# SOCIOEMOTIONAL FUNCTIONING IN CHILDREN DIAGNOSED WITH ALCOHOL RELATED NEURODEVELOPMENTAL DISORDER (ARND): PROFILE ON THE CHILD BEHAVIOUR CHECKLIST (CBCL)

by

Rachel Greenbaum

A thesis submitted in conformity with the requirements
for the degree of Masters of Arts
Department of Human Development and Applied Psychology
Ontario Institute for Studies in Education of the
University of Toronto

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Rachel Lindsay Greenbaum, Masters of Arts, 2000
Department of Human Development and Applied Psychology
Ontario Institute for Studies in Education of the
University of Toronto

## Abstract

In an effort to better understand the socioemotional challenges of children exposed prenatally to alcohol, children aged 4-18 diagnosed with Alcohol Related Neurodevelopmental Disorder (ARND) were compared to normal controls. The sample of 68 children included 33 with ARND, 33 normal controls matched to the ARND sample for age, gender, and SES, and 2 children with ARND for whom matches were not available. Comparisons were made both quantitatively and qualitatively using a standardized measure of socioemotional functioning. The study investigated the hypothesis that children with ARND would present with a distinct clinical profile of disturbed socioemotional functioning. Furthermore, it was hypothesized that the severity of socioemotional disturbance would reflect familial and background factors. Results indicated that the ARND group did differ significantly than controls in their presentation of socioemotional problems. As well, children in the ARND group presented with a consistent clinical profile of socioemotional functioning on the CBCL.

## Acknowledgements

This research was supported by the Reva Gerstein M.Sc. Studentship and the RESTRACOMP Research Training Studentship through the Research Institute at the Hospital For Sick Children. I am grateful for the financial assistance and opportunities made available to me through HSC.

I would like to extend a deeply heartfelt thank you to the following individuals for their guidance, support, and encouragement throughout this endeavor: My supervisors, Dr. Joanne Rovet and Dr. Jennifer Jenkins; Dr. Gideon Koren; Mohamed Abdolell; Sandra Hepworth and Patricia Arseneau; my family.

An extraordinarily special thanks to my husband, Hal Niedzviecki.

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Term Definitions (adapted from Institute of Medicine [IOM], 1996)

Alcohol Related Birth Defects (ARBD):

Congenital physical anomalies associated with prenatal alcohol exposure, including malformations and dysplasias (i.e. cardiac, skeletal, renal, ocular, auditory abnormalities)

Alcohol Related Neurodevelopmental Disorder (ARND):

This term is applied to individuals with neurodevelopmental problems that are associated with prenatal alcohol exposure. They may or may not present with any or all of the physical manifestations of prenatal alcohol exposure. The diagnosis involves behavioural and/or cognitive abnormalities that are inconsistent with developmental level and cannot be explained by familial background or environment alone.

Fetal Alcohol Effects (FAE):

This term was initially used to describe adverse birth outcome that could be proven to be related to alcohol exposure in utero. The term was meant to apply to animal models of teratogenesis and large prospective group studies of humans exposed to alcohol prenatally, rather than individual patients. However, this term has been used frequently in the lay literature to refer to individuals affected by prenatal alcohol exposure who do not present with full-blown FAS.

Fetal Alcohol Syndrome (FAS):

Patients with this diagnosis present with all the clear phenotypic features including those relating to face, brain, and growth (variations of this diagnosis take into account confirmed versus unconfirmed histories of alcohol exposure).

## Abbreviations

| | |
|---|---|
| ADHD | Attention Deficit Hyperactivity Disorder |
| ARND | Alcohol Related Neurodevelopmental Disorder |
| ARBD | Alcohol Related Birth Defects |
| CNS | Central Nervous System |
| FAE | Fetal Alcohol Effects |
| FAS | Fetal Alcohol Syndrome |
| FARA | Fetal Alcohol Related Abnormalities |
| FIQ | Full Scale Intelligence Quotient |
| IQ | Intelligence Quotient |
| PIQ | Performance Intelligence Quotient |
| SES | Socioeconomic Status |
| VIQ | Verbal Intelligence Quotient |

# Chapter One: Introduction

Alcohol exposure in pregnancy leads to characteristic symptoms including brain dysfunction and growth dysmorphology [known as Fetal Alcohol Syndrome (FAS) and Alcohol Related Neurodevelopmental Disorder (ARND) respectively]. These conditions contribute to many problems including psychopathology, learning disabilities, sexual abuse and sexual deviance, substance abuse, and trouble with the law (Streissguth, Barr, Kogan, and Bookstein, 1996). Until very recently, research has focused specifically on FAS while ARND has been mostly neglected. Cognitive difficulties have been emphasized whereas the mental health issues pertaining to prenatal alcohol exposure have been greatly ignored. It is important to study socioemotional functioning among these children because a high proportion of individuals with FAS/ARND have been found to require mental health services as adults (Famy, Streissguth, and Unis, 1998).

Despite increasing awareness about the relatively poor prognosis among individuals prenatally exposed to alcohol, little is known about the early manifestations of socioemotional disturbance in children with ARND. Since socioemotional deficits may precede the onset of psychopathology, information about these problems can facilitate in early identification and development of prevention/intervention strategies. The purpose of this investigation is to identify the nature and severity of socioemotional disturbance among children with ARND. The present study addresses a gap in previous research, first, by studying ARND specifically, rather than FAS, and second, by studying socioemotional functioning specifically in children with ARND, rather than cognitive/academic functioning.

## Historical Background

Fetal alcohol exposure is the most prevalent single cause of intellectual impairment in children in the western world to date (Kaemingk and Paquette, 1999; Korkman, Autti-Ramo, Koivulehto and Granstrom, 1998). It is also the most common preventable cause of birth defects and the leading cause of mental retardation ahead of Down Syndrome and cerebral palsy (Korkman et al. 1998; Nulman, O'Hayan, Gladstone, Koren, 1998). Of all forms of substance abuse, alcohol represents by far the most serious problem, whether judged by its frequency or capacity to harm the fetus (Institute of Medicine [IOM], 1996).

Although alcohol's role in human teratogenicity was not systematically studied until the late 1970's, adverse effects of alcohol consumption during pregnancy have been noted throughout history (Abel, 1990). Indeed, the first scientific study of children of alcoholic mothers was reported by a British physician, Dr. William Sullivan, in 1899. However, until the last few decades little attention was paid to the plausibility of alcohol's teratogenicity. In 1968, an article in France by Lemoine et al. provided the first description in the medical literature of the effects of alcohol on the fetus. It was not until 1973, with the independent observation of Jones and Smith (1973), that a distinct dysmorphic syndrome associated with gestational alcoholism, Fetal Alcohol Syndrome (FAS), was coined and recognized in the medical literature.

The criteria for diagnosis of FAS is based on the presence of a triad of features: 1) pre and/or postnatal growth retardation (weight, length, and/or height <10th percentile) 2) CNS damage (signs of neurologic abnormality, developmental delay, or intellectual impairment), and

3) characteristic facial dysmorphology (i.e. microcephaly, poorly developed philtrum, thin upper lip, and flattened maxillary area) (Abel, 1990).

However, very few alcohol-exposed children present with the full-blown syndrome, especially all the facial features listed for FAS. Moreover, of the dysmorphic characteristics listed, most are not "disfiguring" and, in fact, many lead to appealing or attractive looking faces. Children under age ten are often described as "elfin" or "pixie-like" (Berg, Kinsey, Lutke, and Wheway, 1995). Further, facial features often tend to fade with age and may become undetectable by adolescence (Berg et al. 1995; Spohr and Steinhausen, 1984,1987). The absence of facial features poses an additional problem in that these children essentially look "normal", and so are expected to be "normal", and yet they will not have escaped the damaging effects of alcohol's teratogenicity (Mattson, Riley, Gramling, Delis, and Jones 1998; Sampson et al., 1997).

Despite its wide-spread recognition, FAS encompasses a relatively small proportion of children prenatally affected by alcohol (Connor and Streissguth, 1996). One estimate is that only 10%-40% of the offspring of alcohol abusing women meet the criteria necessary for a diagnosis of FAS (Roebuck, Mattson, and Riley, 1998). To describe the large number of children affected by prenatal alcohol exposure who do not fit all of the criteria to meet diagnosis of full-blown FAS, terms such as Fetal Alcohol Effects (FAE), Alcohol Related Birth Defects (ARBD), Fetal Alcohol Related Abnormalities (FARA) and Alcohol Related Neurodevelopmental Disorder (ARND) have been used (IOM, 1996). Of the terms to be adopted within this field, ARND is the most recent and perhaps the most encompassing.

The combined incidence of fetal alcohol related abnormalities has been estimated in the general population to be about 0.91 percent, and 10 to 20 percent of the population in some Native communities (Sampson et al., 1997). A similar range, from 1-3 children out of 1000 in general obstetric populations, was reported by Korkman et al. (1998). Incidence appears to vary both within and between countries. For example, the incidence of FAS was found to be more than 20 times higher in the U.S. than in other countries (Nulman et al., 1998).

Much of the research to date has focused on children with FAS specifically to the exclusion of ARND (Korkman et al., 1998), and on their medical or cognitive difficulties rather than mental health issues pertaining to prenatal alcohol exposure (Roebuck, Mattson, and Riley, 1999).

## Diagnosing ARND

In 1996, the National Institute of Medicine formally established the diagnostic criteria for ARND. These include history of prenatal alcohol exposure in conjunction with:

A. Evidence of CNS neurodevelopmental abnormalities, as in any one of the following:

- decreased cranial size at birth
- structural brain abnormalities (i.e. microcephaly, partial or complete agenesis of the corpus callosum, cerebellar hypoplasia)
- neurological hard or soft signs, such as impaired fine motor skills, neurosensory hearing loss, poor tandem gait, poor eye-hand co-ordination ***and/or***

B. Evidence of a complex pattern of behaviour or cognitive abnormalities that are inconsistent with developmental level and cannot be explained by familial background or environment alone, such as:

- learning difficulties; deficits in school performance
- poor impulse control
- problems in social perception
- deficits in higher level receptive and expressive language
- poor capacity for abstraction or metacognition
- specific deficits in mathematical skills
- problems in memory, attention, or judgment (IOM, 1996).

Despite these guidelines, diagnostic issues in the field are far from resolved. The absence of facial characteristics in the majority of fetal alcohol affected children makes identification of the disorder extremely difficult. Without diagnoses, these individuals do not receive the services they require (Streissguth, 1994), and this may exacerbate the existing deficits. Moreover, the cognitive and behavioral difficulties associated with ARND impact detrimentally on interpersonal relationships, particularly within the family system. Subsequently, many individuals with ARND encounter disrupted home lives and ultimately end up isolated with few resources to deal with their problems. At the same time, it has been recognized that even a stimulating environment with sensitive parents or a good institution may not be sufficient to compensate for prenatal damage due to alcohol exposure (Nulman et al., 1998; Spohr, Willms, and Steinhausen, 1993; Steinhausen, Nestler, and Huth, 1982). To complicate matters further, FAS/ARND may be confused with other syndromes that present with similar physical features and/or cognitive and

behavioural profiles including: Aarskog syndrome, Williams syndrome, Noonan's syndrome, Cornela deLange syndrome, Trisomy 21(Down syndrome), Dubowitz syndrome, Stickler syndrome, Bloom syndrome, fetal hydantoin syndrome, maternal phenylketonuria fetal effects, fetal toluene syndrome, fragile X syndrome, velocardiofacial syndrome, Turner's syndrome, Opitz syndrome, and attention deficit hyperactivity disorder (Berg et al., 1995; IOM, 1996, Nulman et al., 1998).

## ARND and the Brain

A misconception exists that FAS is on the extreme negative end of a continuum, with ARND representing relatively less negative effects. However, this does not seem to be the case. A recent longitudinal analysis by Steinhausen et al. (1998) refuted earlier evidence (Spohr, Willms, and Streinhausen, 1993; Steinhausen, Nestler, and Spohr, 1982) and showed no linear relationship between degree of morphologic damage and intelligence. According to these researchers, "this may simply reflect the fact that dysmorphic features may be a crude measure of morphologic damage, especially of the brain" (Steinhausen et al. 1998). Mattson et al. (1998) recently compared children with histories of heavy prenatal alcohol exposure who had defining physical dysmorphology to children without dysmorphology and showed neuropsychological deficits in both groups, regardless of whether physical features were present or not.

Many studies of alcohol teratogenesis suggest that the brain is the most vulnerable body organ to the effects of prenatal alcohol exposure (Aase, 1994; Nulman et al., 1998; Streissguth, 1994). The associated patterning of brain damage related to prenatal alcohol exposure includes reductions or alterations in some or all of the following: basal ganglia, cerebellum, corpus callosum, and vermis. As well, a wide range of impairments at the

molecular and biochemical levels are seen and these contribute to such behavioural manifestations as minor learning disabilities, mental retardation, hyperactivity, distractibility, memory impairments, poor judgment and adaptability, impaired social skills, hyperresponsiveness to stress, and somatosensory and auditory problems (Nulman et al., 1998). Recent work has implicated early damage in the prefrontal cortex to impaired judgment and moral/social reasoning (Anderson, Bechara, Damasio, Tranel, and Damasio, 1999). These findings are very relevant to the field of fetal alcohol research given that these kinds of problems in judgment and reasoning tend to wreak havoc in the lives of many individuals affected by prenatal alcohol exposure.

## ARND and Cognitive Functioning

For individuals affected by prenatal alcohol exposure, IQ can vary from the very deficient to the average range. Subjects in Ann Streissguth's pivotal longitudinal study represented a gradually accrued group that began with the first patients diagnosed FAS in 1973 by Jones and Smith and ended with those who came to the University of Washington FAS Diagnostic Clinic between 1993 and 1995 (Streissguth et al., 1996). The clients (aged 3-51 years) were largely ascertained through clinical referral across a 22-year period and diagnosed by a small group of dysmorphologists. Results indicated a mean IQ of 79 for the FAS group and 90 for the FAE group.

Perhaps what was most notable among Streissguth's findings was that having a higher IQ did not assure these individuals a higher level of well-being. In fact, high IQ was found to be disadvantageous since correlational analyses revealed that ***low IQ*** had a "protective effect". Clients classified as mentally retarded, had lower rates of alcohol and drug problems, disrupted school experience, trouble with the law, and confinement

compared to individuals with IQ's above 85. However, despite this apparent "protective effect", all low IQ clients were in dependent living situations and had employment problems. Regardless of their IQ, groups obtained equally low scores on measures of adaptive functioning.

A recent study by Korkman et al. (1998) tracked expectant mothers who were "heavy" alcohol abusers (>10 drinks per week) while simultaneously supporting them in their efforts to abstain from drinking. It is important to note that the amount of alcohol consumed during for damage to occur has never been definitively established (Nulman et al., 1998). Accordingly, a >10 drinks per week cut-off is somewhat arbitrary. Korkman et al.'s (1998) study compared neuropsychological characteristics of three subgroups of prenatally exposed children and compared them to non-exposed controls.

In this study, children whose mothers were able to stop or reduce drinking during trimester I were compared to children exposed to alcohol during trimesters I and II, and to children who were exposed to alcohol throughout pregnancy. Although the groups did not differ significantly with respect to age and gender distribution, they did differ with respect to maternal education. The mothers of children in the exposed groups had significantly less education than mothers of children in the non-exposed control group. As such, the results of this study are somewhat ambiguous. It cannot be assumed that differences found between the subject and control groups are due to alcohol exposure alone. There is no way to determine the extent to which neuropsychological differences seen between exposed versus non-exposed children are related to the differences in maternal education either apart from or in combination with alcohol exposure in utero.

Nevertheless, consistent with previous reports (Streissguth, 1996) Korkman et al.'s results indicated impairments on composite scores of naming, receptive language, attention, and visual-motor production as well as a significant split between Verbal IQ (VIQ) and Performance IQ (PIQ), favouring the latter. Full scale IQ's (FIQ) were within the average range. Based on their findings, the authors concluded that the neuropsychological characteristics of children exposed to alcohol in utero seem to include linguistic problems in naming and phonological analysis that predispose to verbal learning problems at school, problems in attention and executive functions, difficulties in complex tasks of visuo-motor production (design copying), and problems with learning manual motor series. Relative assets were long-term memory as well as sensorimotor differentiation and precision.

With respect to the effects of duration of exposure, the group exposed throughout pregnancy was impaired in all the aforementioned domains. For children of mothers who were able to stop drinking during trimester II, naming was the only significantly affected composite test score. In contrast, exposure during trimester I only did not produce any significant impairment. Korkman et al.'s study should not, however, be interpreted to mean that the fetus exposed to alcohol during this period is impervious to the effects of alcohol teratogenicity. The study's findings should be considered in light of Nulman et al.'s (1998) critical review of the literature, which suggests that the fetal brain is vulnerable to the harmful effects of alcohol in the first trimester.

Presently, the exact temporal window of fetal vulnerability is not known but there is some evidence that a number of FAS expressions each have their own unique critical periods. First trimester alcohol exposure is critical for organogenesis and the distinctive

FAS facial dysmorphology. This is alarming because 50% of North American pregnancies are unplanned and a majority of women are not aware of their pregnancy during the first 4-6 weeks of gestation (Nulman et al., 1998). Subsequently, Nulman et al. (2000) studied the effects of exposure to binge drinking in the first trimester (>5 drinks per occasion) in non-alcoholic women. Results indicated that although cognitive development was not found to be significantly affected, behavioural dysfunction reflecting disinhibition was more common in this group.

In a comprehensive review of the research examining the cognitive and adaptive functioning of individuals exposed to alcohol prenatally, Kaemingk and Paquette (1999) conclude that, "there is converging evidence that prenatal alcohol exposure adversely impacts response inhibition, visuomotor abilities, and visual memory". They claimed further that even individuals with FAS or FAE who have "average" intellectual abilities still may have academic problems and may not be able to live independently as adults. To the question "Is prenatal alcohol exposure associated with a specific pattern of deficits?", the authors respond that, "interpretation of findings across studies is not parsimonious, and inconsistencies could be attributable to sample characteristics, methodological differences, and exposure factors". Regarding the issue of timing, the effects of a teratogenic agent are said to be "exquisitely related" to the period of development when exposure occurred (Kaemingk et al., 1999). Thus, it would not be surprising if prenatal alcohol exposure resulted in a variety of neuropsychological presentations depending on specific exposure characteristics and CNS vulnerability at the time of exposure (Kaemingk et al., 1999).

It is well-known that prenatal alcohol exposure is related to cognitive and behavioural deficits throughout childhood and adolescence. To date most research has focused on understanding and quantifying the cognitive profile of children with FAS with relatively less focus on ARND or behavioural or psychosocial adjustment. Very recently, research efforts have begun to provide empirical evidence that in addition to previously reported cognitive impairments, heavy prenatal alcohol exposure is related to significant impairments in psychosocial functioning, and, furthermore, even children without alcohol-related physical anomalies suffer from impaired psychosocial functioning (Roebuck, Mattson, and Riley, 1999; Thomas, Kelly, Mattson, and Riley, 1998). Thomas et al. (1998) broached the question of "whether social skills deficits in children with FAS are due to their general deficits in intelligence or whether the problems with social performance can be separated from IQ". They approached this question by comparing children with FAS to children, with similar deficits in intelligence, who were not exposed to alcohol prenatally.

The children (aged 5-12 years) were matched for Verbal IQ and compared using the Vineland Adaptive Scales. The higher SES among the normal control group represents a major confound to the study. The authors concluded that "social deficits in children with FAS are beyond what can be explained by low IQ scores", and yet the underlying cause of poor social behaviour in children with FAS (not to mention ARND) remains to be determined. For instance, in this study, social deficits may have been related to the differences in socioeconomic status apart from or in addition to, the effects of prenatal alcohol exposure. The authors also recognized the possibility that specific cognitive deficit such as "theory of mind" may underlie poor social skills. Theory of

mind is a higher level, metacognitive process involving in part, the ability to consider and anticipate the experience and perceptions (and subsequent beliefs, thoughts, and feelings) of others separately from the experience of the self. It seems likely that children with limited skills in this regard would be more prone to misinterpret events (others' actions, reactions, and intentions) and subsequently to respond in ways that are deemed to be socially inappropriate. Further study in this area is warranted given its potential for pin-pointing a major source of social and adaptive skills impairment among individuals affected by prenatal alcohol exposure.

## ARND and Attention

Problems with attention surface early and affect approximately 70% of children with ARND (Berg et. al., 1995). Hyperkinetic disorders were the most frequent type of psychopathology at both the preschool and early school age period in a subgroup of 27 children that were included in Steinhausen et al.'s research (1993; 1994) in Berlin. Oesterheld, Kofoed, Keppen, Johnson, and Skorey-Solberg (1998) estimated the prevalence of attention deficit hyperactivity disorder (ADHD) in 67 children with FAS using Conners Parent and Teacher Rating Scales. According to these authors, the prevalence of Attention Deficit Hyperactivity Disorder (ADHD) in the general population ranges between 3 and 6 percent. Based on their findings, Oesterheld et al. (1998) estimated that the prevalence of ADHD in children with FAS is at least 3 times greater and may be as high as 9 times greater than that found in the general population. It has also been suggested that because attention disorders tend to predispose to alcoholism, part of the attention disorders observed in children exposed to alcohol may actually stem from a genetic predisposition (Korkman et al. 1998; Steinhausen, 1982).

Many children exposed prenatally to alcohol are diagnosed with ADHD. Nanson and Hiscock (1990) reported no differences between parental ratings of children with FAS/E and ADD on standardized behavioural rating scales suggesting the social behaviour of these 2 groups is similar and that these groups of children are more hyperactive and inattentive than are control children. However, a fair degree of controversy surrounds the issue. Many professionals question the appropriateness of this diagnosis given that there seem to be significant qualitative differences between the attention problems in alcohol exposed versus "pure" ADHD samples.

Coles, Platzman, Raskin-Hood, Brown, Falek, and Smith (1997) investigated the extent to which children with documented prenatal exposure and physical features associated with FAS and fetal alcohol effects (FAE) show the same neurocognitive and behavioural characteristics as children with ADHD and no alcohol exposure. Coles et al. compared groups on traditional measures of cognitive abilities and behaviour and a profile analysis of different types of attention processes.

The results indicated that children with FAS and children with ADHD have distinct attentional profiles (Coles et al., 1997). Even though both groups were equally impaired intellectually, there was little similarity in their pattern of responses. Children with ADHD were less able to focus and sustain attention whereas children with FAS/FAE were less able to encode information and use it meaningfully in problem solving. Consequently, these researchers called into question the assumption that behaviours seen in children with FAS result from the same neurocognitive deficits as those seen in individuals with ADHD.

This research has important implications for treatment of attention problems in children with prenatal alcohol exposure. Medications such as methylphenidate which are used to treat ADHD symptoms, tend to act on and improve sustained attention (Coles et al., 1997). Stimulant medications, while helping to focus attention, may not improve learning or problem solving and this might explain why medications do not seem to work with many of these children. Although alcohol exposed children may share some behavioural characteristics with children diagnosed with attention deficit disorders, current reports on the behavioural profile of alcohol exposed children are not conclusive.

One seemingly important aspect that has not yet been explored in this field, is the relationship between attention deficits and other areas of functioning. In particular, no study has compared children with FAS/ARND to children with "pure" attention deficits on measures of socioemotional functioning. There may be much insight to be gained in such an endeavor in light of the fact that socioemotional problems have been associated with attention problems in non-alcohol exposed children (Barkley, 1997, 1998; Farone, Biederman, Weber, and Russell, 1998).

## ARND and Attachment

According to Steinhausen, Nestler, and Sphor (1982), children with FAS tend to experience: "a chain of detrimental circumstances, including feeding problems, failure to thrive, and repeated hospitalization in the neonatal and infancy periods", as well as higher rates of separation from their parents, "a factor generally believed to be deletrious to development". These children are more likely than others to suffer disturbances in bonding and other deficits in the mother-child relationship (Steinhausen et al., 1982).

Infants born to mothers who report drinking large quantities of alcohol during pregnancy show signs of CNS dysfunction at birth. They demonstrate increased irritability, autonomic instability, a decreased sucking response, motor immaturity, slow habituation, low levels of arousal, distorted sleep patterns, and withdrawal symptoms (Streissguth, Barr, and Martin, 1983; Coles, Smith, Fernhoff, and Falek, 1984; Coles, Smith, and Fernhoff, 1985). High-pitched crying, disturbed sleep, and feeding difficulties often follow withdrawal symptoms and may persist for days and weeks (Coles and Platzman, 1993). Behavioural difficulties often continue into the preschool period, reflecting difficulties in cognitive functioning and sustained attention, increased activity level, emotional instability, rigidity, and irritability (Landesman-Dwyer, Ragozin, and Little, 1981). These neurobehavioural effects may adversely impact on mother-infant interaction and future attachment relationships (Meares, Penman, Milgrom-Friedman, and Baker, 1982). Black, Bucky, and Wilder-Padilla (1986) described children of alcoholics as ignoring, withdrawing, and avoiding conflict. These children were self-reliant and unable to trust other people when they needed help and they grew up perceiving adults as uncaring and insensitive (Cork, 1979).

O'Conner, Sigman, and Kasari (1992), proposed that alcohol consumption following pregnancy was directly related to the mother's interaction with her child and this resulted in a negative affective response in the child and in insecure attachment. They tested the hypothesis that three independent and direct paths could be drawn between prenatal drinking and infant negative affect, maternal behaviour, and attachment behaviour respectively. The model was based on the possibility that alcohol consumption affected mother and infant independently. The results indicated that this group contained

a high number of disorganized infants (32%) and that the mothers of these infants were the heaviest drinkers. Mothers who drank more had infants who displayed more negative affect in interaction, and expressed insecure attachment behaviour. The mothers of these infants were less stimulating in the interaction process.

The effects of alterations in infant behaviour on infant attachment have been deemed the most significant result of prenatal exposure to alcohol (Nulman et al., 1998) while emotional and social aspects associated with heavy maternal drinking also conspire to weaken the maternal-infant bond. Poor quality of early attachment and daily care was cited first among a list of reasons for behavioural difficulties in a twelve-year follow-up of children exposed to alcohol in utero (Autti-Ramo, 2000). Research beyond the field of prenatal alcohol exposure suggests a possible link between insecure attachment in infancy and child behaviour problems (Campbell, 1995; Goldberg, 1997), thus highlighting the need to examine pathways for later maladaptation.

It must be noted that children of alcoholic mothers (many of whom are adversely affected by their exposure to alcohol in utero) are, for obvious reasons, frequently removed from their mother's care. In a German sample of 158 children (Steinhausen, Willms, and Spohr, 1993), 24.1% were living with foster or adoptive parents, 25.5% were living in institutions, and 24.1% had experienced various changes of their domestic status over time. These authors observed that, "diversification of domestic environment reflects the consequences of maternal alcoholism and disorganized family milieu". These circumstances point back to the higher risk for attachment problems among alcohol exposed children given the high proportion of disruptions within the context of their earliest (albeit possibly already disturbed) relationships.

Further, Steinhausen, Willms, and Spohr (1994), reported that milieu was a highly significant variable with children in institutions having by far the highest rates of psychopathology. A complicating factor was that children with the most severe cognitive impairment and dysmorphological damage were most likely to be institutionalized, and were more likely to have had both mothers and fathers who were alcoholic. To date, no study has yet succeeded in disentangling the effects of the teratogenic and environmental risk factors on the child's development (Nulman et al., 1998; Steinhausen and Sphor, 1998).

## ARND and Psychopathology

It is well known that children with FAS are at high risk of psychopathology. Steinhausen, Nestler, and Huth (1982) were one of the first groups of researchers to study psychopathological symptoms in children affected by prenatal alcohol exposure. Steinhausen et al. (1998) reported that 63% of their FAS sample suffered from one or more psychiatric disorders and the types of disorders shifted with age. For example, while hyperactivity and attention deficit were common both during preschool and school age, problems were not restricted to these core symptoms (Steinhausen, 1998). In the preschool period, eating disorders, enuresis, speech delay, and stereotyped habits (facial tics, nail biting, hair plucking) also occurred (Steinhausen et al. 1993, 1994, 1998; Streissguth, 1994). Later, during early school age, problems such as speech delay and stereotyped habits were even more common, and problems such as anxiety or sleep disorders emerged (Steinhausen et al., 1993, 1994,1998).

Steinhausen (1993, 1998) described hyperkinetic disorder as the predominant psychiatric syndrome of alcohol exposed children and social relationship problems as the

second most frequent problem. In their clinical studies, Steinhausen et al. (1998) showed that the prevalence of general psychopathology in children with FAS exceeded by far the rates of psychopathology in epidemiological studies. Further, the prevalence of general psychopathology was also higher than in controls matched for IQ and social background.

A more recent study of individuals with FAS/FAE by Streissguth et al. (1996) also reported high rates of severe mental health problems. To address the long-term outcome of gestational alcohol, Streissguth et al. (1996) define as primary disabilities those that reflect the FAS or ARND diagnosis. Secondary disabilities are those that an individual is not born with and could presumably be prevented through better understanding and appropriate intervention. Mental health problems were found to be the most prevalent secondary disability recorded by Streissguth et al. (1996). Ninety percent (426) of 473 subjects presented with one or more psychiatric conditions. The most frequent mental health problems for children and adolescents in Streissguth's group were attention deficit (61%), depression (50%), suicide threats (43%), and psychotic symptoms (29%).

In a 30 year follow-up of 28 of Lemoine's original patients from France (Lemoine and Lemoine, 1992), who had mild FAS in childhood, 2 had committed suicide as adults, and 5 others had attempted suicide (Streissguth, 1994). What is striking in the aftermath of these findings is that despite similarly high rates, research efforts have continued to focus more on attention deficits while depression, suicide, or more importantly it would seem, the precursors of depression and suicide in children affected by prenatal alcohol exposure have been largely ignored.

Many of the secondary disabilities associated with ARND create barriers to achieving a functional lifestyle later in life as indexed by: unstable employment, impaired relationships, lack of sound money management, poor care of possessions, and unproductive use of time. Social isolation, depression, suicide, lack of birth control, sexually transmitted diseases, and substance abuse have been identified as issues commonly faced by practitioners working with young adults with FAS/FAE (Connor and Streissguth, 1996; Smitherman, 1994).

The secondary disabilities observed in children and adolescents affected by prenatal alcohol exposure have now been shown to persist into adulthood and severely impede adaptive functioning. Moreover, adults with FAS or ARND (or FAE) suffer from high rates of psychiatric illness. A recent study by Famy, Streissguth, and Unis, (1998) of adults (19-51 years of age) with FAS (11) or FAE (14) showed that 23 (92%) received a DSM IV axis I diagnosis. The most common axis I diagnoses included major depressive episode, psychotic symptoms, and brief psychotic disorder. The most common axis II diagnoses were avoidant personality disorder, antisocial personality disorder, and dependent personality disorder. One subject each had paranoid, schizotypal, and borderline personality disorder. According to self-report, 18 (72%) subjects had already received some form of psychiatric treatment and 6 (24%) had required hospitalization in a psychiatric institution. Fifteen (60%) subjects met criteria for current or past alcohol or drug dependence. The authors claim this to be the first formal study of the psychiatric diagnoses of subjects with FAS/FAE. This work has provided strong evidence that children with alcohol exposure are at great risk for multiple debilitating psychiatric problems throughout life.

Despite the rapidly growing awareness about the relatively poor prognosis among individuals exposed to alcohol in utero, much less is known about the early manifestation of socioemotional disturbance in children with ARND. Since problems in socioemotional functioning may precede the onset of severe psychopathology, information of children's socioemotional functioning may facilitate early identification and lead to prevention/intervention strategies. According to Roebuck et al. (1999):

> Given the increased risk for emotional and social adjustment problems in alcohol-exposed children it is important to understand and document their behavioural and psychosocial profiles. This is especially true given that the effects are not seen just in childhood, but progress into adulthood where these problems are likely to continue to present challenges (p. 1071).

Furthermore, many practitioners in the field have recognized the lack of systematic research around prevention and intervention of secondary disabilities in children with ARND (Connor et al., 1996; IOM, 1996; Smitherman, 1994; Weiner and Morse, 1994). Indeed, the committee members appointed by the U.S. congress to study FAS/ARND through the Institute of Medicine (IOM, 1996) articulated the urgent need for further research in the field to assess the clinical expression and specificity of emotional, social, behavioural, and cognitive deficits of these syndromes across the lifespan.

Effective preventative and remedial methods will necessitate our gaining a more comprehensive and integrated understanding of the problems associated with prenatal alcohol exposure. Future research will need to identify highly specific information about the kinds of social relationship problems these children are having as well as their impact on emotional functioning. The interaction between impairments across domains such as cognitive deficits, self-regulatory and behavioural problems (i.e. attention), and

socioemotional functioning also needs to be better understood in order to make informed and targeted prevention and intervention efforts possible.

## Rationale and Purpose

The present study addresses a gap in previous research, first, by studying ARND rather than FAS, and second, by studying socioemotional functioning specifically in children with ARND, rather than cognitive/academic functioning. Having been identified first historically, FAS has been studied for a longer period of time than ARND. To date, FAS remains the most well known of the conditions associated with prenatal alcohol exposure. The fact that children with FAS have received much more attention than children with ARND also seems related to their being a more accessible population. Although the full-blown syndrome represents a relatively small proportion of children affected by prenatal alcohol exposure, FAS is more readily detected given the associated physical anomalies. In addition, there exists a much more explicit, pathognomonic, and well-established set of diagnostic criteria for FAS than for ARND, again making FAS much easier to identify and study. The American Academy of Pediatrics Committee on Substance Abuse and Committee on Children with Disabilities (2000) recently acknowledged that, "the lack of specificity and absence of definitive diagnostic criteria have made research and classification difficult" in ARND.

Similarly, to account for the disproportionate focus on cognitive versus socioemotional functioning in this field, it may also be the case that the cognitive domain represents a much more clear-cut and accessible area from a research perspective. In contrast, the area of socioemotional functioning, which historically has received less attention in terms of systematic scientific inquiry, continues to represent a more daunting

and subsequently neglected topic of research. At the same time however, there is still consensus among professionals that socioemotional problems are common and profound in this population and represent a major area of concern.

The purpose of this investigation is to identify the nature and severity of socioemotional disturbance among children with ARND. This paper summarizes efforts at identifying the characteristic socioemotional profile of ARND in relation to socioeconomic and demographic characteristics of our client population. To accomplish this, the following 3 questions were addressed:

1. Do children who have been diagnosed with ARND present as a group with a clinically significant and distinct profile of social and emotional problems?
2. Is there a significant relationship between the severity of socioemotional difficulties among children with ARND, and environmental factors such as SES and adoption history?
3. Do qualitative responses on the Child Behaviour Checklist (CBCL; Achenbach, 1991) indicate consistent and meaningful themes that are specific to this disorder?

## Hypothesis

Children with ARND will present with a distinct clinical profile of disturbed socioemotional functioning. Furthermore, the severity of their socioemotional disturbance will reflect familial and background factors.

## Chapter Two: Method

### Procedures

#### The Diagnostic Process

The present study is part of a larger ongoing project through the Motherisk Follow-up Program, at the Hospital for Sick Children in Toronto. The Motherisk Follow-up Program, has for the past 6 years been studying children prenatally exposed to alcohol. The children were mostly brought to our clinic by foster or adoptive parents for concerns about suspected alcohol exposure while a small proportion of children were brought by their biological parent(s) or relatives who claimed alcohol was abused by the mother during the pregnancy, and were concerned whether current learning and behavioural problems were due to this alcohol consumption.

Based on descriptions by parents attending ARND support groups, our previous clinical experience with these kinds of children, and descriptions in the literature of disabilities, we defined a cluster of specific ability deficits and assets that appeared to characterize these children. This process culminated in a diagnostic checklist comprised of 21 deficits and 6 assets. This procedure has been deployed previously by neuropsychologists diagnosing other populations of children, including children with nonverbal learning disabilities (Rourke, 1995). It is important to clarify that assets are personal strengths and refer to areas of functioning in which the child is better at in relation to other aspects of his or her own neuropsychological functioning. This does not necessarily mean that the child is performing above standardized norms or other samples of non-clinically referred children.

Each child was administered a battery of comprehensive neuropsychological tests which included standardized measures of intelligence, language, memory, attention, academic, visuospatial, visuomotor, and socioemotional functioning. (See Table 1).

This assessment took place over a full day period or two half-day periods depending on the child's endurance. Assessments for pre-school children (4-5 years of age) lasted on average 4.5 hours while assessments for school-age children lasted approximately 6.5 hours. In a small number of cases where a previous assessment used several of the same tests within a 1-2 year period, the results were incorporated into our diagnostic process rather than repeating the particular measures.

Following test administration and scoring of each child's results, the checklist of deficits and assets was completed in order to determine how well an individual child's neurobehavioural profile resembled our ARND profile. For each child the diagnostic checklist was completed independently by the examiner and the supervising clinical psychologist. Previous analyses revealed that the ratings of the examiner and psychologist were highly correlated (deficits: r= 0.93 strengths: r=0.67) (Greenbaum, Nulman, Rovet, and Koren, 2000). It must be noted that the psychologist's ratings are based on the information and test scores derived from the examiner's assessment of each child. The diagnostic checklist is presented in Table 2.

The criteria for assigning children to the ARND group was set at minimum of 60% of deficits (13 or more out of 21) and 50% of assets (3 or more out of 6). These cut-offs were based on our observations that many children with ARND do not present with the entire range of problems (or strengths) associated with the disorder, but rather a proportion of the features with varying degrees of severity. As such our sample included

a range whereby some children presented with 75%-95% of weaknesses while other profiles comprised of relatively fewer (i.e. 60%) weaknesses were also deemed consistent with ARND given the severity of the problems based on our clinical judgment (Greenbaum et al., 2000).

Each child was seen by the physician on our team who obtained growth measurements and evaluated facial features for dysmorphology. Dysmorphology was evaluated using a 3-point scale ranging from 1 (absence) to 3 (largely present). Demographic information was obtained from caregivers using questionnaires. Socioeconomic status (SES) was determined using the Hollingshead Four Factor Inventory (Hollingshead, 1975) which is based on the education and occupation levels of both parents. Where applicable, the SES score was based on the foster or adoptive family with whom the child was residing at the time of testing.

Psychological reports were written for each child summarizing the test results. Also indicated in each report was the extent to which the child in question met or did not meet our diagnostic criteria, and, subsequently, whether he or she received a diagnosis of ARND. All assessments conducted by the Motherisk Follow-up Program were reviewed and children who received a diagnosis of ARND (versus those who were not given a diagnosis) were selected for the study. The diagnosed children were then matched to control subjects on a best fit basis on age, gender, SES, with the exception of 2 children for whom controls could not be obtained (due to their relatively older ages). The final sample included a total of 33 matched pairs. The non-matched ARND subjects were included, however, in the qualitative portion of the study, which examined the ARND group only (n=35), not in comparison to the control group.

It should be noted that this diagnostic process is still considered experimental. However, to date there exists no better alternative to identify children with ARND, that is children affected by prenatal alcohol exposure who do not present with all of the physical characteristics to qualify for the full-blown syndrome (FAS). It must also be acknowledged that the present sample of children diagnosed with ARND includes children who were referred for diagnostic assessment based on the knowledge and/or suspicion of prenatal alcohol exposure combined with presenting learning and/or behavioural problems. Therefore, in order to study the behavioural (and socioemotional) problems associated with ARND, one must inevitably rely on samples of children who have been brought to professional attention at least in part due to the presence of behavioural (and socioemotional) problems. This represents a major confound for the study but one that is inherent in our present state of knowledge about the clinical problem. It is also one that we make some attempt to address in our results.

## Participants

For this study, a sample of convenience was used that includes the total number of diagnosed ARND cases seen by the Motherisk Follow-up Program at the Hospital For Sick Children in Toronto since 1996. Out of 61 children referred between 1996 and 1999, 35 children (19 boys) were diagnosed with ARND through the Motherisk Program. Referrals were made by biological, foster, or adoptive parents, or social agencies. The children ranged in age from 4 to 18 years of age. Because many children were not in the care of their biological parent(s) at the time of the assessment, specific details about the history of alcohol exposure (timing, amount, poly-substance use) were not always available. However, in most cases, heavy alcohol use by the mother during pregnancy

was highly suspected. While these referrals were made for a clinical service (diagnostic assessment), consent was obtained from parents/guardians permitting the clinical information to be used for research purposes.

Controls were selected from the databases of several preexisting studies in which they had previously served as control participants. Controls were individually matched to the ARND group on a best fit basis on age, gender, and socioeconomic status (SES). Pairs were matched within 6 months of each child with ARND, except for 11 children who were matched within 7-9 months of age. There were no differences among the matched pairs with respect to gender or SES.

Tests and Measures

Child Behaviour Checklist.

The Child Behaviour Checklist (CBCL; Achenbach, 1991) is the primary measure of socioemotional functioning in this study. It is designed to record in a standardized format children's competencies and problems as reported by their parents or parent surrogates. The CBCL is a structured rating scale for 4-18 year old children asking parents to rate their child's social and emotional problems. It consists of 118 behavioural and emotional problems that are rated using a "0" (not true), "1" (sometimes true), or "2" (very true) response set. All ratings are based on parent judgments at the present time or within the past 6 months. Where an older scoring version had been used, the completed CBCL forms were re-scored using the 1993 computerized norms to ensure comparability of all protocols in the study.

The development of the CBCL was prompted by the lack of satisfactory constructs and operational definitions for childhood disorders (Achenbach, 1991). The

ultimate goal of this task was the identification of, "reliable profile patterns that characterize clinically-referred boys and girls" (Achenbach and Edelbrock, 1980). The CBCL has evolved over multiple editions (1978, 1979, 1983, 1991, 1993). On this task, problems and competencies are grouped according to dimensions that qualitatively reflect a child's deviance from normative groups. In the standardization sample, the parents' ratings on the CBCL were factor analyzed separately for different age intervals (4-5, 6-11 and 12-18). These age groups were chosen to reflect of developmental changes in cognitive, biological, and psychosocial functioning as well as major transitions in schooling.

CBCL behaviour problem scales.

Task scoring is based on normative data provided by Achenbach and is used to convert raw scores to T-scores (mean=50, SD=10). The task is computer-scored to yield a Total Problems Score, two broad-band factor scores Internalizing and Externalizing, and eight narrow-band scales: Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behaviour, and Aggressive Behaviour. A Sex Problems scale is also scored for parents' ratings of children aged 4-11. The narrow-band syndromes can be viewed as subtypes of the broad-band syndromes (Achenbach and Edelbrock, 1980). The problem scales are scored negatively with higher T-scores reflecting more problems. A T-score of 63 or more represents the clinical range for the problem scales.

Internalizing symptoms are those associated with overcontrolled tendencies whereas externalizing symptoms are those associated with conduct problems or undercontrolled behaviours. Among the eight narrow band scales Withdrawn, Somatic

Complaints, and Anxious/Depressed belong to the Internalizing category, while Delinquent and Aggressive Behaviour belong to the Externalizing category. Although Social Problems, Thought Problems, Attention Problems, and Sex Problems do not fall under either the Internalizing or Externalizing broad-band categories, these scales do load into the Total Problems score.

CBCL social problem scales.

Also provided by the CBCL is an overall social competence rating comprised of three scales assessing school performance, involvement in activities, and social relationships. In contrast to the problem scales, the competence scales are scored positively, so that a higher score reflects better competence. A T-score of 40 or less represents the clinical range for the competence scales.

CBCL diagnostic classifications.

In addition to scale scores, the CBCL provides a diagnostic classification that assesses the degree of similarity between the individual child's profile and profiles of patients with differential psychopathological diagnoses. Children are assigned to diagnostic classifications by computerized scoring and the algorithm of Edelbrock and Achenbach (1983). This procedure derives from their studies of consistent profile types among large groups of behaviour-disturbed children using hierarchical cluster analysis techniques. Computerized scoring of each child's protocol provides six or seven (depending on the child's age and sex) intraclass correlation coefficients (ICC), which represent the degree of correspondence between that child's profile and that of children in each of the profile subtypes determined by clustering techniques. According to Edelbrock and Achenbach's algorithm, any child with a Total Behaviour Problem score between 25

and 100 and a positive ICC above 0.20 is assigned to the typology of the highest ICC. For the purposes of the present study, a cut-off point of 0.25 was used to discriminate between significant and non-significant ICC profiles. This information was entered into the database as a discrete variable, that is either yes, the child had at least one positive ICC profile typology or no, he did not. Also entered into the database were variables indicating which of the seven ICC classifications were applicable (>0.25) to each child (Withdrawn, Somatic, Social, Delinquent-Aggressive, Withdrawn-Anxiety-Depression-Aggressive, Social-Attention, and Delinquent).

CBCL psychometric properties.

Adequate reliability and validity coefficients are reported by Achenbach (1991). For instance, inter-interviewer and test-retest reliabilities of CBCL item, scale, competence, and problem scores were supported by strong intra-class correlations. Content validity for the CBCL is demonstrated by the finding that nearly all CBCL items discriminated significantly between demographically matched referred and non-referred children. As well, construct validity is supported by numerous correlates of CBCL scales, including significant associations with analogous scales on the Conners Parent Questionnaire and the Quay-Peterson Revised Behaviour Problem Checklist. The CBCL's quantitative scale scores discriminated between referred and non-referred children after demographic effects were partialled out providing strong evidence for criterion-related validity. All competence scales were scored higher and all problem scales were scored lower for non-referred than referred children at $p<.01$. Clinical cut-points on the scale scores were also shown to discriminate significantly between demographically matched referred and non-referred children.

Qualitative Data

For the ARND group only, qualitative responses to items requesting parents/caregivers to describe the child's greatest strengths and their greatest concerns about their child were examined systematically. The written responses from all the CBCL forms were transcribed collectively into separate lists of greatest strengths and greatest concerns. The responses were then organized into meaningful clusters or overlapping themes totaling 6 for strengths and 6 for concerns. A tally was taken manually of the total number of responses fitting into each of the categories of strengths and concerns. This tally was then converted into a percentage of the children in the ARND group who were identified/reported by their parent/caregiver as having each of the 12 attributes.

Composite Scores

Ten environmental factors were selected from a standard case history form completed by the parent or caregiver for each child in the ARND group. It should be noted that this list of environmental factors was not intended to be exhaustive; selection of these ten factors was based on the literature pertaining to risk in children, which indicated that these were the most salient among many environmental influences in the lives of the children (Jenkins and Keating, 1999); Rutter et al., 1997; Sameroff and Fiese, 2000; Sameroff and Seifer, 1982). Rather than including this information as 10 separate variables in the database (given the already high number of variables in the study), the information was divided into categories resulting in 2 composite scores. Items comprising the Home and Social Background composite score include the following information: number of homes lived in prior to testing, amount of time spent in current (at time of testing) home, number of other children living at home, history of abuse and/or neglect,

and socioeconomic status. Items comprising the Educational and Treatment History composite score include: repeating a grade, receiving treatment and/or modified programming in school, receiving treatment outside of school, and medication related to attention and/or other behavioural/emotional issues. It was anticipated that higher composite scores would be associated with greater severity of socioemotional problems. Table 3 outlines the scoring breakdown for the composite scores.

Data Analysis

t-tests were used to compare the ARND and control group on all continuous variables including demographic characteristics and broad and narrow-band scales on the CBCL. t-tests were also used to compare the number of children in the ARND and control group presenting with scores in the clinical range (>63) on the CBCL broad band scales (Total, Internalizing, and Externalizing Problems). For the broad and narrow-band scales, 10 variables were selected a priori based on clinical judgment: Total Behaviour Problem, Externalizing and Internalizing problems, Attention, Sexual, Social, and Thought Problems, Withdrawal, and Aggressive and Delinquent behaviour. For these 10 a priori variables, a Bonferroni adjustment by a factor of 10 was applied to the p-values obtained for each test. Therefore, p-values were considered significant if they were less than or equal to 0.05/10 ($p<.005$). For the remaining CBCL narrow-band and competence scales (n=6), a Bonferroni adjustment by a factor of 120 was applied to the p-values obtained for each test (to adjust for the high number of variables in the study which resulted in multiple analyses). All clinical elevations for narrow-band scales were rank-ordered from highest to lowest. The highest mean T scores were taken to represent

problem areas of greatest severity in the ARND group and the lower scores, while still clinically significant (T>63) were deemed less severe.

t tests were also used in post hoc analyses to determine whether the children in the ARND group represented a homogeneous sample given that there was more certainty in some cases than in others about the history of prenatal alcohol exposure. In a number of cases, despite the absence of physical anomalies, there was a high degree of certainty that the child had been heavily exposed to alcohol in utero. Exposure history was considered to be confirmed with certainty when one or more of the following was true: the child was still in the care of a biological parent who could report first-hand on the exposure history; the reason for the child's placement into care was due specifically to the maternal alcohol abuse; the child suffered alcohol withdrawal at birth. In contrast, among the other cases, despite strong suspicions there was not the same kind of conclusive evidence to substantiate the claims about prenatal alcohol exposure. Therefore, the sub-group analyses compared the children in the ARND group on select variables on the basis of having a confirmed versus unconfirmed exposure history. The comparisons were made for SES, age at testing, and Total, Externalizing, and Internalizing Problem Scores on the CBCL.

In an effort to gain a greater understanding of the socioemotional problems among children with ARND, exploratory analyses were conducted on the 118 individual items of the CBCL. Given the matched pair design, McNemar's test was used to compare ARND versus controls on these items. McNemar's test requires cell sizes greater than or equal to 5. Due to low frequencies in some cells, it was deemed both useful and clinically sound to collapse categories as this would achieve increased cell frequencies. Originally, these

items were rated on a 3-point scale (0=Not True, 1=Somewhat True, 2=Very True). The "Somewhat True" and "Very True" categories were combined into a single True category, thus creating a 1=True versus 0=Not True dichotomy.

Multiple regression analyses were used to determine the relationship (if any) between the presence/severity of socioemotional problems in children with ARND and external factors. The ultimate goal was to determine the extent to which these other (i.e. environmental) factors influenced/accounted for poorer socioemotional outcome in this sample of children with ARND. The Total Problems score on the CBCL constituted the primary outcome variable. Since this score is continuous, multiple linear regression was used for modeling. Predictors considered for this model were: child's age at testing (subsequently the age at which the child was diagnosed with ARND), 2 composite scores (home/social background and education/treatment history), and overall dysmorphology score. Although it is now recognized that children exposed prenatally to alcohol manifest problems regardless of the presence or degree of physical abnormality, we chose to provide further confirmation of this in the present study by using overall dysmorphology as a predictor variable with the expectation that it would not significantly influence outcome on the CBCL. The secondary outcome variable was ICC. Given that it is a dichotomous variable, multiple logistic regression was used for this modelling. The same predictors were considered for this model.

Post hoc analyses were conducted using 3 of the 7 specific ICC profile types (Social, Delinquent, and Delinquent/Aggressive) as outcome variables with the same predictors used previously in the regression modelling. These ICC profile types were

selected because they were found to be the most frequently represented profile types within the ARND subject group.

Two covariates were added on a post hoc basis to the multiple regression analyses. After the first set of analyses revealed no significant correlations between Home/Social Background and the outcome variables (Total Problems and ICC), SES and history of abuse/neglect were selected from among the 5 variables comprising the composite score Home/Social Background given that these two variables have been frequently found to be associated with increased symptomatology in children (Rutter, 1991). It was hypothesized that the original method of combining a number of environmental variables into the one Home/Social Background composite score (ultimately to limit the total number of variables in the study), may not have been sensitive enough to detect the influence of individual variables on the socioemotional outcome of the children over and above the effects of prenatal alcohol exposure.

Percentages were tabulated manually for the qualitative data in order to describe the proportion of children in the ARND sample presenting with each of the 12 defining attributes as reported by parents/caregivers (6 into strengths and 6 into concerns).

## Chapter Three: Results

### Demographic Information

Results from the t tests confirmed that the matched-sample design employed in the study ensured that there were no significant differences between the ARND and control subjects with respect to demographic characteristics including age, gender, and SES. Table 4 provides a summary of the demographic information. The quantitative part of the study included 33 pairs of 19 males and 14 females ranging in age from 4 to 14 years. The qualitative part of the study involved only the ARND subjects and included the same 33 children from the matched sample plus two ARND subjects for whom matches were not available (n=35). Results from the sub-group analyses revealed no differences in SES or age at testing between ARND subjects with confirmed (n=17) versus unconfirmed exposure histories (n=18).

### Descriptive Statistics

All of the 10 scales selected a priori, remained significantly different between the ARND and control group after correcting for multiple testing (Bonferroni). In contrast to controls, children in the ARND group presented with significantly elevated scores on scales of Total Problems, Externalizing and Internalizing Problems, Attention, Sexual, Social, and Thought Problems, Withdrawal, and Aggressive and Delinquent behaviour (adjusted p=.001). The majority of scores for the ARND group were within the clinically significant range. When ranked according to severity by the degree of T-score elevation, the following scales emerged in order as the most significantly problematic in the ARND group: Attention Problems, Social Problems, Total Problems, Delinquent and Aggressive behaviour, Thought Problems, and Externalizing Problems. This is in contrast to the

control group for whom none were found to be clinically significant. Table 5 and Figure 1 show mean differences between children in the ARND group and controls on the CBCL broad and narrow band scales. After statistical adjustment to correct for the number of variables in the analysis, the following 6 scales did not remain significant: Withdrawal, Anxiety/Depression, Somatic Complaints, Social Competence, School Competence, and Activities. However, the mean T-scores for children in the ARND group were within the clinical range (T<40) for the Social and School Competence scales.

Significant differences were indicated between the ARND and control groups with respect to the number of subjects obtaining clinically elevated scores (T>63) on the broad-band scales of the CBCL. Compared to controls there were significantly more children in the ARND group who obtained clinically elevated scores on the Total Problems, Internalizing, and Externalizing Problems scales ($p<.0001$). Table 6 shows the differences between children in the ARND group and controls with respect to clinical elevations on the CBCL broad-band scales.

## McNemar's Test

McNemar's test indicated a significant difference between the children in the ARND group and controls ($p=0.001$). With respect to the ICC scores, 61% of children in the ARND group had at least one ICC above the 0.25 cut-off compared to 12% in the control group. Examination of the seven specific profile types revealed that those relating to Social problems, Delinquency, and Delinquency/Aggression were most frequently represented in the ARND group. The profile type relating to Withdrawal was the most frequently represented profile in the control group (n=2). Table 7 summarizes the frequencies of the 7 ICC profiles for both groups.

Item Analysis

McNemar's test of the individual CBCL items (which collectively comprise the broad- and narrow-band scales) were conducted on an exploratory basis in an effort to understand the specific kinds of socioemotional problems characterizing ARND and establish if a consistent socioemotional profile exists. Without correcting for multiple testing (Bonferroni), the McNemar's Test revealed significant differences between children in the ARND group and controls on 62 (52.5%) of the 118 individual items of the CBCL. For these 62 items, the following 7 items were endorsed for more than 80% (ranging up to 90-97%) of children in the ARND group:

- acts too young for his/her age
- argues a lot
- can't concentrate/can't pay attention for long
- can't sit still/restless or hyperactive
- disobedient at home
- impulsive or acts without thinking
- showing off/clowning

The following items also discriminated between the 2 groups (unadjusted p=0.001) based on the number of ARND versus controls who had endorsements on:

- can't concentrate/can't pay attention for long
- impulsive or acts without thinking
- doesn't seem to feel guilty after misbehaving
- lying or cheating
- doesn't get along with others

- confused
- sudden changes in mood or feelings
- disobedient at school

Table 8 and 9 summarize the significant differences between ARND and controls on the 118 individual items of the CBCL.

## Multiple Regression Analyses

Bivariate correlations revealed that age at testing, gender, and the composite variable of Educational/Treatment history were significantly correlated with severity of outcome as reflected by the primary outcome variable, CBCL Total Problems. The multivariate linear analyses revealed that while age at testing was no longer significant, there was a significant interaction between gender and Educational/Treatment history in the prediction of Total Problems on the CBCL (p=.04). This interaction shows that the gender difference is marked at low levels of intervention and there is no difference in total problems between boys and girls at high levels of intervention. As shown in Figure 2, this interaction reflected the tendency for boys to have high Total Behaviour Problems scores regardless of Educational/Treatment History, whereas only girls with high Educational/Treatment scale scores had elevated Total Behaviour Problems scores. For males the slope for Education/Treatment in the prediction of Total Problems was 0 .46. For females, the slope for Education/Treatment in the prediction of Total Problems was 1.46. Figure 2 shows the interaction effect between gender, Education/Treatment, and outcome on the Total Problems scale of the CBCL.

Multivariate logistic analysis was carried out to examine the predictors of a positive ICC score. This analysis indicated that age at testing (odds ratio =1.41, p=.02) and gender

(odds ratio=0.12, p=.02) were significantly correlated with positive ICC. For every increase of one year in age (at the time of testing, and subsequently the time of ARND diagnosis), there was a 40% higher chance/risk of having a positive ICC profile. Also, males with ARND were 8 times more likely than females to have a positive ICC profile. Post hoc analyses of the 3 specific ICC profiles, Social, Delinquent, and Delinquent/Aggressive showed a similar pattern as that resulting from the logistic regression for the single ICC variable. For instance, age was correlated with the ICC profile Social Problems (.05) and with the profile Delinquent/Aggressive (.04). No other significant correlations were indicated.

Neither the composite score pertaining to home and social background factors (Home/Soc) nor overall dysmorphology were significantly correlated with the presence or severity of socioemotional problems as reflected by the primary and secondary outcome variables, CBCL Total Problems and ICC. Post hoc analyses also indicated that neither SES nor history of abuse/neglect were correlated with outcome in this sample.

## Qualitative Information

One of the goals of this study was to determine whether the qualitative responses on the CBCL were able to generate consistent and meaningful themes that are characteristic of ARND. Manual analysis of the qualitative data revealed 12 meaningful themes (6 for the item "greatest concerns" and 6 for "best things"). Themes applying to the highest proportion of children in the ARND group were deemed to be most characteristic of the disorder. With respect to "greatest concerns", cognitive problems described 62% of the children in the ARND group, while concerns about adaptive life functioning/the child's future and behavioural concerns/self-regulatory problems were

described in 52% of the cases. Socioemotional problems were concerning to parents/caregivers in 41% of the children.

With respect to "best things" identified by parents about their children, 64% of the children in the ARND group were described as having a loving nature, 49% were described as being helpful and trying hard (positive approach), 36% as having a zest for life and sense of humor, and 33% as having a hobby. Tables 10 and 11 summarize the proportion of children in the ARND group who are represented in each of the 12 characterizing categories.

## Chapter Four: Discussion

The initial hypothesis of this study proposed that children with ARND would present with a distinct clinical profile of disturbed socioemotional functioning. The second hypothesis proposed that the severity of their socioemotional disturbance would reflect familial and background factors. Present findings supported the first but not the second hypothesis. Results revealed that the children with ARND presented with significantly higher levels (many of which were also clinically significant), on a wide range of problems as indicated on the broad and narrow- band scales of the CBCL. In keeping with this, children in the ARND group were much more likely than control subjects to present with a positive ICC profile typology, particularly those profiles associated with social problems, delinquency, and aggression. Exploratory analyses of the individual items on the CBCL identified 13 specific problems that characterize a majority of the children in the ARND sample. Finally, systematic scrutiny of the qualitative information gleaned from the CBCL resulted in themes that are highly consistent with the results from the narrow-band scales, ICC profiles, and individual CBCL items that differentiated the ARND and control groups.

While the overall aim of the present study was to expand upon what is currently understood about socioemotional functioning in children with ARND, these findings have also served to confirm findings from previous research in the field. At the most general level of analysis, the present findings are consistent with previous studies showing that as a group, children affected by prenatal alcohol exposure present with significant impairments in socioemotional functioning (Roebuck et al., 1999; Steinhausen et al., 1998; Thomas et al., 1998), and furthermore, that prenatal alcohol exposure

independent of the physical features required for a diagnosis of FAS leads to psychosocial impairments (Roebuck et al., 1999).

More specifically, as was evident in Steinhausen et al.'s (1998) longitudinal sample of children with FAS, peaks in the areas of attention deficit problems and social relation problems also characterized the CBCL profiles in the children with ARND in the current sample. These findings were corroborated by teacher ratings in the Steinhausen et al. study. The range of mean T-scores ($\underline{M}$=50, $\underline{SD}$=10) on the CBCL narrow band scales was also fairly consistent with Steinhausen et al.'s results (1993) with mean T-scores ranging between 52-68 as compared to 58-75 in the present sample.

While the study by Roebuck et al., (1999) comparing 32 alcohol exposed children to well-matched controls on the Personality Inventory for Children (PIC) showed the Hyperactivity Scale was the least elevated of the scales, 19 of the 32 children (59.4%) in the alcohol group had T-scores on the Hyperactivity scale above the cut off score reported to discriminate ADHD from other groups of children. Therefore, while their overall PIC profiles differed from ADHD groups, the authors did agree that, "a large portion of the children in the alcohol group may exhibit behaviours typically associated with ADHD" which is consistent with the present findings.

The present study is highly comparable to Roebuck et al.'s (1999) in that their sample was also drawn from a larger ongoing study, the primary purpose of which was to assess neuropsychological impairments in alcohol-exposed children. Their alcohol group had significantly higher T-scores than controls on all substantive scales of the PIC (Roebuck et al., 1999), with clinical elevations on 5 of these scales: Achievement, Intellectual Screening, Development, Delinquency, and Psychosis. The authors concluded

that the highest elevations on the Delinquency and Psychosis scales, "suggest emotional lability, poorly developed social skills, social withdrawal, and history of developmental delay". These findings definitely correspond with our findings using the CBCL, which is a more widely used test. We found the children with ARND differed from controls on Social Problems, Withdrawal, Thought Problems, Aggression, and Delinquency CBCL behaviour problems scales. They also scored lower on School and Social Competency scales. As well, among the 7 ICC profiles, those relating to Social problems, Delinquency, and Delinquency/Aggression were the most frequently obtained by children in our ARND group.

The Sex Problems narrow band scale appears to be a unique and important feature of the CBCL. That this scale was significantly higher in the ARND versus control group would seem to constitute a warning sign. It has become general knowledge among professionals in the field that children with ARND may be at higher risk both as potential victims and perpetrators of sexual exploitation/abuse. While this topic has been discussed in the lay literature (Berg et al., 1995), it has not yet been addressed in a more formal, systematic study. Berg et al. (1995) explained that, "Inappropriate sexual behaviour can become offender behaviour and younger children may be at risk. As well, [children with ARND] may be at a very real and substantial risk of being victimized themselves." The present discussion may be one of the first to highlight the issue from an empirically-based stand-point and suggests that further and more specific inquiry in this area is warranted. The exploratory adjusted chi square analysis (McNemar's Test) of the 118 individual CBCL items was undertaken in an effort to glean more information about the specific kinds of problem behaviours contributing to the socioemotional difficulties in children

with ARND. The results expand what has previously been documented about the specific nature of negative behaviours characterizing children with ARND. The following items consistently characterized a majority of the children in the ARND group: acts too young for his/her age, argues a lot, can't concentrate/can't pay attention for long, can't sit still/restless or hyperactive, impulsive or acts without thinking, showing off/clowning, doesn't seem to feel guilty after misbehaving, lying or cheating, doesn't get along with other kids, confused, sudden changes in mood or feelings, and disobedient at home and school. With replication, the specificity of these findings may be useful in helping to direct both diagnostic and rehabilitative efforts with these children.

The results of the multiple regression analyses indicated that at low levels of intervention (i.e. through modified education programming, treatment outside of school, and/or with medication), males show higher levels of problems than females. However, at high levels of intervention, males and females have similar rates of Total Problems. Children who had received more in the way of intervention were also found to have higher Total Problems scores on the CBCL. Males were 8 times more likely than females in the ARND group to have a positive ICC profile suggesting that male sex may be a risk factor for poor sociobehavioural outcome in children with ARND. Among the children in the ARND group, for every increase of one year in age (at the time of testing, and subsequently the time of ARND diagnosis), there was a 40% higher chance/risk of having a positive ICC profile. This shows the dramatic increase in behavioural problems with age in this population. Factors relating to home and social background (Hom/Soc) were not correlated with outcome.

One might have expected that children receiving more intervention would be better off (i.e. have less Total Problems) than children receiving less intervention. However, the opposite was true for the children in this sample. It is possible that among children in the ARND group, those who received more intervention actually had more (Total) problems to begin with. Furthermore, their higher number of Total Problems may also have meant that these children were harder to treat and subsequently required and received more intervention than the children with fewer Total Problems.

The finding that males were 8 times more likely than females to have a positive ICC profile, agrees with other scientific observations showing that in this population, the male sex is associated with increased psychosocial vulnerability (Steinhausen et al., 1982,1984,1994). At the same time, however, Steinhausen et al. (1994) found that despite the fact that boys obtained higher Total Problems scores than girls, he claims, "there is no significant differentiation between the 2 sexes with regard to the various psychiatric syndromes". In the present sample, males and females did not differ with regard to their Total Problems score. However, the finding that males in this sample were more likely than females to present with positive ICC profiles, means essentially that they are more likely to present with discernible clusters of characteristics that are consistent with other populations of disturbed children, in contrast to females who are less likely to demonstrate such common profiles.

Older children in the ARND group were more likely than younger children to have a positive ICC profile. More specifically, for every increase of one year in age (at the time of testing, and subsequently the time of ARND diagnosis), there was a 40% higher chance/risk of having a positive ICC profile. This finding is in keeping with the

findings of Thomas et al. (1998) who showed that social skills deficits in children with FAS become more pronounced over time such that older children, adolescents and adults with FAS may be more socially impaired than young children with FAS. They suggest that this may be due, at least in part, to the fact that it is often difficult to discern social handicaps in young children with FAS as they tend to be talkative, affectionate, and outgoing, and furthermore, that compared to young children, adolescents and adults face a higher standard for acceptable social behaviour.

It is also possible that the older children in our ARND sample were essentially worse off, with respect to their socioemotional problems, than their younger counter-parts because they had received a diagnosis later in life (in this study their older age at testing also necessarily reflected their older age at diagnosis). This is supported by the longitudinal work of Streissguth et al. (1996) which found that the best prognostic factor among individuals affected by prenatal alcohol exposure is early diagnosis (before age 6). In a Canadian study identifying FAS in the juvenile criminal justice system, Fast, Conry, and Loock (1999) found that, only 3 of 67 youths had been diagnosed with FAS/FAE before the study. The average age of this sample was 14.8 years. While this study supported the original contention that this group is disproportionately represented in the juvenile justice system, it also highlighted the fact that many children with FAS/ARND are either not being diagnosed early enough or not at all. These authors noted that, "by adolescence the opportunity for successful preventative interventions has diminished". At the same time, however, it also possible that the finding that older children were more likely to have a positive ICC profile may simply reflect a measurement issue, to the extent that problems may cluster with one another more for older than for younger

children. In this way one would expect to find a stronger relationship between social problems and hyperactivity and aggression with age. This may be understood if we consider that various behavioural problems (i.e. externalizing) are more similar to the behaviours of many young children and as such tend to be tolerated more among younger children. In contrast, the same kinds of problem behaviours in older children tend to differ considerably from what is typical. Their problematic behaviours may be more aversive and subsequently less well tolerated among older than younger peers.

The second hypothesis of this study proposed that the severity of socioemotional disturbance in children with ARND would reflect familial and background factors. The fact that social background factors such as SES and adoption history were not correlated with severity of socioemotional outcome is consistent with Steinhausen et al.'s (1982) finding that SES and social environment had no effect on the psychiatric status of the children among children with FAS. This finding is somewhat promising diagnostically, in that it suggests that the effects of prenatal alcohol exposure without physical dysmorphology are just that, the effects of alcohol toxicity rather than the effects of negative environmental factors. Alternatively, it is also possible that the methods employed presently which combined a number of environmental variables into a single composite score, may not have been sensitive enough to detect the influence of individual variables on the socioemotional outcome of the children over and above the effects of prenatal alcohol exposure. Although neither of individual background variables (SES and history of abuse/neglect) correlated with socioemotional outcome in this sample, this may also have reflected a limitation in the sensitivity of the scales used for these variables.

Close and systematic scrutiny of the qualitative information from the CBCL has helped confirm and extend what was found quantitatively. The themes that emerged from the majority of parent/caregiver responses to the question, "What concerns you most about your child?", included attention/impulsivity, cognitive problems, socioemotional functioning, behavioural /self-regulatory problems, and delinquency, all of which are highly consistent with the results from the narrow-band scales, ICC profiles, and individual CBCL items that differentiated the ARND and control groups. Also identified as a major theme was the concern about poor adaptive functioning and the children's future. This represents an important issue that may not be readily raised or recognized within the quantitative portion of CBCL and similar measures. Our attention might also be drawn to the fact that what represents one of the greatest concerns to caregivers of children with ARND, is a problem not readily addressed, particularly within the context of the education system which tends to be the focus of remedial efforts in children with FAS/ARND.

It is typically assumed that children with Alcohol Related Neurodevelopmental Disorder are better off than children with Fetal Alcohol Syndrome, in part because as a group they tend to present with a higher level of intellectual functioning. However, in light of recent research findings, it is becoming harder to deny the fact that prenatal alcohol exposure children, regardless of their IQ display many functional impairments, even though they lack the physical characteristics required for a diagnosis of FAS (Roebuck et al., 1999; Streissguth, et al., 1996; Thomas et al., 1998). The present finding that many parents/caregivers were most concerned about adaptive life skills in their children with ARND is in keeping with results from Streissguth et al.'s (1996)

longitudinal study, which indicated that despite the differences in IQ between FAS and ARND, adaptive scores were equally low for both subgroups. In the present study, some of the specific concerns described by parents about their child with ARND included: having poor judgment, no sense of danger, being uninhibited with strangers, and easily influenced by others. Given this range of problems, it does not seem too far a stretch to make a connection between these problems in adaptive life skills and Conry et al.'s work (1999) showing that a disproportionately large number of youth and adults with FAS/FAE are coming into conflict with the legal system. This highlights the need for professionals working with these families to take their problems in adaptive life skills very seriously. Additionally, it suggests that remedial efforts need to be comprehensive and move beyond the context of the classroom supporting both the home and family system.

Positive attributes or areas of strength are not the first aspects researchers tend to concern themselves with in their work with disturbed populations. And yet, this information yields much in the way of potential clinical value. In this respect, the results from the qualitative portion of the CBCL study have helped to extend what has been documented previously in the scientific literature and described anecdotally by parents. The following themes were identified as the "best things" about this sample of children with ARND: their loving nature, zest for life and sense of humour, hobbies including athleticism and creativity, positive approach (willingness to help others, trying hard), outgoing nature, and specific academic or cognitive traits. With replication, these results should offer important direction to professionals in their work with these children,

specifically in terms of creating effective programs that utilize and build upon these positive attributes.

## Methodological Limitations

To date, research efforts investigating the effects of prenatal exposure to alcohol have been limited by a number of factors, the present study being no exception. For instance, few studies have been able to include specific measures of both dose and timing of alcohol exposure during pregnancy. The main measure for the majority of studies consists simply of knowledge that clients were in fact born to mothers who abused alcohol. In some cases, among adopted samples for example, even this may not be certain. In addition to the lack of specificity around dose and timing of alcohol exposure, there is often even less specific information available around polysubstance exposure. Women who abuse alcohol very commonly also abuse other illicit drugs, especially cocaine and cigarettes. Cocaine in combination with alcohol produces cocaethylene, an exceptionally active neurotoxic substance. High rates of nicotine abuse alone have been correlated positively with reduced birth weight, and retarded intra uterine growth (Persson, Grennert, and Gennser, 1978; Rantakallio, 1979; Steinhausen, Nestler, and Sphor,1982) while cigarette smoking combined with alcohol is known to increase the risk for low birth weight, microcephaly, and hearing difficulties (Nulman et al., 1998). As well, other factors such as amount/quality of prenatal care and maternal nutrition are scarcely considered in the research. Such information is often not easily accessed, for instance when a child is no longer in the care of his or her biological parents. This is true for many of the foster and adopted children in the present study. As such the extent to

which these factors contribute to the outcome of children prenatally exposed to alcohol remains unknown and represents a serious confound.

Another factor that has been largely overlooked in the extant literature pertains to the likely occurrence of co-morbidity with respect to mothers' (and/or fathers') substance abuse and psychopathology. Many children born to substance abusing parents may also be genetically predisposed to develop psychopathology quite separately from that which has been considered to be secondary to the effects of prenatal alcohol exposure. The fact that a majority of alcohol-exposed children do not remain in the care of their biological parents, makes this information very difficult to obtain. Nonetheless, the paucity of information in this regard would seem to constitute another major confound within the existing literature.

Despite the recent establishment of relatively uniform criteria to guide professionals in the field, both clinical and research methods have been subject to temporal changes in terminology, interpretation and referral patterns. As mentioned previously, the lack of specificity and absence of definitive diagnostic criteria for ARND have made research and classification very difficult. Comparisons of findings have been limited to some extent by differences in the methods/measures used, both diagnostically and for research purposes.

For obvious reasons, none of the studies involving children exposed to alcohol in utero have involved random assignment. That is, studies have not involved random assignment of subjects to the alcohol-exposed versus non-exposed conditions prior to or during pregnancy, while holding all other variables constant. Needless to say, circumstances beyond the control of researchers continue to prevent this from happening.

As such, issues of covariance (e.g. maternal nutrition, prenatal care during pregnancy, quality of home environment) are difficult to account for in studies such as this one. As well, in many studies such as this one, researchers have not been blind to group membership; in some instances, the professionals responsible for the original diagnosis of FAS/ARND were actively involved in research procedures with these same individuals. With respect to the statistical analyses employed in ARND/FAS research to date, there is no real possibility of assessing causation and as such, the interpretations that can be made are merely correlational in nature.

A major limitation of the present study also pertains to the use of a normal control group. While comparing alcohol-exposed children to age and gender matched controls may be informative, this does not provide information about how alcohol exposed children differ behaviourally from other developmentally delayed groups (Roebuck et al. 1999). Until very recently, children with FAS/ARND had rarely been compared to other clinically diagnosed groups, especially with respect to socioemotional functioning. Instead, most studies measured intelligence, motor coordination and attention span in alcohol-exposed children and compared results only with societal norms (Weiner et al., 1994). Other efforts have used inappropriate comparison groups such as children with Down Syndrome or children born to mothers with Epilepsy (Mattson and Riley, 1999; Steinhausen, Nestler, and Huth,1982). Despite the difficulties inherent to the task, it is important that we strive to determine how the problems associated with prenatal alcohol exposure differ from those found in other clinically similar populations. In a previous study, El-Guelbaly and Offard (1977) had difficulty differentiating between the adjustment of children with schizophrenia, those of depressive, and those of alcoholic

parents. As well, Greenbaum et al. (2000) were unable to discriminate statistically between children with ARND and other clinically-referred children on measures of attentional, behavioural, and socioemotional problems. Results from these studies suggest that a more fine-grained approach is needed in our efforts to understand more completely the nature of the problems relating to ARND. It is worth noting that clinicians have had limited success in treating the effects of prenatal alcohol exposure using methods derived from work with other clinical populations. Therefore, it is important that future research studies compare children with ARND to other similar clinical populations in order to identify the problems that are unique to prenatal alcohol exposure. This information will direct efforts to develop interventions that are specifically targeted to meet the needs of this currently under-serviced population.

Further limitations of this study are the small sample size and reliance on parental report, which may be subject to some bias. Although the clinically-referred sample may limit the generalizability of the findings given that these cases may represent extremes with respect to symptomatology, realistically, no alternative exists for identifying children with ARND in the general population. Despite these limitations, the results of this study represent a substantial contribution within the field of ARND research.

Replications of these findings are needed before firm conclusions can be made. Efforts to replicate this study would likely benefit from a larger sample size to allow for a more detailed exploration of the relationship between environmental factors like adoption and abuse history and socioemotional outcome in ARND. Such efforts would also be improved by including multiple informants to ensure reliability (i.e. teachers, self-report). A larger-scale qualitative study specifically, comparing children with ARND to other

clinically referred children and normal controls, might also be able to elaborate on the themes derived in the current study with respect to areas of greatest concern and positive attributes.

## Future Directions

It is not yet clear how the timing of alcohol exposure, prenatal care, maternal health, genetic susceptibility, and concomitant exposures contribute to FAS/ARND. As well, little is known about paternal alcohol abuse and its impact both in terms of influencing maternal alcohol consumption and/or biological repercussions on the developing fetus. Future research will need to address these issues utilizing consistent, rigorous, and systematic methods of inquiry.

It has been observed (IOM, 1996) that children with ARND have rarely been compared to other clinically diagnosed groups to identify factors specific to those who have been exposed to alcohol. It may indeed be possible to design targeted prevention efforts that will help to avoid the more negative outcomes, if the specific problems of children with FAS/ARND are identified. At the same time, many practitioners in the field have expressed concern about the curious lack of enthusiasm for targeted efforts directed at the prevention of secondary disabilities in children with ARND (Connor et al., 1996; Smitherman, 1994; IOM, 1996; Weiner and Morse, 1994; (IOM, 1996). The view that intervention may not be applicable to children affected by alcohol is inconsistent with the approach taken toward other groups of high risk and disabled children.

Greatly needed are efforts directed at early identification, prevention, and treatment that include support for children with FAS/ARND as well as for biological, foster, and adoptive family members. It is notable that, although many health professionals

recognize that fetal alcohol exposure is a grave problem (IOM, 1996; Jones et al., 1998; Sampson et al., 1997), there are few, if any, programs in Canada for specifically treating children with FAS or ARND. Furthermore, all efforts to date to prevent secondary disability in these children have been deemed "insufficient and inadequate" (IOM, 1996). There is also no available source of systematically compiled information describing the number of people with FAS/ARND receiving services or the kinds of services received by individuals with FAS or other alcohol related deficits (IOM, 1996).

Despite the high prevalence and severity of ARND, there currently exists minimal community support and extremely limited resources to meet the specialized social, emotional, and academic challenges of these children and their families. Further studies such as this are greatly needed to address the diverse range of problems associated with ARND and help direct treatment-oriented research. Such efforts may potentially help to circumvent many of the serious secondary problems which pose an enormous cost to individuals with ARND, their families, and society.

Table 1

Measures Used as Part of the ARND Diagnostic Test Battery

| Measure | Age Group |
|---|---|
| Intellectual Functioning | |
| Wechsler Intelligence Scale for Children (WISC-III: 13 subtests) | school |
| Wechsler Preschool and Primary Scale of Intelligence (WPPSI-R): 7 subtests | preschool |
| Wechsler Adult Intelligence Scale-Third Edition (WAIS-III): 11subtests | adult |
| McCarthy Scales of Children's Abilities: 20 subtests | preschool |
| Memory | |
| Wide Range Assessment of Memory and Learning (WRAML): 9 subtests | school |
| Denman Neuropsychological Memory Test: 2 subtests | school/adult |
| Language | |
| Peabody Picture Vocabulary Test-Third Edition (PPVT-IIIR) | all |
| Expressive One Word Picture Vocabulary Test (EOWPVT-R) | preschool/school |
| Preschool Language Scale-Third Edition (PLS-3) | preschool |
| Token Test for Children | preschool/school |
| Test for the Reception of Grammar (TROG) | preschool/school |
| Test of Language Development-2 (TOLD2): 1 subtest Grammatic Comprehension | school |
| Test of Language Competence (TLC): 2 subtests | school/adult |
| Visual-Motor Co-ordination | |
| Beery-Buktenica Developmental Test of Visual-Motor Integration-4th Edition (VMI) | all |
| Grooved Pegboard | school/adult |
| Attention/Executive Functioning | |
| Connor Continuous Performance Test (CPT) (kiddie/ standard versions) | all |
| TRAILS A&B | school/adult |
| Wisconsin Card Sorting Test | school/adult |
| Verbal Fluency Test | school/adult |
| Academic Functioning | |
| Wide Range Achievment Test (WRAT-3) | school/adult |
| EINSTEIN | preschool |
| Woodcock Reading and Mastery Tests: 1 subtest Passage Comprehension | school/adult |
| Keymath: Time and Money | school/adult |
| Behaviour and Socioemotional Functioning | |
| Conners' Parent Rating Scale (CRS-R) | all |
| Carey Temperament Scales | preschool/school |
| Werry Weis Peters Activity Scale | preschool/school |
| Child Behaviour Checklist (CBCL) | all |

Note. preschool=4-5 years, schoolage= 6-16 years, adult=>16 years

Table 2

Diagnostic Criteria Checklist Used with the ARND Sample

| Deficits | Assets |
|---|---|
| decreased intelligence | (relatively) good visuospatial skills |
| poor math | good face recognition |
| poor reading comprehension | air of competence/self-confidence |
| chattiness | good rote memory |
| anomia | good verbal fluency |
| poor comprehension | good immediate object memory |
| problems with word meanings | |
| difficulty with sentence structure | |
| problem with pragmatics | |
| perseverative | |
| poor gross & fine motor | |
| poor time management/planning | |
| poor organization/planning | |
| poor memory | |
| poor associative learning | |
| concrete thinkers | |
| poor social skills | |
| behaviour problems | |
| poor attention/ADHD | |
| high activity | |
| poor adaptive skills | |

Table 3

Scoring Breakdown for Composite Scores

| | | | |
|---|---|---|---|
| **Home/Social Background** | | | |
| Number of homes lived in prior to testing | 0= | 2= | 3= |
| | 1 | 2-3 | 4+/orphanage |
| Amount of time spent in current home | 0= | 2= | 3= |
| | 4+years | 2-3 years | <1 year |
| Number of children living in current home | 0= | 2= | 3= |
| | 1-2 | 3 | 4+ |
| Abuse/Neglect history | 0= | 2= | 3= |
| | No | Suspected | Yes |
| SES | 0= | 2= | 3= |
| | 1-2 | 3-4 | 5 |
| | Home/Social Background score: | | /15 |
| Education and Treatment History | | | |
| Repeated a grade | 0= | 2= | 3= |
| | No | -- | Yes |
| Received treatment in school | 0= | 2= | 3= |
| (i.e. modified program) | No | Pending | Yes |
| Received treatment outside of school | 0= | 2= | 3= |
| | No | Pending | Yes |
| Diagnosed with ADHD: | 0= | 2= | 3= |
| | No | -- | Yes |
| Treated with medication: | 0= | 2= | 3= |
| (related to attention and/or | 1-2 | 3-4 | 5 |
| other behavioural/emotional issues) | | | |
| | Education and Treatment History score: /15 | | |

Table 4

Demographic Information for the Matched Pair Sample

| | Group (n) | |
|---|---|---|
| | ARND (33) | Control (33) |
| Gender | 19 males | 19 males |
| | 14 females | 14 females |
| Mean and Range of Age at Testing in years | 8.36 (4-14.83) | 8.37 (4.17-14.25) |
| Distribution of matched pairs among SES categories | | |
| Major business and professional | | 1=21.2% (n=14) |
| Medium business, minor professional, technical | | 2=21.2% (n=14) |
| Skilled craftsmen, clerical, sales workers | | 3=30.3% (n=20) |
| Machine operators, semi-skilled workers | | 4=18.2% (n=12) |
| Unskilled laborers, menial service workers | | 5=09.1% (n=06) |
| Confirmed Exposure History in ARND group (n=35): | 17 | |

Note. 2 female subjects ages 16 and 18 years were included in the (non-matched pair) multiple regression sample (n=35)

Table 5

Mean T scores on CBCL Broad Band, Narrow Band, and Competence Scales

| | Group | | |
|---|---|---|---|
| | ARND | Control | Adjusted pvalue $p \leq 0.05/10=0.005$ |
| Broad Band Scales | M (SD) | M (SD) | |
| Total Problems | 68.33 (8.64) | 45.21 (8.69) | 0.001 |
| Externalizing Problems | 64.64 (10.78) | 46.21 (9.37) | 0.001 |
| Internalizing Problems | 60.06 (12.35) | 46.42 (7.77) | 0.001 |
| Narrow Band Scales | | | |
| Withdrawn | 60.30 (8.80) | 52.03 (4.08) | 0.001 |
| Somatic Complaints | 58.85 (9.99) | 51.85 (3.31) | |
| Anxiety/Depression | 58.67 (9.74) | 52.42 (3.87) | |
| Social Problems | 68.82 (10.94) | 52.73 (5.35) | 0.001 |
| Thought Problems | 64.88 (12.10) | 53.12 (6.03) | 0.001 |
| Attention Problems | 75.12 (11.63) | 52.64 (4.21) | 0.001 |
| Delinquency | 66.42 (9.35) | 51.82 (3.59) | 0.001 |
| Aggression | 65.48 (10.85) | 52.39 (4.70) | 0.001 |
| Sex Problems | 61.34 (12.35) | 50.00 (0.00) | 0.001 |
| Competence Scales | | | |
| Activities | 43.64 (6.81) | 48.13 (8.16) | |
| School Competence | 27.33 (4.65) | 45.68 (6.30) | |
| Social Competence | 35.29 (8.30) | 46.04 (7.67) | |

Note. For broad and narrow band scales a T score $\geq 63$ is considered to be in the clinical range. For competence scales a T score $\leq 40$ is considered to be in the clinical range.

Table 6

Group Differences in Clinical Range Scores on the CBCL Broad-Band Scales

| | Group | | |
|---|---|---|---|
| | ARND n=35 | Control n=33 | p value |
| Broad Band Scales | | | |
| Total Problems | 27 (77%) | 0 | <.0001 |
| Externalizing Problems | 26 (74%) | 2 (6%) | <.0001 |
| Internalizing Problems | 11 (31%) | 0 | 0.0004 |

Table 7

Group Differences on CBCL Intraclass Correlation Coefficients (ICC profiles)

| | Group | | |
|---|---|---|---|
| | ARND | Control | McNemar's *p* |
| Number of children with positive ICC profiles | 57% (n=20/35) | 12% (n=4/33) | 0.001 |
| Frequencies of positive ICC profiles | | | |
| ICC-Withdrawn | 1 | 2 | |
| ICC-Somatic | 2 | 1 | |
| ICC-Social | 12 | 0 | |
| ICC-Social/Attention | 4 | 1 | |
| ICC-Withdrawn/Anxiety/-Depression/Attention | 0 | 0 | |
| ICC-Delinquent/Aggressive | 6 | 0 | |
| ICC-Delinquent | 7 | 1 | |

Table 8

Most Meaningful Differences Between Matched Pairs on the 118 Individual CBCL Items Based on Frequencies and Ratios

| Item | Group (n) | |
|---|---|---|
| | ARND (33) | Control (33) |
| Acts too young for his/her age[a] | 28 | 6 |
| Argues[a] | 28 | 20 |
| Can't concentrate/poor attention[a] | 32 | 13 |
| Can't sit still/restless/hyperactive[a] | 28 | 13 |
| Confused/seems to be in a fog[b] | 23 | 1 |
| Disobedient at home[a] | 30 | 18 |
| Disobedient at school[b] | 22 | 3 |
| Doesn't get along with other kids[b] | 24 | 3 |
| Doesn't feel guilty after misbehaving[b] | 26 | 2 |
| Impulsive/acts without thinking[a] | 29 | 10 |
| Lying or cheating[b] | 26 | 6 |
| Poor schoolwork[b] | 26 | 2 |
| Showing off/clowning[a] | 27 | 16 |
| Sudden changes in mood or feelings[b] | 22 | 3 |

Note. [a]Over 80% of the ARND sample had endorsements on these items
[b]At least 58% more of ARND subjects than controls had endorsements on these items

Table 9

Significant Differences in Frequencies Between Matched Pairs on the Individual CBCL Items

| | GROUP | | |
|---|---|---|---|
| | ARND | Control | unadjusted p value |
| acts too young for his/her age | 28 | 06 | .001 |
| argues a lot | 28 | 20 | .03 |
| bragging, boasting | 20 | 08 | .005 |
| can't concentrate, can't pay attention for long | 32 | 13 | .001 |
| can't get mind off certain thoughts; obsessions | 17 | 07 | .025 |
| can't sit still, restless, or hyperactive | 28 | 13 | .001 |
| clings to adults or too dependent | 21 | 11 | .03 |
| confused or seems to be in a fog | 23 | 01 | .001 |
| cries a lot | 18 | 06 | .003 |
| cruelty, bullying, or meanness to others | 19 | 02 | .001 |
| day-dreams or gets lost in his/her thoughts | 22 | 06 | .001 |
| demands a lot of attention | 25 | 14 | .008 |
| destroys his/her own things | 19 | 04 | .001 |
| destroys things belonging to others | 19 | 02 | .001 |
| disobedient at home | 30 | 18 | .001 |
| disobedient at school | 22 | 03 | .001 |
| doesn't eat well | 11 | 08 | .044 |
| doesn't get along with other kids | 24 | 03 | .001 |
| doesn't seem to feel guilty after misbehaving | 26 | 02 | .001 |
| easily jealous | 18 | 10 | .046 |
| fears he/she might think or do something bad | 11 | 03 | .021 |
| feels he/she has to be perfect | 03 | 14 | .005 |
| feels or complains that no one loves him/her | 11 | 04 | .035 |
| feels others are out to get him/her | 10 | 03 | .035 |

Significant Differences in Frequencies Between Matched Pairs on the Individual CBCL Items (continued)

| | GROUP | | |
|---|---|---|---|
| | ARND | Control | unadjusted p value |
| feels worthless or inferior | 10 | 03 | .035 |
| gets hurt a lot, accident-prone | 17 | 04 | .002 |
| gets in many fights | 19 | 02 | .001 |
| gets teased a lot | 19 | 06 | .005 |
| hangs around with others who get in trouble | 12 | 02 | .004 |
| impulsive or acts without thinking | 29 | 10 | .001 |
| lying or cheating | 26 | 06 | .001 |
| bites fingernails | 14 | 06 | .021 |
| nervous, high-strung, or tense | 15 | 04 | .008 |
| nervous movements | 12 | 03 | .007 |
| not liked by other kids | 17 | 01 | .001 |
| constipated, doesn't move bowels | 01 | 05 | .046 |
| too fearful or anxious | 13 | 05 | .021 |
| aches or pains | 06 | 01 | .025 |
| headaches | 11 | 03 | .005 |
| problems with eyes | 07 | 01 | .034 |
| picks nose, skin, or other parts of body | 22 | 08 | .003 |
| poor school work | 26 | 02 | .001 |
| poorly coordinated or clumsy | 18 | 04 | .001 |
| prefers being with younger kids | 21 | 12 | .039 |
| refuses to talk | 13 | 03 | .004 |
| repeats certain acts over and over; compulsions | 11 | 01 | .004 |
| screams a lot | 13 | 05 | .046 |
| showing off or clowning | 27 | 16 | .002 |
| sleeps less than most kids | 15 | 06 | .02 |

Significant Differences in Frequencies Between Matched Pairs on the Individual CBCL Items (continued)

| | GROUP | | |
|---|---|---|---|
| | ARND | Control | unadjusted p value |
| speech problems | 18 | 03 | .001 |
| stares blankly | 13 | 01 | .001 |
| steals at home | 11 | 01 | .004 |
| strange behaviour | 13 | 01 | .001 |
| stubborn, sullen, or irritable | 24 | 11 | .003 |
| sudden changes in mood or feelings | 22 | 03 | .001 |
| sulks a lot | 19 | 03 | .001 |
| swearing or obscene language | 15 | 01 | .001 |
| teases a lot | 19 | 08 | .005 |
| temper tantrums or hot temper | 23 | 10 | .002 |
| trouble sleeping | 13 | 03 | .008 |
| unhappy, sad, or depressed | 12 | 02 | .004 |
| unusually loud | 19 | 09 | .008 |
| whining | 18 | 07 | .002 |
| withdrawn, doesn't get involved with others | 12 | 01 | .008 |

Table 10

Frequencies Across Qualitative Categories: Greatest Strengths Characterizing the ARND Group

| Strengths Categories (n=33) | Example Descriptors | % of ARND Group | Males: Females |
|---|---|---|---|
| Loving Nature | compassionate, affectionate, big heart loving/loveable, warm-hearted | 64 | 13:8 |
| Zest for Life/Sense of Humour | positive, happy, full of life, joyful to be around, enthusiastic, passionate, quick to smile, funny, happy-go-lucky, humour, laughter | 36 | 8:4 |
| Hobbies/Creativity/Athleticism | love for sports/outdoors/ nature/horses, crafts, artistic/creative, wonderful imagination | 33 | 6:5 |
| Academic/Cognitive Trait | reads well, memory, good at French, visual memory; people/places, smart | 15 | 2:3 |
| Positive Approach/Motivation & Helping Behaviour | willing to try, caring, generous, kind, willing to help smaller children, loves/tries hard/wants to please, loves to work, persistent | 49 | 8:8 |
| Outgoing Nature | active, adventurous, go-getter, friendly, outgoing personality, inquisitive | 15 | 4:1 |

Table 11

Frequencies Across Qualitative Categories: Greatest Concerns Characterizing the ARND Group

| Concerns Categories (n=29) | Example Descriptors | % of ARND Group | Males: Females |
|---|---|---|---|
| Attention/Impulsivity | distractible, impulsive, hyperactivity, short attention | 28 | 5:3 |
| Cognitive Problems | language/speech, learning, misinterpretation of information, math/times tables, listening, lack of reasoning and logic, intellectual problems, memory, ability to learn from past mistakes | 62 | 9:9 |
| Adaptive Life Skills & Future | poor life skills, self-care, money, hygiene, poor sense of time/space/money, future/independence, follow basic routines, safety, no sense of danger, uninhibited with/no fear of strangers, easily influenced by others, daily life stuff/can't remember if he ate or not | 52 | 10:5 |
| Socioemotional Functioning | sudden rages, frustration, social skills, emotional insensitivity, tantrums, mood, goes from one extreme to another, low self-esteem, manipulative, narcissistic, laughs inappropriately | 41 | 8:4 |
| Behaviour & Self-Regulation | noise level/loud, startles easily, behaviour, perseverative, activity, does not complete tasks, loses control, cannot stop irritating behaviour on request, unpredictable, balance/clumsiness | 52 | 8:7 |
| Delinquency | obsession with death/killing, lies, aggression, scratch/bite, noncompliance, manipulative/dishonesty | 17 | 4:1 |

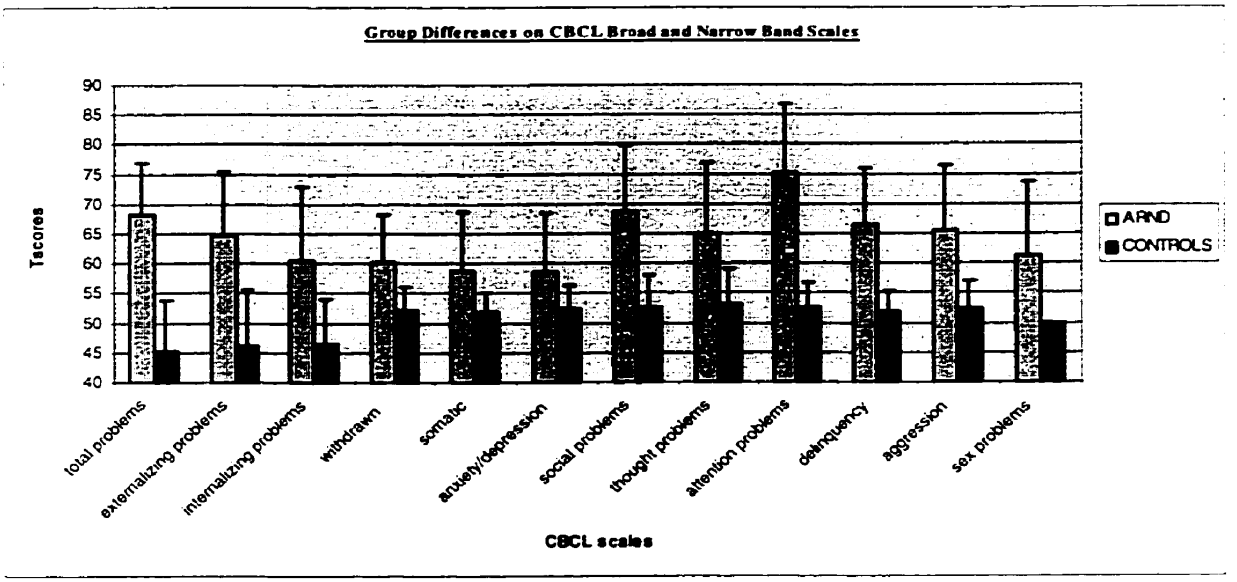


Figure 1. Group Differences in T scores on CBCL Broad and Narrow Band Scales

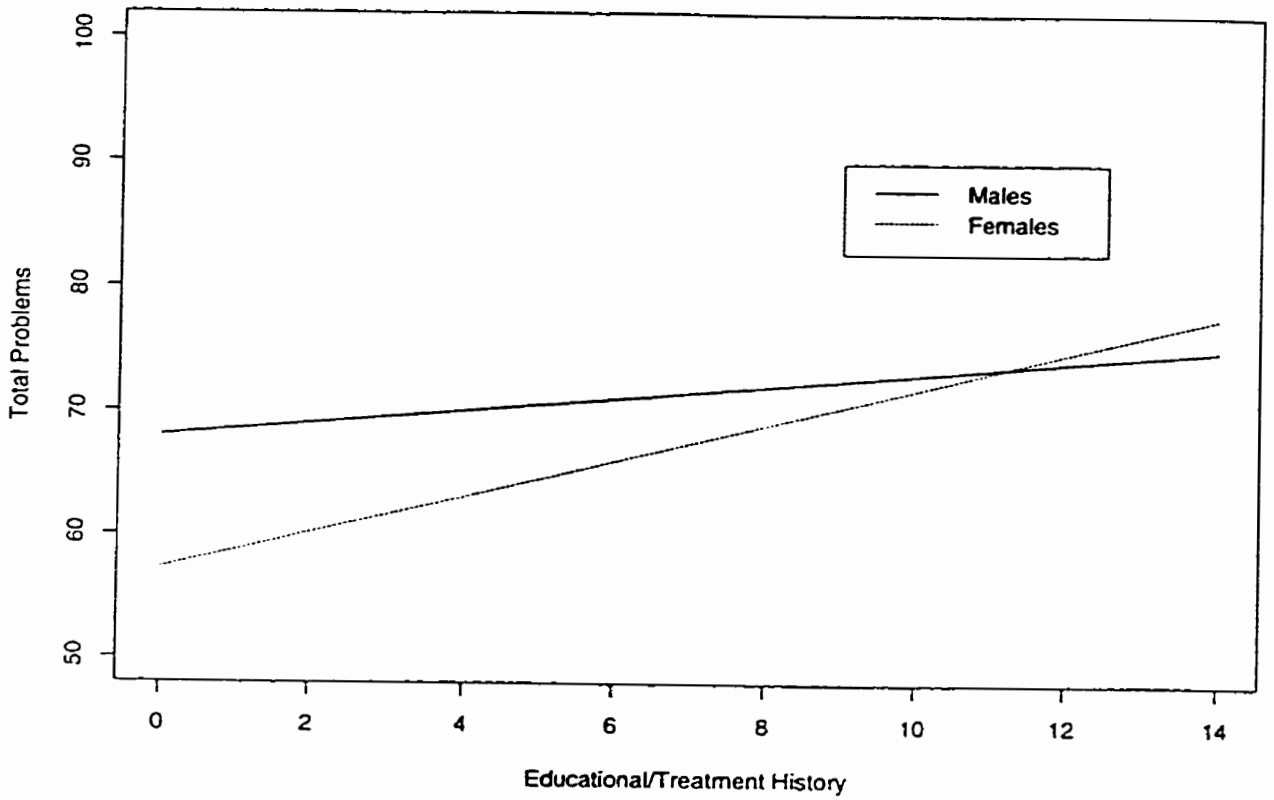


Figure 2. Interaction between Gender and Educational/Treatment History in the Prediction of Total Problems on the CBCL

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