting they would recommend it to a friend. Of the 46 per cent of the PCIT parents who completed the program all felt that their presenting problems had been improved, with 70 per cent of PSM parents reporting the same outcome.

There are several limitations of this study; the small sample size, the high drop-out rate and reduced examination of all potentially important outcomes. It is indeterminate as to whether the improvements in parent report measures can be translated to other environments such as home and school.



## Adult Interventions

### Parent Child Assistance Program

One study evaluated the effectiveness of intervention with adults with FASD<sup>455</sup>.

A 12 month pilot community intervention for 19 young women with FASD in Washington State and Seattle, USA was developed from interventions with high risk women, to determine if these interventions could be applied to the FASD population. The intervention model used in this study was the Parent Child Assistance Program (PCAP), a three year case management model that was expanded to include women diagnosed with FASD. The study aimed to describe how PCAP and community service providers collaborated to better understand and serve clients with FASD; describe demographic and psychosocial characteristics of the 19 PCAP clients with FASD and compare client connection to key services at baseline versus at conclusion of the 12 month pilot intervention.

Improved outcomes, including decreased alcohol and drug use, increased use of contraceptives and medical and mental health care services, and stable housing, were obtained by implementing a community intervention model of targeting education and collaboration with key service providers, and by using advocate case managers as facilitators.

### Step by Step Program

The effectiveness of a community based intervention for parents with FASD utilised the Step by Step program in which mentors work with parents affected by FASD on a one to one basis<sup>456</sup>. The program is designed to work with a client for a maximum of 3 years. The mean length of time spent in the program was 24.5 months. Mentors helped clients identify and work towards meeting their needs and achieving their goals. A retrospective analysis of 24 closed client files revealed very high and significant needs of the clients identified upon intake to the program, with 71 per cent of the clients reporting experiencing at least one type of abuse in either childhood or adulthood or both. Sixty five per cent reported two or more types of abuse; 63 per cent of the sample reported past or present alcohol/and or drug use. The three most common categories of help clients identified as needed were: parenting; relationships and peers; and behavioural problems.

There was a significant reduction in the clients' needs from pre to post program ( $F(1,23)=10.75, \{<.01$ ). Clients' goals scores also increased significantly pre to post program. Goals included parenting, self-reliance and community connection.

## Clinical trials and pilot studies

### Infancy/Early Childhood Interventions

#### Attachment Focussed Intervention

Another clinical trial evaluating the effectiveness of an attachment focussed intervention in preschool children (2-5 year olds) with FASD is currently underway in the USA. This intervention will use the Circle of Security (CoS) in a home based setting, with an aim of improving caregiving and child outcomes in families with children with FASD. The outcomes hypothesised are a change in Attachment classification as a measure of psychological adjustment of the child, a change in the Parenting Scale (which is a measure of dysfunctional parenting practices), as well as a change in the parenting stress index (which is a measure of parental stress and the parent child interaction style).

There will be 36 sessions carried out over 9 months with a 3 month follow up, comparing the treatment group receiving CoS with a wait list control group<sup>457</sup>.

## **School Aged Interventions**

### **The Fitzroy Valley Alert Program®**

The Alert program® cluster randomised control trial is being conducted to develop, implement and evaluate a teacher delivered curriculum version of the program across nine primary schools in the Fitzroy Valley, a remote location in the Kimberley region of Western Australia. A pilot study was conducted at another Fitzroy Valley school in 2015 with the purpose of conducting a process based evaluation of the program training, delivery, resources and outcome measures. Between 2016 and 2017 all children in primary school at the study sites will receive The Alert program® in their regular classroom. Participants in the study will be predominately Aboriginal children who are enrolled in years one to five at a study school and who have attended school 20 per cent of the time preceding data collection. A family workshop will also be delivered three times within each community during the intervention period to support parents/caregivers to develop knowledge of The Alert program® concepts. The fourth year (2018) of this project will focus on translation of the findings to other sites.

### **GoFAR - Intervention for Affective and Metacognitive Control in FASD**

The GoFar program focuses on a core area of deficit identified in FASD; that is, affective and cognitive control that are central to behavioural and adaptive disorders from infancy through young adulthood. GoFAR is developing methods to address these problems in young children by bringing together computer game technology and behavioural techniques for affective and cognitive control. The methodology is based on previous work that suggests that a metacognitive technique (FAR: 'F' Focus/Plan, 'A' Act, 'R' Reflect) is helpful in improving behavioural and educational outcomes in FASD. An important element is the GoFAR computer game, a 'serious game' that supports the understanding and development of metacognitive control techniques and reinforces its use by clinically-referred children, ages 5 to 10 years. This game is incorporated into a 10 week, manualized intervention program which focuses on improving adaptive functioning and behaviour.

Currently, a clinic trial of a pilot group of 30 alcohol-affected children and their caregivers is being carried out. Outcome measures will include both standardized assessments suitable for a clinical setting and measurement of changes in physiological response. Pre and post testing allow a preliminary understanding of the usefulness of the program elements, including the GoFAR game, as well as the suitability of the outcome measures selected. More comprehensive evaluation of the efficacy of this methodology in treating the neurodevelopmental disorders associated with FASD is planned<sup>458</sup>.

### **Computer game interventions for FASD**

Direct targeted interventions have been shown to improve attention and working memory. Underlying brain functions have been shown to be responsive to change in other areas of disability such as ADHD. These have taken account of the plasticity of the brain and neurocognitive advances in rehabilitation of the brain<sup>459</sup>.

There is a pilot study (2010) and an efficacy study (2011) with results reported using a computerised training program called Cognitive Carnival, which combines working memory, inhibitory control and attention training with the use of metacognitive strategies<sup>460</sup>. Strategies utilised include: rehearsal, visualisation and reducing speed. Scaffolding techniques are used to help facilitate performance which involve helping children to think about how they may improve their performance by repeating sequences back in their mind before attempting to answer. The domains targeted for intervention in this study build on previous studies which have utilised computerised attention training for children such as the CPAT in children with FASD<sup>461</sup> also included in this critical review.

Children aged 6-12 years (n=18) were randomly assigned to either the intervention group n=9 (Cognitive Carnival) or control group n=9 (educational computer games) and participated in 24 sessions for 30 minutes over 12 weeks. Outcomes were reported with clinically significant improvements evident in memory, attention, inhibition and quantitative tasks. Significant limitations identified include low power due to small group size and pre and post measures not being sensitive enough.

A further study utilising a delayed treatment control approach with 6-15 year olds (n=21) was completed. Subjects were tested at 3 time points – 0 weeks, 12 weeks and 24 weeks. Metacognitive strategies were used among the children and adolescents to aid their performance. Improvements were seen on executive functioning tasks and increased mastery of metacognitive strategies. There were large differences in performance found both between and within individual participants across time, which underscored the variability within this population.

## **Adolescents/Young Adult Interventions**

Two projects, funded by the Centre for Disease Control in the USA are described in detail below.

### **Project Step Up (University of California)**

Fifty two young people aged 13 -18 years were included in a group intervention. Project Step Up<sup>462</sup> was designed to provide parents with tools to facilitate their teens' difficult transition to adulthood and to better understand the neurocognitive deficits associated with prenatal alcohol exposure and alcohol misuse in adolescents. The aims were to achieve a decrease in alcohol risk scores and alcohol related negative behaviours in the intervention compared to the control group. The intervention consisted of six 60 minute sessions delivered over the course of 6 weeks. Caregivers and adolescents completed concurrent sessions with pre and post intervention measures completed as well follow up at three months post intervention.

Results showed that adolescents reported consuming lower levels of alcohol and fewer high risk behaviours following intervention in the light/moderate lifetime use than those in the light/moderate control group. Outcomes were fewer drinks consumed on the weekend and over the last month in the light/moderate lifetime use group compared with the control group. However, these results were not sustained over the 3 month follow up period.

### **Partners for Success (St Louis University)**

This project aims to reduce maladaptive behaviours among adolescents and young adults (16 -25 years of age) with FASD. The therapy is anticipated to reduce these behaviours for participating youth with FASD, reduce stress among their families, and improve the quality of life for all participants. It is a six month intervention, with a three month follow up. There are two groups in the randomised control

trial - community standard care alone (CS) and Partners for Success<sup>463</sup> (PFS) utilising a local agency in St Louis, USA.

The intervention uses a team of clinicians, therapists, and mentors to:

- Engage each family in family therapy during a 12-month period (in biweekly sessions) to address their unique needs.
- Provide life coaching to youth and young adults with FASDs during a 12-month period (in biweekly sessions).
- Online and in person caregiver support group.
- Mentor the young adult (towards decreased maladaptive behaviours, increased adaptive behaviours, increased self-esteem, and improved school/work performance).

Results support some improvement but not statistically significant compared to controls. Further research and replication required in other areas and with other agencies.

### **Distance learning parent training interventions**

The Strongest Families program uses input from key stakeholders to determine the need for services and supports and develop and evaluate a distance training program for parents/caregivers. The purpose of the study is to see if a new Strongest Families program, made especially for children with FASD, works to help families deal with difficult behaviours. Two hundred parents/caregivers of children ages 4-12 years with a FASD diagnosis and mild to moderate behaviour problems, who are living in Canada, will take part in this study.

The Strongest Families FASD intervention will be delivered over the Internet via a website using IRIS (Interactive Research and Intervention Software). This program eliminates the need for families to travel to an intervention site on a regular basis, as the intervention can be completed in families' own homes and participants can be recruited from across the country<sup>464</sup>.

Strongest Families modules for behaviour and anxiety disorders as well as night time bedwetting have been rigorously tested in randomized trials and shown to be effective with lasting effects one year later, targeting mild and moderate conditions<sup>465</sup>. The Strongest Families program is serving families across Canada, in Finland and soon in Vietnam.

## Appendices

**TABLE 8: Domains of Impairment**

Author/Year/ Location	Recognised Neurocognitive behavioural impairments		Standardised assessment of impairment		Formal diagnosis related to FASD (recognised criteria)		Sample size (population, or selected) and age
	Score	Impairments	Score	Assessment	Score	FASD Criteria	
Tamana et al 2014 Canada	2	Academic achievement, language, verbal memory, Executive Functioning, visual motor integration, motor abilities	2	WRAT, PPVT-R, CAVLT, VMI, Halstead-Reitan Battery, WISC, WAIS, WRIT	2	Institute of Medicine Criteria	117 (All ARND) 5-17 years
Rasmussen 2011 Canada	2	Executive functioning Working memory, visual memory, and attention	2	CANTAB	2	4 digit diagnostic code	24 (12 FASD)/26 control 6-17 years
Rasmussen 2009 Canada	2	Executive Functioning and age related differences	2	D-KS Executive Functioning	2	4 digit diagnostic code	29 cases 8-16 years
Rasmussen 2006 Canada	2	Intelligence, Memory, executive functioning and attention	2	WISC-III, WPPSI-R, CMS, BRI Executive Functioning, CRS-R	2	4 digit diagnostic code	50 6-15 years
Janzen et al 1995 Canada	2	Intellectual functioning, visual perceptual, fine motor, speech and language, behavioural problems	2	MSCA, Wisconsin Fine Motor Steadiness Battery (Grooved Peg Board component)VMI, Florida Kindergarten Assessment Battery (RD), (TELD), (CBCL)	2	Diagnosis by criteria of the Research Society on Alcoholism	10 Preschool FASD/10 controls 3.5-5 years
Green et al 2009 Canada	2	Executive Functioning, attention, planning and spatial working memory	2	CANTAB	2	Canadian Diagnostic guidelines	89 FASD/92 Controls 8-15 years
Kerns 1997 Canada	2	Complex attention, verbal learning and Executive Functioning	2	WISC-R, WAIS-R, WRAT-R, CVLT, CTT, APT, COWAT, RFF	2	US Diagnostic guidelines at University of Washington	16-27 Young adults
May 2014 USA	2	Verbal IQ, Working Memory, General and conceptual ability, adaptive behaviour	2	VMI, Differential Ability Scales- 2 <sup>nd</sup> ed, VABS	2	IOM diagnostic guidelines related to FASD	36 FASD First graders ~ 7 Years Controls 168 Population sample

Mattson et al 2013 USA	2	Neurobehavioural profile - Executive Functioning Differences between ADHD and AE	2	CANTAB, D-KEFS, WISC-IV	2	Diagnosis made by member of CIFASD Dysmorphology Core	468 AE = 209, Controls = 185 ADHD = 74 8-17 years
Streissguth 2007 USA	2 range	Attention, arithmetic skill, spatial -visual memory and IQ	2	<b>Neonates:</b> Brazelton Neonatal Assessment Scale <b>8 months:</b> Bayley Scales of Infant Development <b>4 yrs:</b> WISC Wisconsin Motor Steadiness Test <b>7-8 yrs:</b> WISC-R, WRAT-R, PRS, CPT, CMT 7 <b>11-14yrs:</b> WISC-R, WRAT- R, CPT, WCST 21 yr: WAIS-R, TONI-2, WRAT-R WCST, CPT	2	Dysmorphology examination to determine diagnosis	500 0-25 years
Astley 2010 USA	2	CNS Functional Profiles – Intelligence, Visual Motor Sensory Executive Functioning/Memory and Behavioural	2	WISC, VMI, RCFT, CBCL	2	4-digit diagnostic code	1400 Newborn to adult
May 2013 South Africa	2	Verbal and non verbal ability and digit span performance, behavioural problems.	2	WISC-IV, Achenbach Teacher Report Form	2	IOM Diagnostic System	157 First graders ~ 7 years
Mattson & Riley 1998 USA	2	Memory	1	Tests selected from 5 mutually exclusive sets of 10 words. Word Stem Completion Test, Free Recall and Recognition Memory Test, Controlled Oral Word Association Test , Category Fluency Test	1 18 with FAS	Not specified	21 18 with FAS, , 3 with heavy prenatal exposure 8-18 years
Roebuck-Spencer et al 2003	1	Corpus Callosum anomalies Attention and problem solving	2	Bimanual Coordination Test (c-BCT)	2	Dysmorphology examination confirmed diagnoses	21 FASD 10-19 years

USA							13 = FAS 8 = PAE
Roebuck 2002 USA	1	Interhemispheric transfer of information in the corpus callosum	1	WISC-III, Finger localisation task?	1 14 FAS; 8 PAE	Dysmorphology examination confirmed diagnoses and full neuropsychological test battery.	24 FAS/PAE 20 control 8-15 years
Ware et al 2012 USA	2	Executive functioning and its relationship to Adaptive Behaviour	2	VABS D-KEFS	2	Clinical Examination by a member of the Collaborative Initiative on Fetal Alcohol Spectrum Disorders (CIFASD) Dysmorphology Core	357 8-18 years 3 groups 142 = Heavy Prenatal Alcohol Exposure (38 FAS) 60 (ADHD) ADHD group (82) 133 = CON group
Carr et al 2010 Canada	2	Sensory processing Adaptive behaviour	2	SSP, ABAS-II, WISC-IV/WPPSI-III	2	Canadian Guidelines for diagnosis (Chudley, 2005)	46 3 groups 15 pFAS and 16 ARND 15 PEA 3-14 years
Jirikowic et al 2008 Canada	2	Sensory Processing Sensory Motor Performance	2	SSP, BOTMP, QNST-II NEPSY Sensorimotor and Visuospatial Processing, SIB-R, WRAT-3, SSRS, TONI-2	2	4 digit diagnostic code, 2004 version (Astley, 2004)	25 5-8 years Clinic referred
Jirokovic, Kartin & Olson 2008 Canada	2	Sensory processing Adaptive behaviour School performance	2	SIB-R, TONI-2	2	4 digit diagnostic code	25 FASD 23 controls 5-8 years
Wengel et al 2010 Canada	2	Sensory Processing Sleep disturbances	2	Sensory Profile, CHSQ	2	Canadian Guidelines for Diagnosis related to FASD	31 19 FASD/12 control 3-6 years
Thomas et al 1998	2	Adaptive behaviour – social skills	2	WISC-R, VABS	2	Research society on Alcoholism criteria	45 3 groups (n=15) (15 with FAS) 5-12 years
Fagerlund et al 2012 Finland	2	Adaptive behaviour (communication, daily living skills and socialisation)	2	VABS, WISC-III, WAIS-III, LIPS-R (Brief IQ Screener)	2	Dysmorphology examination confirmed diagnoses using modified Institute of Medicine Criteria	73 FASD, Learning disordered children IQ matched n = 30 and 40=control

							Children and adolescents
Kodituwakku et al 1995 USA	2	Self-regulation, working memory	2	ACAT, Controlled Oral Word Association Test WRAML, WGTA, WCST	1 8 FAS; 2 FAE	Diagnostic criteria proposed by Fetal Alcohol Study Group of the Research Society on Alcoholism	10 FAS/FAE/10 control 9-18 years
Lucas et al 2014 Australia	2	Gross motor	2	GMDS: Locomotion GMDS: Locomotion M-ABC: Ball skills P-CTISB-2: Static Balance RT & movement	2		5 studies pooled Sample size range: 10 to 45 in intervention group Total sample sizes: 20 to 400 7months-13 years
Doney et al, 2014 Australia	2	Visual motor skills (visual-motor integration, eye hand coordination, and visuomotor precision Fine motor dexterity Grip strength, finger tapping & praxis	2	Beery VMI Paediatric Environmental Neurobehavioural Test Battery: VMI test GMDS Eye & Hand Coordination Subscale NEPSY, M-ABC, Pegboard tests, Detroit Tests of Learning Aptitude, WFMSB	1	FASD diagnostic criteria provided in studies various	22 studies FASD 4 to 12 years
Streissguth et al 2004 USA	2	Secondary disabling effects Sexual behaviour Disrupted school experience Trouble with the law Confinement Alcohol and drug problems	2	WISC WAIS-R WRAT VABS	2	Dysmorphologists at the University of Washington diagnosed FAS or FAE.	415 6-51 years 155 with FAS and 260 with FAE
Spohr et al 2007 Germany	2	Life course impairments Disruptive and externalising behaviours Attention problems Limited occupational options Dependent living	2	YABCL	2	Diagnosis made by Research Society on Alcoholism diagnostic criteria	37 ~0 to 30 years (not reported accurately)



**TABLE 9: Interventions (Quantitative)**

Author/ year	Study type and purpose	Discipline providing intervention	Specificity What is involved, who is it for, why is it important	Sampling strategy # participants	Outcome measures/impact	Limitations	Kmet Score as %
<b>ALERT INTERVENTION PROGRAM – Level 1 Evidence</b>							
Nash et al 2014	RCT Treatment intervention	Occupational therapists	Alert Program (improving targeting self-regulation which is part of Executive Functioning) for children with FASD	25 children (14 test; 15 control) 8 to 12 years	12 week program Treatment effects on cognitive and socioaffective Executive Functioning domains. At 12 week follow up - Treatment group displayed significant improvements in social cognition and inhibitory control. Parents also reported improved behavioural and emotional regulation. These behavioural improvements continued and were stable at the 6 month follow up.	Non randomised, small sample, no control for confounding	19/24  79%
Wells et al 2012	RCT	Psychologists	Neurocognitive habilitation group treatment approach (ALERT) for children with either FAS or ARND	78 children (40 treatment; 38 control) 6 to 11 years	Measures were BRIEF – a self-administered (caregiver) questionnaire assessing Executive Functioning behaviours in home and school environments. Children in the intervention group demonstrated significant improvements in executive and emotional functioning compared with the control group. The Roberts Apperception testRATC	Differences in outcomes between the treatment and control groups impacted by extensive feedback and comprehensive recommendations made by assessing psychologist following initial screening	23/24  99%
<b>MATH INTERACTIVE LEARNING EXPERIENCE INTERVENTION PROGRAM (MILE)</b>							
Kable et al 2014	RCT Treatment Intervention	Speciality Centre, Community based and Parent Instructors	MILE for children with FASD, delivered by different instructors including parents for 15 weeks.	60 children (20/19/21) across 3 intervention types	Intervention comparison (speciality centre vs community instruction vs parental instruction) pre and post test. Community translation of the MILE program and MILE	More difficult to ensure intervention efficacy (ie completion within a given time frame) when completed in	22/24  91%

				(final numbers with attrition 18/15/19) not statistically significant 6 year olds	instructor training program was well received.	community due to time and family based needs.	
Kable et al 2007	RCT Intervention program with 3 components	Paediatric geneticist, psychologists and educators	Parental education for all (workshops) and then randomisation to either the intervention group with specialised tuition focused on math (MILE) or the standard psychoeducational care groups	61 children 3 to 10 years	Parental (satisfaction); children pre and post test for gain with caregivers reporting few problem behaviours. Both randomised groups showed gain, but children in focussed math tuition group demonstrated a gain of over 1 SD on any of 4 math outcome measures	Sample group is not population based so this restricts the generalizability to the whole population or other cultural groups. Costly as the program is delivered 1:1, may not translate to a school environment.	21/24 87.5%
Coles et al 2009	Pre and Post	Teachers	Persistence of effects of a math performance and child functioning (using MILE) at 6 months post intervention	54 children (28 intervention; 26 contrast) 3 to 10 years	Intervention targeted at improving behaviour and pre-academic and academic (specifically math) functioning in children and supported this as effective in remediating academic deficits associated with prenatal alcohol exposure		19/24 79%
<b>WORKING MEMORY STRATEGIES – Level 1 Evidence</b>							
Loomes et al 2008	Pre and Post		Executive Functioning of children diagnosed with FASD involved in brief rehearsal training to improve memory (numbers based). Important background for tailoring interventions aimed a memory improvement	33 children (16 control; 17 experimental) (19 aboriginal) 4 to 11 years	The experimental group showed a significant increase in digit span score across the three sessions and on post test 2. Rehearsal training was effective in improving memory for numbers	Small sample, very brief and specific intervention. No non FASD control group.	19/22 86%
<b>CHILDREN'S FRIENDSHIP TRAINING (CFT) INTERVENTION – Level 1 Evidence</b>							
O'Connor et al 2012	RCT intervention study (with random	Multidisciplinary professionals (psychiatrists,	CFT versus standard of care (SOC) for children with Prenatal Alcohol	85 children (41 CFT group; 44 SOC group)	Children in CFT group showed significantly improved knowledge of appropriate social skills, overall	Absence of independent evaluation of child	20/24 83%

	assignment to CFT or SOC)	psychologists, social workers, family therapists, speech therapists)	Effects (PAE) or without PAE. 12 sessions of 90 mins duration.	6-12 years Attrition: 67 completed study (CFT 32; SOC 35)  6 to 12 years	self concept compared to the SOC group. Parent satisfaction was higher in the CFT group (90.7%) compared to 68.6% in the SOC group, in relation to their confidence in the child's ability to get along better with other children.	behaviours in a natural setting. Possible overestimation of children's behaviour change by parents in the CFT group. Potential difficulty generalising to children outside of the study group.	
O'Connor et al 2006	RCT Intervention study (with random assignment to CFT or DTC)	Psychologists	Testing the efficacy of child friendship training (CFT) with FASD children using intervention and delayed treatment control (DTC) groups.	100 children (51 to CFT; 49 to DTC)  6 to 12 years	Test of Social Skills Knowledge (TSSK) and Social Skills Rating System (SSRS). No statistical significance between the groups at baseline; CFT showed significantly improved knowledge of appropriate social skills compared with DTC group at testing.	Treatment performed in a highly controlled setting and effectiveness requires testing in more typical community settings.	20/24  83%
Frankel et al 2006	Pre and Post	Psychologists	Role of community psychopharmacological management of children with FASD and capacity for peer social interactions after exposure to Children's Friendship Training (CFT)	77 children  5 to 11 years	Children grouped according to medication status (stimulants, neuroleptics, antidepressants, and non stimulants). Social Skills Rating System – 55 items, evaluated by parent and teachers. Combined with CFT, children prescribed neuroleptics showed greater improvement on all outcome measures. Combined with CFT, children prescribed stimulant medication failed to show improvement or showed poorer outcomes.	Significant effects on 3 out of 6 outcome measures, may have been due to higher baseline values for children prescribed neuroleptics. Study investigators did not formally verify medication compliance as there are no checklists specifically for FASD. (they exist for ADHD)	20/24  83%
Bertrand 2009	RCT	Mental health providers	Evaluate an intervention FMF designed to improve caregiver self-efficacy,	52 children (26 FMF intervention	As tested, FMF is a feasible, low-intensity, sustained model of supportive behavioural	Some outcome measures were created for the study	19/24  79%

(Study 5)	Stratified randomised block design	meet family needs and reduce child problem behaviours for children with FASD when compared with community standard of care	group; 26 community standard group)  5 to 11 years	consultation lasting 9-11 months – 16 fortnightly sessions of 90 min duration. FMF intervention showed significantly improved sense of parenting self-efficacy and reduced child disruptive behaviour immediately post treatment. A greater percentage of caregivers parents in FMF group reported engaging in more self care behaviours than in community group 72% vs 44% p <.05 . There was a Highly significant effect (p<.01) in that the FMF group reported that family needs were met more often than in the community group.
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**MILE INTERVENTION PROGRAM – Level 1 Evidence**

Kable et al 2012	RCT (treatment condition) Training Intervention	Parents – Web based group Investigators or graduate clinical psychology students – parent workshop group	Parents of children with diagnosed FASD received one of three educational interventions (community group as standard; workshop; or web based) aimed at improving child functioning 6 week intervention	59 children (18/23/18) across 3 intervention types 75% of community group and 79.3% of workshop group completed post testing Internet group highest attrition rate (9 of 11 did not log in at all) not statistically significant.	Knowledge and satisfaction (parents); pre and post test against – caregiver advocacy, and behavioural regulation (as measured using Child Behaviour Checklist). All forms of parent education have some benefits dependent on degree of alcohol related impairment, severity of behavioural symptoms and overall goals of intervention.	Only individuals with alcohol-related dysmorphology included, therefore not generalised across spectrum of alcohol affected children Sample group is not population based so this restricts the generalizability to the whole population or other cultural groups.	22/24  91%
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**LANGUAGE AND LITERACY TRAINING – Level 1 Evidence**

Adnams et al 2007	Pre and Post (with randomisation of FASD diagnosed participants to intervention or control groups and a non-exposed control group)	Teachers	FASD children assigned to language and literacy training (LLT) or FASD controls (20 each),	65 children 40 FASD children intervention or control groups; 25 non-alcohol exposed group  9 year olds	General cognitive and scholastic tests for baseline performance and longitudinal change (at nine months) using teacher and parent questionnaires, classroom observations and LLT tests with participants	Cognitive mechanisms not adequately assessed prior to intervention.	19/24  79%
<b>COMPUTER GAME INTERVENTION – Level 1 Evidence</b>							
Coles et al 2006	Pre and Post (with randomisation of subjects to one of two groups)	investigators	Effectiveness of computer games as teaching devices for children with FAS or pFAS diagnosis	32 children (16 each in two exposures – fire or street safety  4 to 10 years	Children were both test and control after exposure to one game. All children pre tested on both safety skill sets then randomised to one experimental condition, Children showed significantly better knowledge of the fire and street safety game immediately and at 1 wk follow up. 72% were able to generalise all four steps within a behavioural setting.	Games developed relevant to a specific diagnosis so not generalizable to other developmental disabilities and not compared to other methods of safety education	18/24  75%
<b>COMPUTERISED ATTENTION TRAINING INTERVENTION (CPAT) – Level 1 evidence</b>							
Kerns et al 2010	Pre and post	Assistant teachers	Efficacy of direct CPAT intervention on improving attending in children with FASD	10 children with FASD  6 to 15 years	16 hours over ~ 9 weeks at school Pre and post assessments included descriptive, working memory, attention and academic measures – maths and reading fluency with demonstrated significant gains. Greatest improvements were noted on decreased scores on distractibility test and decreased reaction times on the ANT-C and increased scores on auditory sustained attention test.	Small sample, no control group, and observed changes could be due to CPAT training or other effects.	15/20  75%
<b>LANGUAGE AND LITERACY TRAINING – Level 2 Evidence</b>							
Timler et al 2005	Case study	Psychologist	Improving social communication capacity (linguistic and social cognitive skills) based on 2	1  9 year old	Promoted mental state verb production in the subject, with overall performance		14/22  63%

individual and 4 group sessions to improve responses to social situations using modelling of socially appropriate responses based on linguistic cues

demonstrating changes in cognition.

**COGNITIVE CONTROL STRATEGIES - Level 2 Evidence**

Adnams et al 2003 In: Riley et al, 2003	Pilot study – pre test/ post test controlled intervention	Therapists	Determine the feasibility of applying intervention strategies to children with FAS in an educational setting. Using CCT or usual classroom determine the effect on cognitive processing and behaviour of children with FAS.	10 children with FAS 5 intervention group 5 control group  8 year olds	Significant results in behavioural domain.	As it was a pilot study, limited by small sample size. Duration of intervention was less than what has been shown to be effective in other studies. Lack of suitable tools to measure outcomes for qualitative evidence.	10/24  42%
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**VIRTUAL REALITY INTERVENTION – Level 2 evidence**

Padgett, Strickland & Coles 2006	Pre and post case series		Pre-post case series design to evaluation VR format as an instructional tool with children with FAS or pFAS	5 children  4 to 7 years	All five children correctly identified fire safety components 100% of the time and four correctly performed all three safety steps in both real world and picture sequence test 100% after training	Very small sample size, brief follow up time (1 week)	14/22  63%
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**PARENT CHILD INTERACTION THERAPY (PCIT) AND PARENTING SUPPORT AND MANAGEMENT (PSM) – Level 2 evidence**

Bertrand 2009 (Study 4)	RCT	Parents	To evaluate two group based interventions for children with FASD to reduce behaviour problems and decrease parenting stress using PCIT and PSM	46 (treatment and comparison)  3 to 7 years	Prior to intervention – parents completed questionnaires (PSI; ECBI); and observation of parent-child interactions with DPICS-II). PSI and ECBI completed each week. Improvements noted over time across outcome measures and intervention conditions, but not statistically significant.	Small sample size, and not all potentially important outcomes were examined.	16/24  67%
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**FAMILY SUPPORT AND MANAGEMENT – Level 2 evidence**

Leenaars et al 2012	Pre and Post Intervention	Community based social services	Efficacy of and satisfaction with support, education, advocacy and referrals for families with children with FASD as provided through a specialised family support agency	186 families (30% foster family, 23% adoptive, 19% biological, 9% permanent guardianship <1 to 23 year old dependents 66% confirmed FASD diagnosis, 27% possible FASD	Pre-test included needs assessment (37 item) and goals development, Stress Scales (for parents) and anonymous completion of satisfaction surveys by parents Significant decrease in needs from caregivers and increase in goal attainment pre to post program. Significant decrease in caregiver stress and an overall high satisfaction from families with the program.	Family stress scales not psychometrically tested; program specific	14/22 63%
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**PARENT CHILD ASSISTANCE PROGRAM (PCAP) REMODELLED – Level 2 evidence**

Grant et al 2004	Intervention study (nested)	Case manager	Young women diagnosed with FASD enrolled in a pilot intervention remodelling a standard PCAP to meet the specific needs of the FASD group	19 young women	Aimed at improving diagnosis of FASD and comorbidities, addressing substance misuse, improving birth control, all of which showed improvements	Case specific, with small sample and no control.	14/22 63%
Denys et al 2009	Retrospective case note review	n/a	Retrospective review of closed cases from the Step by Step program modified from the PCAP for parents diagnosed with FASD. Client matched with mentor for 3 years.	24 adults(closed case files) 19 to 47 years	Based on the outcomes in the Hull Outcome Monitoring Evaluation system (HOMES) inclusive of needs (40 items), goals (21 pre-written plus mentor designed) and progress assessments. Of 17 clients 82% agreed the mentor helps learn to solve problems. 100% agreed that the mentor motivates them to take care of their own health and 62% reported an improvement in health over the past year.	Small sample size, some measures based on self-report, not all participants with a definitive FASD diagnosis	15/24 62%

**TABLE 10: Interventions (Qualitative)**

Author year	Study type and purpose	Specificity What is involved, who is it for, why is it important	Sampling strategy # participants	Outcome measures/impact	Limitations	Kmet Score as %
Petrenko, Tahir, Mahoney & Chin 2013	Investigating systems-level barriers contributing to secondary conditions and interfering with prevention and treatment	Focus groups and individual interviews with parents and providers to establish the most commonly identified barriers to accessing the required services and supports to prevent secondary conditions related to FASD. Relevant to assisting with planning approaches	25 parents of children with FASD 18 service providers (n=43)	Part of a larger study investigating the wants and needs of the community for prevention intervention for children with FASD. Demonstrated an overarching construct of lack of knowledge of FASD across multiple systems, and, five specific systems level barriers including: delayed diagnosis, qualifying for services, availability of services, implementation of services and maintenance of services	Findings may not reflect the views of all families and providers. Did not include teachers as service providers.	18/18 100%
Petrenko, Tahir, Mahoney & Chin 2014	Investigating the elements required in intervention programs for individuals with FASD from the perspective of those with a lived experience (parents and providers)	Focus groups and individual interviews with parents and providers to establish characteristics for preventing secondary conditions in individuals with FASD	25 parents of children with FASD 18 service providers (n=43)	Five ideal program characteristics for the prevention of secondary conditions in individuals with FASD are: 1) Availability of programs across the lifespan; 2) Programs have a prevention focus; 3) Are individualised; 4) Are comprehensive; 5) Are coordinated across systems and developmental stages  These are consistent with the Positive Behaviour Support (PBS) framework.	None noted	18/18 100%



Pei, Job, Poth & Atkinson 2013	Assessments for educational interventions of children with FASD	Semi-structured focus groups with providers and caregivers to identify comprehensive picture of assessment requirements and intervention implications. Highlighted that a) assessment and diagnosis are only as useful as the accommodations and supports that follow; b) varied learning and behavioural needs of children with FASD require assessments that are geared toward informing intervention practices in order to achieve best possible results; c) inclusion of teachers in assessment process.	31 teachers, 7 administrators, 16 allied health professionals, 6 caregivers (n=60)	Three major findings: Increasing the scope of the assessment process to reflect the <i>whole child</i> ; the <i>assessment process</i> needs to be responsive and find success (cannot be solely deficit focussed); <i>building school capacity</i> via communication of assessment outcomes and relevant recommendations for learning and development to all stakeholders including teachers.	Potential community bias as a result of snowball sampling. Lack of geographic and cultural representative-ness	17/18 94%
Rutman and Van Bibber 2009	Examining the challenges, accomplishments and support needs of adults with FASD in relation to parenting their children	Semi-structured interviews, and focus groups using video ethnography (video and audio taping) to establish the challenges and successes of adults with FASD in relation to their parenting.	15 adults with FASD; 8 support people; 36 service providers (various) (n=59)	Identified the main themes as: 1) Parenting hopes and goals; 2) Parenting accomplishments and strategies; 3) Parenting challenges and barriers; 4) Challenges associated with societal attitudes or expectations 5) Policy related barriers for parents with FASD Attesting to the need for ongoing support for adults with FASD, with support not gated by IQ-related criteria, but rather by functional need.	None noted	16/18 89%

Burnside and Fuchs 2013	Investigating the lived experiences of youth with FASD transitioning from child welfare care to adulthood	Semi-structured interviews with youth living in care to establish their concerns as they transition from education into adulthood, employment and alternative living arrangements	20 youth aged 16-21 years (10 females; 10 males) (n=20)	<p>Main themes identified were:</p> <ol style="list-style-type: none"> <li>1) Role of FASD in the lives of youth in care;</li> <li>2) Importance of relationships;</li> <li>3) Adolescent development for youth with FASD in care;</li> <li>4) Additional challenges during adolescence;</li> <li>5) Preparing for age of majority</li> </ol> <p>Highlighting the dysfunction which has affected their lives (including separation from family of origin and multiple care placements over time); difficulties in planning for the future; limited opportunities to gain skills required for independent living; and the self-reported presence of a range of secondary behaviours typically associated with FASD (anger, aggression, anxiety, poor self-concept, addictions, and mental health problems).</p>	Small group of participants who identified as having FASD. Not generalizable to other populations – although consistent with the known literature.	16/18 89%
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**TABLE 11: Emerging evidence**

Author year	Study type and purpose	Discipline providing intervention	Specificity What is involved, who is it for, why is it important	Sampling strategy # participants	Outcome measures/impact	Limitations
<b>CLINICAL TRIALS</b>						
Infancy/Young children						
Hanlon-Dearman, A University of Manitoba	Evaluating the Effectiveness of an Attachment-Focused Intervention in Preschool Children with FASD. Randomised control trial – waitlist control		Circle of Security (COS) Family Intervention in improving caregiving and child outcomes in families with children with FASD delivered in a home based setting.	N = 12 2 to 5 years	Change in Attachment Classification – measure of psychological adjustment of children. Change in the Parenting Scale – measure of dysfunctional parenting practices Change in the parenting stress index – measure of parental stress, parent-child interaction style	
Children						
Fitzpatrick, J 2015	Pilot study The Alert program®: A tailored evidence based treatment program for Aboriginal children living with FASD	Teachers and education assistants	Develop, Implement and, evaluate a school based therapeutic program to improve impairments in executive functioning and self-regulation for Aboriginal Children in the Fitzroy Valley.	Children born between 2003-2007 attending 8 primary schools in the Fitzroy Valley		
Pei et al, 2010	Pilot study using Computerised intervention training program Randomised control trial	Psychologists	Cognitive Carnival – combines working memory, inhibitory control and attention training as well as metacognitive strategies. 24 sessions for 30 mins over 12 weeks (experimental received Cognitive Carnival intervention) Control group	6 to 12 years 18 children 9 experimental 9 control	Measures: Developmental Neuropsychological Assessment (NEPSY). Working Memory Test Battery for Children (WMTB-C), the BREIF MRIs to look at measurable changes in the brain. Clinically significant gains made pre to post test – improvements most significant in memory,	Small sample size, hence lack of statistical power. Pre and post measures not sensitive enough to detect changes. Large differences between experimental and control group.

			played educational computer games.		attention, inhibition and quantitative tasks.
Pei, et al 2011	Efficacy study using the Computerised intervention training program Delayed treatment control approach	Psychologists	Cognitive Carnival – combines working memory, inhibitory control and attention training as well as metacognitive strategies 24 sessions for 30 mins over 12 weeks. All received the Cognitive carnival intervention.	6 to 15 years 21 children	Measures: Tasks of Executive Control (TEC), Test of Performance-Children’s Version (KITAP), Spatial Span (WISC IV). Measures taken pre, mid (12 weeks) and post test (24 weeks). Some preliminary significant gains pre to post intervention and continued improvement at 12 weeks post intervention. Improvements seen on executive functioning tasks and increased mastery of metacognitive strategies.
Coles & Kable (2015)	Clinical trial GoFAR program		Parent training, child training and behaviour analogue sessions where the parent and child get to implement their new knowledge in the context of learning basic life skills. Includes a computer game that teaches children to control impulsive and problematic behaviour. 10 weeks of 60 min sessions.	5 to 10 years	To improve self-regulation and adaptive living skills through teaching children metacognitive control techniques – F – A – R (Focus/Plan) (Act) (Reflect) can help in improving behavioural and educational outcomes.

Adolescents/Young Adults

O’Connor et al (n.d.) <sup>466</sup>	Project Step Up		Group intervention that addressed strategies to cope effectively with the pressures associated with alcohol misuse for	N = 52 13-18 year olds	Decrease in alcohol risk scores and alcohol related negative behaviours in the intervention compared to the control group. Positive treatment response was	More frequent intervention required during the follow up period.
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adolescents and their caregivers – coping with stress, handling parenting challenges, attitudes to alcohol and alcohol related social pressures.  
6 x 60 minute concurrent sessions over 6 weeks.  
Pre and Post intervention plus 3 month follow up.

not maintained over the 3 month follow up period

Tenkku et al (n.d.) 467	Partners for Success (PFS)	Psychologists	For Youth/young adult and parents utilising Combination of In home family therapy, Online/In-person caregiver support group using Cognitive Behavioural Therapy (CBT) and Solution Focused Therapy (SFT) as well as Mentorship of young adults modelled on the P-CAP program  6 month intervention with 3 month follow up	N = 42 13-25 year olds 2 group community standard (CS) alone or CS + PFS	Setting: 25 mile radius around City of St. Louis. Adolescents: To increase # of adaptive behaviours (decrease # of maladaptive behaviours), Improve self esteem, improve performance at school and work Parents – decrease parental stress, increase parent empowerment, coping and problem solving skills. No significant differences noted between 2 groups although the PFS group did improve or remain stable.	Small sample size Utilisation of local agency that was not currently a significant provider of interventions to adolescents with FASD
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Parent/Family Interventions

Turner et al (n.d.) <sup>468</sup>	Parenting training for Children with FASD. Randomised control trial.		Internet based intervention program for parents/guardians of children with FASD. Based on the Strongest families program.	4 to 12 years	A change on the Child Behaviour Checklist from baseline to 11 months. Measure of adaptive functioning and problems. A change on the Depression Anxiety & Stress Scale Short Form. Measure of parental distress.	
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**TABLE 12: Kmet et al (2004) Checklist for assessing the quality of quantitative studies**

	Criteria	Yes (2)	Partial (1)	No (0)
1	Question/objective sufficiently described?			
2	Study design reported and appropriate?			
3	Method of subject/comparison group selection OR source of information/input variables described and appropriate?			
4	Subject (and comparison group, if applicable) characteristics sufficiently described?			
5	If interventional and random allocation was possible, was it described?			
6	If interventional and blinding of investigators was possible, was it reported?			
7	If interventional and blinding of subjects was possible, was it reported?			
8	Outcome and (if applicable) exposure measure(s) well defined and robust to measurement/misclassification bias? Means of assessment reported?			
9	Sample size (appropriate)			
10	Analytic methods described/justified and appropriate			
11	Some estimate of variance is reported for the main results?			
12	Controlled for confounding?			
13	Results reported in sufficient detail?			
14	Conclusions supported by the results?			

**TABLE 13: Kmet et al (2004) Checklist for assessing the quality of qualitative studies**

	Criteria	Yes (2)	Partial (1)	No (0)
1	Question/objective sufficiently described?			
2	Study design evident and appropriate?			
3	Context for the study clear?			
4	Connection to a theoretical framework/wider body of knowledge?			
5	Sampling strategy described, relevant and justified?			
6	Data collection methods clearly described and systematic?			
7	Data analysis clearly described and systematic?			
8	Use of verification procedure(s) to establish credibility?			
9	Conclusions supported by the results?			
10	Reflexivity of the account?			

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