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LANGUAGE AND LITERACY OUTCOMES FROM A PILOT INTERVENTION STUDY FOR CHILDREN WITH FASD IN SOUTH AFRICA

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Abstract

This pilot study investigated the efficacy of a classroom language and literacy intervention in children with fetal alcohol spectrum disorders (FASD) in the Western Cape Province of South Africa. The study forms part of a larger, on-going study that includes metacognitive and family support interventions in addition to language and literacy training (LLT). For the LLT study, 65 nine year old children identified as either FASD or not prenatally exposed to alcohol, were recruited. Forty

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children with FASD were randomly assigned to either a Language and Literacy Training (LLT) intervention group or FASD control group (FASD-C). Twenty-five non alcohol-exposed children were randomly selected as non-exposed controls (NONEXP-C). Prior to intervention and after 9 school-term months of treatment, general scholastic tests, teacher and parent questionnaires, classroom observations and specific language and literacy tests were administered to the participants. The 9 months assessment reflects the mid-point and the first assessment stage of the overall study. At initial diagnosis and prior to commencement of the interventions, participants with FASD were significantly weaker than non-exposed control children in reading, spelling, addition, subtraction, phonological awareness and other tests of early literacy. Teachers rated a range of adaptive behaviors of children with FASD as significantly worse than non-exposed controls. Mean scholastic and language and literacy scores for all groups showed improvement over baseline scores after 9 months of intervention. The mean test scores of children with FASD remained lower than those of nonexposed controls. Comparison of mean baseline to post-intervention score changes between the LLT, FASD-C and NONEXP-C groups revealed that although there were no significant gains by the LLT intervention group over control groups on the general scholastic assessment battery, significantly greater improvements occurred in the LLT intervention group compared to the FASD-C group in specific categories of language and early literacy. These categories were syllable manipulation, letter sound knowledge, written letters, word reading and non-word reading and spelling. In spite of cognitive and classroom behavioral difficulties, children with FASD from a vulnerable environment demonstrated significant cognitive improvements in specific areas targeted by classroom interventions. To our knowledge, this is the first report of a systematic classroom intervention and resultant cognitive response in children with FASD.

Keywords

Fetal alcohol spectrum disorder (FASD); fetal alcohol syndrome; education; classroom interventions; South Africa

Introduction

The term fetal alcohol spectrum disorder (FASD) encompasses a continuum of mild to severe physical and neurobehavioral effects in children exposed to alcohol in the prenatal period. Although considerable attention has been devoted to delineating the neurobehavioral functioning of children with FASD, relatively little is known about effective interventions for learning and behavioral problems in these children. In 1996, a fetal alcohol syndrome (FAS) study committee of the Institute of Medicine (IOM) recommended the evaluation of effectiveness of educational interventions on children pre-natally exposed to alcohol (Stratton et al., 1996). Even before the IOM report, the need for developing intervention programs for alcohol-affected children had been recognized (Carmichael-Olson et al., 1992), yet few systematic outcomes studies of intervention programs exist. Studies have been reported by O'Connor et al., 2006, Premji et al., 2007, and Riley et al., 2003.

The lack of research on effective interventions to ameliorate behavioral and learning problems in children with FASD can be attributed to a number of factors. First, despite extensive literature on the neurobehavioral functioning of children with FASD (Connor et al., 2000; Kodituwakku et al., 2001; Mattson et al., 1999; Mattson and Riley, 1998; Riley & McGee, 2005) there is no consensus yet among researchers on their cognitive and neurobehavioral phenotype. Designing effective treatment for a given disorder requires specifying central (core) and peripheral deficits and strengths associated with that disorder. Second, researchers with a specialty in intervention have not had access to large groups of well-diagnosed children within the FASD continuum, thus preventing the design of systematic outcome studies that employ rigorous scientific

methodology. Third, there has been minimal exchange of ideas between educators, clinicians, and basic scientists with regard to development of effective intervention programs.

Although the core deficits in FASD have not been clearly delineated, sufficient information exists about areas of deficit (general intelligence, language, executive function, memory, attention, gross and fine motor skills) and relative potential strengths, to provide a base for development of appropriate interventions (Kalberg & Buckley, 2006). In studies conducted in South Africa (May, et al., 2000; Viljoen, et al., 2005; May et al., 2007), we also found that children with FAS were impaired not only in verbal and non-verbal IQ, but also in tasks that measured fundamental language skills such as grammar comprehension and memory (Adnams et al., 2001; Kodituwakku et al., 2006).

An intervention study has the potential to shed light on the cognitive phenotype of children with FASD. Interventions aimed at certain deficits may effect improvements in other deficits, thus helping to identify primary (core) and secondary deficits. Identification of the mechanisms underlying neurobehavioral change in children with FASD should eventually lead to the development of effective intervention methods (Kalberg & Buckley, 2006).

FASD constitutes a significant public health problem in high risk populations in South Africa. Previous epidemiology studies have indicated a high prevalence of FASD in school children aged 5 to 9 years in this high risk region in the Western Cape (May et al., 2000; Viljoen et al., 2005; May et al., 2007). This has yielded a large cohort of children diagnosed within the FASD continuum, as well as controls from a similar ethnic, socioeconomic, language and educational background. Previous neurobehavioural studies in this region demonstrated that besides deficits in verbal and non-verbal intelligence, children with FASD in this community have fine motor skill, visuo-spatial and behavior problems compared to controls (Adnams et al., 2001; Viljoen et al., 2005; May et al., 2007).

It is known that deficits in academic and adaptive skills are associated with impoverished living conditions (McLoyd, 1990; Bradley et al., 2001; Barnett, 1998; Liddell & Rae, 2001). Poverty, low maternal education, and depression are all contributory factors that have prevailed in the cohort studied here. The children identified in these studies are Coloured (mixed ancestry of Black African, White, and Malaysian decent) and have inherited an educational system that continues to suffer from the legacy of South Africa's apartheid era. More recent changes in the country's national education policy, consistent with the international trend towards inclusive education, provide for inclusion of children with mild intellectual and learning disabilities within the mainstream system (South African Government Department of Education, 2001). Most children with FASD fall in the category of mild intellectual disability and remain in mainstream education in South Africa, in spite of the fact that they may have significantly disabling behavioral, attentional, and executive functioning problems (Kodituwakku et al., 2001). The burden of FASD in school-aged South African children thus falls heavily on a poorly resourced state education system. Given the high prevalence and the socio-political context of FASD in the Western Cape region, it is not only a research, but an ethical imperative to address systematic interventions aimed at amelioration of learning and behavior disabilities in children identified with FASD. A previous pilot intervention study undertaken in this community demonstrated improvements in self-regulation and behavior in children with FASD (Riley et al., 2003).

South African school education is reliant on the medium of spoken language for most instruction and this is likely to exacerbate primary deficits of language in children with FASD. Spoken language can be analysed into large speech units (words and syllables e.g. "pram") intermediate speech units (onsets, /pr-am/ and rimes, /pr-am/) and small speech units, (phonemes e.g. /p-r-a-m/). Phonological awareness refers to a person's awareness that

language has a sound structure. It is the ability, aside from ascribing meaning, to recognise and manipulate words into smaller units such as syllable, intrasyllabic structures and phonemic segments which can be blended, deleted and inserted. Speech, reading and phonological awareness all share the same speech processing system. Language deficits may undermine the complex processes involved in literacy acquisition and children presenting with deficits in vocabulary, phonology and expressive language have difficulty learning to read, spell and engage in creative writing activities. There is now an established causal link between early phonological awareness and reading achievement (Stahl, 1994) and there is a strong body of evidence that phonological training not only results in improved awareness, but also in better reading and spelling skills in at risk children (Lundberg et al., 1998, Ball & Blachman, 1991). Wimmer et al., 1991, reported that the predictive value of phonological awareness before instruction in first graders was independent of IQ and initial differences in letter knowledge and reading ability. Although phonological awareness is crucial to literacy development, successful reading outcomes depend on the learners' ability to link their phonological awareness skills to letter knowledge and reading experience. This is referred to as the phonological linkage hypothesis (Hatcher et al., 1997).

Since many of the mental representations involved in literacy are the same as those involved in receptive and expressive language activities, it is necessary to tailor a language and literacy program to address both the language and literacy deficits in children. Many programs implementing phonological awareness training to children at risk for reading difficulties have been developed over the past two decades. Few such programs have been developed and tested in the South African context. The multi-sensory phonological awareness and letter knowledge program was devised by Nadler-Nir, 1997, who conducted a study on disadvantaged first graders in the Western Cape Province of South Africa. The intervention focused on improving various components of phonological awareness, including segmentation, blending, manipulation and letter knowledge. Results of the study indicated that the program was highly effective in improving phonological awareness, letter knowledge, reading and spelling skills.

Although there are many tests of phonological awareness, the use of a variety of measures to tap both simple and compound phonological awareness skills is desirable. The phonological awareness tests used in this study (Table 1), conform to a range of levels of difficulty from the most basic, or lower order, to complex higher order phoneme manipulation.

The overall study was undertaken to evaluate the efficacy of three intervention methods on children with FASD from a community in South Africa. The aim of this study focuses on outcomes of one of the interventions. Given the background of language deficits and thus risk for literacy disability in children with FASD, the specific aim was to determine the degree to which a classroom language and literacy intervention program improved academic skills in children exposed to high levels of alcohol. We hypothesise that the core, or fixed deficit in children with FASD is ameliorable to a degree yet unknown, and that children with FASD can improve in specific academic skills after targeted interventions.

Materials and Methods

Participants

The participants recruited in the larger intervention study were 105 third grade children (53 males and 51 females) from 10 schools in a high risk region in the Western Cape. For the present study, 40 of the above children with FASD and 25 non-exposed controls participated. The children with FASD were assigned to LLT intervention (20) and FASD controls (20). The participants were similar in age, socio-economic status and first language (Afrikaans). All were originally identified from an epidemiological study in which children were diagnosed by a 2 tier method of active case ascertainment (May et al., 2007). In the first tier, 818 children were

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screened for height, weight or head circumference $\leq 10^{\text{th}}$ centile. In the second tier these children underwent a dysmorphology examination and neuropsychological assessment, and their mothers were interviewed regarding maternal drinking during the child's gestation. The neuropsychological assessment at diagnosis consisted of a verbal and a non-verbal test and a teacher rated measure of behavior. A dysmorphology score was assigned, psychological test results reviewed, maternal history of alcohol use and abuse and factors of susceptibility and risk assessed, and a diagnosis made via a multidisciplinary case conference using revised 1996 Institute of Medicine (IOM) diagnostic criteria for FAS (Hoyme et al., 2005). The revised IOM criteria clarify the IOM diagnostic categories that were proposed in 1996 and represent the continuum of effects within FASD. The diagnostic scheme of the revised IOM categories is as follows: Children with FAS (with or without confirmed maternal alcohol exposure) must have a combination of abnormalities in all domains, i.e., facial dysmorphic features, growth, and brain growth or structure. In the partial fetal alcohol syndrome (PFAS) category (with or without confirmed maternal alcohol exposure), children must display typical facial dysmorphic features and abnormalities in 1 of the other domains, namely growth (height or weight) or central nervous system structure or function). Thus, in PFAS abnormalities in all domains are not necessary. The central nervous system function abnormalities in FAS and FASD is evidenced by patterns of behavioral and cognitive abnormalities including impairments in the performance of complex tasks, higher-level receptive and expressive language deficits, motor dysfunction, poor academic performance and deficient social interaction. The deferred category was reserved for those children who, when seen early in the study, received a medical evaluation and a preliminary diagnosis of 'possible FAS'. They were subsequently not placed into the diagnostic categories FAS or Partial FAS at the case conference stage.

Maternal interviews were obtained on all children except where they were no longer in the care of their birth mothers or families. In most of these cases there was collateral validation of heavy maternal alcohol consumption during pregnancy and the children had been placed in foster or adoptive care or in a state children's home. Informed consent was obtained from all parents or legal guardians. In the case of non-literate caregivers, the consent was read and explained verbally. Active case ascertainment in the epidemiology study yielded 75 children with FASD who received a diagnosis as FAS, PFAS or Deferred. Forty (40) of these children with FASD were randomly assigned to the treatment group and a control group (FASD-C) of the language and literacy intervention. There is robust literature reporting the detrimental effect of poverty and socio-economic disadvantage on IQ, school achievement and socio-emotional functioning (McLoyd, 1990). The FASD-C group was therefore included to account for the influence of these factors on the classroom cognitive and adaptive functioning of the children with FASD who received interventions. Of the 40 prenatally alcohol-exposed children in the language and literacy intervention study, 27 had a diagnosis of FAS, 8 had a diagnosis of Partial FAS and 3 were designated as Deferred due to positive stigmata on initial 2nd tier dysmorphology screen. A further 2 alcohol-exposed children were included in the FASD sample in the study, but did not meet criteria for diagnosis of FAS, Partial FAS or Deferred and were excluded from the analysis in this report. Therefore, study participants included in the analysis represent the most severe levels of the FASD continuum.

The non-exposed controls were recruited from 1) 193 children who were randomly selected in the beginning of the epidemiologic study and then confirmed as unexposed and, 2) children who did not demonstrate features of FAS, Partial FAS or Deferred during the 2nd tier of screening for dysmorphology. The latter were also eligible as non-exposed controls if maternal histories reported no alcohol intake during pregnancy. Following a second random selection from all non-exposed children, 25 were included as non-exposed controls (NONEXP-C). (See Participant characteristics Table 2). By virtue of selection from the 1st tier of screening, the NONEXP-C group included children whose growth was $\leq 10^{th}$ centile. Forty one (41) % of children in the randomly selected control group demonstrated similar growth impairments.

Given the high (25%) prevalence of growth stunting in all South African children under 5 years (Monson et.al., 2006; Labadorios, 1999), the NONEXP-C group could be reasonably expected to represent that general local population, in which low socio-economic circumstances and under-nutrition prevail.

Study Design

This is a randomized, controlled intervention study comparing a FASD intervention group with FASD and non-exposed controls. Following randomization to participant groups, scholastic and language and literacy tests were administered to all participants in order to establish a baseline for intervention. To exclude sensory impairments which may impede learning progress, visual acuity was screened by the Snellen method (Snellen, 1862). A Snellen standard block letter and 'Falling E'' eye chart at a distance of 20 feet were used for acuity evaluation. The visual acuity of all participants tested in the normal range. Similarly, participants' hearing was tested using free field audiometry. No significant hearing impairments were detected. The duration of the intervention program was 9 school-term months after which the baseline scholastic and language and literacy tests were repeated. Assessments were administered by Afrikaans speaking test administrators who were blinded to diagnosis and intervention group assignment.

Language and literacy intervention

Language and literacy intervention commenced in the third formal year of school when the participants were between 9 and 10 years old. The program was administered for half an hour twice a week by an experienced speech and language therapist. A total of 38 hours of therapy was undertaken over the 9 month period (19 hours of language therapy alternating with 19 hours of phonological awareness and literacy training). Intervention sessions took place at a designated classroom of one of the participating schools and were administered to children in groups of five. Groups were initially constituted according to location of school attended but were re-arranged according to children's abilities. The program focused on training in phonological awareness and acquisition of other pre- and early literacy skills necessary for competency in reading and spelling (see Table 3).

Neuropsychological Assessment at Diagnosis

Tests were administered to participants at the time of diagnosis, 2 years prior to commencement of the intervention, in the first school grade (aged 7 years), and constituted the 2nd tier neuropsychological assessment of the initial diagnostic phase:

- 1. *Test for the Reception of Grammar (T.R.O.G.,* Bishop, 1989). This test consists of identification of 80 items depicting increasing complexity of syntactic and lexical grammar, and has been shown to be useful as a measure of verbal abilities in children with developmental disabilities. Because TROG is designed to provide qualitative information about which aspects of grammar give difficulty, as well as an overall score, TROG items are grouped in blocks of four. The TROG has been used in previous studies on children with FASD in South Africa. The score is expressed as a percentile.
- 2. *Raven Colored Progressive Matrices* (Raven, 1947). This cross-cultural test assesses non-verbal problem solving based on matching components of patterns and is normed for children. It has been used widely in cognitive research on developing countries. This test relies on percentile scores for interpretation. Above the 95th centile is intellectually superior; above the 75th centile is above average intellectual capacity; between the 25th and 75th centiles is intellectually average; below the 25th centile is

below average intellectual capacity and at or below the 5th centile is intellectual impairment.

3. Personal Behaviors Checklist - 36 (PBCL – 36). (Streissguth et. al., 1998).

This measure is a checklist which assesses 36 behavioral features that typically characterize persons with FASD. This instrument is not normed. Behaviors for a child are rated by an observer, either the child's caregiver, or as in this study, teacher. A higher score denotes behaviors characteristic of children with FASD.

Teacher Reports

Teacher questionnaires were administered to all participants before the intervention.

Achenbach Teacher Rating Scales (Achenbach and Rescorla, 2001)—These teacher report questionnaires rate behaviors in six Diagnostic and Statistical Manual-IV oriented categories. The categories reported here are: 1) Attention Deficit and 2) Hyperactivity Problems. A higher score in any category indicates a greater problem behavior related to that DSM-IV category.

Classroom Behaviors (Wood et al., 2002)—The Wood Teacher Questionnaire consists of 10 items describing classroom behavior and adaptive functioning. The raters observe the following on-task academic behaviors in the classroom: 1.) being in one's chair or otherwise appropriately out of seat; 2.) using materials appropriately; 3.) working on the assigned task; 4.) following the teacher's directions; 5.) appropriate participation in class discussions; 6.) paying attention; 7.) starting assignments as soon as the teacher finishes instructions; 8.) asking for teacher assistance if needed; 9.) accepting teacher feedback appropriately; and 10.) not talking to or touching other students or their materials unless directed by the teacher. A higher score represents better behaviour and adaptive functioning. The questionnaire included relative frequency of positive behaviors and the total score was expressed as a percentage.

Children's Perceived Self-Efficacy Scale (Bandura et al., 1996)—The adapted 47 item questionnaire of general and classroom self-efficacy was administered to teachers. The scale is designed to assess self-efficacy representing 8 domains of functioning: enlisting resources, academic achievement, self-regulated learning, extracurricular activities, self-regulatory efficacy, meeting other's expectation, self-assertive efficacy and enlisting others' support. Self efficacy function was reflected as a composite score. A higher score represents greater self-efficacy. We have used this scale previously with children in South Africa, and have found it to be a relevant and valuable measure. In this study, because the intervention outcomes focused on classroom parameters, questionnaires were completed by teachers rather than by children's caregivers.

Intervention Outcome Measures

The general cognitive and scholastic tests were administered to evaluate baseline performance and longitudinal change in key school-based learning areas: reading, spelling and mathematics addition and subtraction. Given the predominant use of spoken language as a medium of instruction in the South African education system, the scholastic battery provided a means to assess whether an improvement in language, specifically targeted by the intervention, might influence other areas of classroom learning and performance.

The UCT (University of Cape Town) Reading Test and UCT Spelling Test—The UCT (University of Cape Town) Reading Test and UCT Spelling Test are graded tests of reading and spelling, normed in 1985 for Afrikaans speaking Coloured learners in the Western Cape by the Child Guidance Clinic of the University of Cape Town. The norms for boys and

girls range from 6 years - 14.5 years. The tests are commonly used in the Western Cape for educational and neuropsychological testing.

The Ballard One-Minute Addition and Subtraction Arithmetic Tests—The Ballard One-Minute Addition and Subtraction Arithmetic Tests are scholastic tests of subtraction, each consisting of 28 graded items and including basic calculations using single and double digit numbers.

Phonological Awareness and Early Literacy Test (PAELT)—This consists of detailed standardized and adapted non-standardized language subtests As no standardized Afrikaans Phonological Awareness test exists, the components follow a similar set of subtests of an English battery developed by Byrne and Fielding-Barnsley, 1993, to measure lower order (prelinguistic) as well as higher order composite literacy skills. The English tests were adapted and translated into Afrikaans (Sorour, P., Hugo, H., & Pieterse, H., 2001, unpublished; Nadler-Nir, E., 2001, unpublished) and this battery underwent further modification by authors of this study. (Table 1). The PAELT subtest scores are expressed as percentage correct of total for that subtest, with a maximum score of 100 for each test.

Note

South African normed tests were used for the scholastic battery and other assessments where possible. Tests were translated where necessary and administered in Afrikaans. Raw scores are reported in this study and were used for analysis for the cognitive and scholastic tests, including those tests normally expressed as age equivalents. The rationale for this is twofold. First, this was to ensure consistency in analysis of study data. Some tests used in the study are not normed for South African children and others have different norms for girls and boys. Second, it was necessary to maximally capture pre- to mid- test score gains. In the scholastic tests conventionally reported in terms of age equivalents, at pre-intervention assessment there was a "floor effect" in scores of the children who were weakest on those tests, particularly when the test floor was set at an age equivalent of 6 years. The extent of any post-intervention score gains in performance from a "below the floor" baseline in weaker children would thus not be reflected in post-intervention scores.

Statistical analyses

Data were analyzed using the Statistical Package for the Social Sciences (SPSS 10.0) (2000). Grouped data were expressed as means and standard deviations. Analysis of variance (ANOVA) was performed to compute between-group effects when participant characteristics for three groups (LLT, FASD-C, NONEXP-C) were compared. Pearson's correlation was used to examine associations between sample characteristics (head circumference, dysmorphology score and verbal cognition and behaviour at diagnosis). The efficacy of treatment with LLT was evaluated using the independent t-test to compare the mean change in intervention to postintervention PAELT scores of LLT with non-treatment groups (LLT vs. FASD-C) and (LLT vs. NONEXP-C). For independent t-test, Levene's test was used to determine equality of variances. Where homogeneity of variance could not be assumed t-test was reported with adjusted df. The standardized mean difference treatment effect size d, was corrected for bias using Hedges' modification (Hedges and Orkin 1995). The rule of thumb of Cohen (1988) was used to interpret the effect size as follows: 0.20 = a small change; 0.50 = a medium change and 0.80 = a large change. Wolf (1986) suggests that a standardized mean effect size of 0.25 is educationally significant and 0.50 is clinically significant.

Results

Participant Attrition—At post-intervention assessment four children were lost to the study (2 of 20 FASD children in the LLT group and 2 of 25 non-exposed controls). Three of these children moved away from the area, and the parents of the fourth control child withdrew consent. The 2 FASD-C children who did not meet diagnostic criteria for FAS, Partial FAS or Deferred were excluded from the statistical analysis in order that this report reflected only those participants with a diagnosis on the more severe spectrum of FASD. The diagnosis of participants who were included for final analysis was as follows: LLT: n = 18 (FAS = 13, PFAS = 3, Deferred = 2); FASD-C: n = 18 (FAS = 13, PFAS = 4, Deferred = 1); NONEXPC: n = 23.

Comparison of LLT, FASD-C and NONEXP-C Groups

Participant Characteristics—An ANOVA for differences in age at baseline demonstrated no statistically significant difference among the 3 groups [F(2,58) = 0.960, p = 0.389]. At the time of diagnosis in the first grade year, there was a significant between-group difference between LLT, FASD-C and NONEXP-C in dysmorphology score [F(2,58) = 51.00, p = .000];head circumference [F(2,58) = 21.07, p = .000]; language (TROG) [F(2,58) = 6.96, p = .002] and behavior (PBCL) [F(2,58) = 13.69, p = .000]. (Table 2). These findings indicate that the children with FASD had significantly more dysmorphic characteristics, abnormalities of brain growth, and impairments in language and behavior than the control group. Prior to intervention, teachers' ratings of classroom behavior showed significant between-group differences for inattention: [F(2,53) = 19.681, p = .000], hyperactivity [F(2,53) = 5.288, p = .008] positive behaviors (Woods): [F(2,58) = 6.887, p = .002] and self efficacy [F(2,58) = 8.447, p = .001]. demonstrating that the control group had significantly fewer behavioral problems noted by their teachers. Independent t-tests yielded no significant differences between LLT and FASD-C groups for these characteristics. The similarity of the LLT and FASD-C characteristics allowed the assumption that these two groups are derived from the same population for age, verbal intelligence and behavior. Thus, no further adjustment for characteristic-derived differences on these parameters was made in other LLT vs. FASD-C comparisons. Given that a gender effect on language might influence the response to the language and literacy intervention, a 1-way ANOVA test was conducted with one between-subject factor (gender) and one within group factor (TROG). This revealed no effect of gender on language in the study population [F(1,58) = .096, p = .758].

Baseline General Scholastic Tests—Independent t-test showed no statistically significant differences between the LLT and FASD-C groups in general scholastic tests of mathematics: addition (t = -.457, p = .651), subtraction (t = .93, p = .926); UCT reading (t = -1.46, p = .152) and UCT spelling (t = -1.44, p = .152). (Table 4). The children with FASD were significantly weaker than non-exposed controls in the same scholastic tests. For these measures ANOVA demonstrated statistically significant between-group differences for LLT, FASD-C and NONEXP-C: addition [F(2,58) = 17,139, p = .000]; subtraction [F(2,58) = 18.676, p = .000; UCT reading [F(2,58) = 16.239, p = .000]; UCT spelling [F(2,58) = 11.576, p = .000]; UCT spelling [F(2,58) = 10.576, p = .000]; UCT spell 000]. There was no effect of gender on reading measured by the UCT Reading Test [F(1,58) = 0.838, p = 0.364] nor on spelling (UCT Spelling Test) [F(1,58) = 1.952, p = 0.168]. The nonexposed control group performed below the age average for most normed tests. We used raw test scores to record results of outcome measures and for analysis. To demonstrate that children with FASD performed "below the floor" on normed tests, the percentage of participants whose baseline scores fell below the test floor (cut off age equivalent = 6.0 years) in the general scholastic tests was calculated for the LLT and FASD-C groups combined (n = 36). The percentage of FASD scores below the test floor in the respective outcome measures were: Ballard Addition: 50%; Ballard Subtraction: 68.4%; UCT Reading Test: 28.9%; UCT spelling: 21%.

Baseline PAELT—The baseline and post-intervention PAELT scores for the LLT, FASD-C and NONEXP-C groups are shown in Table 5. The NONEXP-C group achieved higher scores than the FASD groups on all subtests. There were no statistically significant differences between the LLT and FASD-C groups on subtests of the PAELT administered at baseline assessment. As a measure of composite pre-intervention phonological awareness skills presented by the PAELT subtests, a phonological representation score was computed for each participant, excluding the reading and spelling subtest components. This composite score reflected the sum of scores of 8 phonological awareness subtests: segmentation of first, last, all sounds; blending syllables, blending phonemes, manipulating syllables, manipulating phonemes and letter sounds. The composite phonological representation scores for the groups were: LLT: Mean = 439.8; SD = 211.6; FASD-C: Mean = 511.5, SD = 213.4; NONEXP-C: Mean= 681.3, SD = 73.7. An ANOVA revealed statistically significant group difference between LLT, FASD-C and NONEXP-C [F(2,58) = 10.82, p = 0.000] indicating that the children with FASD had significant phonological representation deficits. Independent t-test showed the LLT and FASD-C group scores did not differ (t = -1.012, p = .32). Given the association of language and phonological function, an ANCOVA was performed to examine the effect of group diagnosis (FASD vs. NONEXP-C) on composite phonological representation. After controlling for verbal intelligence (TROG), the effect of group diagnosis remained significant [F(1,58) = 17.50, p = .000].

Outcomes of LLT Intervention—In order to assess change between baseline and postintervention for each outcome measure of the scholastic and PAELT measures, the mean difference between baseline and post-intervention scores was calculated for each group. At post intervention assessment, each group showed improvements over the baseline assessments in all the outcome measures.

This was expected due to the longitudinal nature of the study. The absolute scores of children with FASD (LLT and FASD-C groups) remained weaker than those of the NONEXP-C group. The pre- and post intervention scores for each group on the PAELT measures are shown in Table 5. Potential increase in achievement by the LLT group over the non-intervention groups was assessed by comparing the baseline to post-intervention score differences of the LLT group with the other groups (Table 6). Independent t-test demonstrated statistically significant gains by the LLT group over the FASD-C group in subtests of the PAELT. These gains were in the domains of syllable manipulation, written letters, word reading, non-word reading and nonword spelling. Pre- to post intervention changes in PAELT scores of the LLT and NONEXP-C group are shown in (Table 6). In the simpler subtests of phonological awareness (first sounds and last sounds), the NONEXP-C group attained the maximum score of 100% at baseline and post-intervention assessment, thus the score change in this group is is reflected as 0. In all other tests, except for one other post-intervention score, maximum scores were not achieved. T-test comparison of the score changes demonstrates significant catch up by the LLT to the NONEXP-C group in several categories of the PAELT subtests, including the more complex early literacy categories such as written letters, reading and spelling of words and non-words.

In the FASD groups there was a significant negative correlation between behavior at diagnosis and positive classroom behavior (Woods): r = -0.58, p = .000; self-efficacy: r = -0.454, p = .000 and the components of language and early literacy in which the LLT group showed statistically significant gains: syllable manipulation: r = -0.48, p = .000; letter sounds: r = -0.46, p = .000; written letters: r = -0.493, p = .000; word reading: r = -.60, p = .000 and nonword reading: r = -.52, p = .000. In other words, these results indicate there was a significant association between behavior problems noted by teachers at the time of diagnosis and a lack of positive classroom behaviors, lower self efficacy and poor language and early literacy function.

Post-intervention, there were no statistically significant difference gains of the LLT group over the FASD-C group in the general scholastic tests. (Table 4). In addition, the LLT and FASD-C groups remained significantly weaker than the NONEXP-C group on the scholastic tests: addition: (t, = -5.44, p = .000); subtraction: (t, -4.41, p = .000); (UCT) reading: (t, -4.74, p = .000); (UCT) spelling: (t, -4.92, p = .000).

Effect Size and Clinical Significance—Functional gains on measurements of PAELT, reflected by effect size d, by the LLT group over the FASD-C group, ranged from 0.31 to 1.2, that is from small to large. (Table 6). Effect size d was large for measurements that were statistically significant. This indicates that the mean gain for the LLT group was 0.31 to 1.2 Standard Deviations above the increase in scores over time of the FASD-C group. Most gains were in the educationally significant range (> 0.25) even when the gain size did not reach statistical significance.

Discussion

The study results confirm the hypothesis that children with FASD can improve in specific skills in response to targeted cognitive interventions. Significant gains over FASD controls were made following administration of a program that targeted literacy and linguistic skills. This outcome is consistent with other intervention studies in environmentally and intellectually vulnerable but non-alcohol exposed children at risk for reading and literacy disabilities (Nadler-Nir, 1997; Conners et al., 2006) and suggests that children with FASD can benefit from language and literacy programs designed for other learning disabled and at-risk children.

The effect size of the gains ranged from small to very large. Functional gains made by the children undergoing LLT intervention compared with FASD-controls, were mostly educationally and clinically significant. The effect size expressed the increase in achievement of the treatment group over the group not exposed to intervention. The greatest effect occurred in the categories of more complex phonological representation and early literacy. Scores for lower order subtests, for example first sounds and last sounds, indicate that, prior to intervention, the FASD groups had acquired phonological skills in these areas. The LLT group also made significant gains over the non-exposed control on some areas of phonological and early literacy function, reflecting some catch-up to the controls. Post intervention gains in non-word reading by the LLT group could possibly mediate other cognitive processes associated with both phonological and working memory.

The study results show that deficits of phonological representation are independent of verbal intelligence in children with FASD. This finding supports evidence from studies of children who are non alcohol-exposed (Wimmer et al., 1991) and suggests that a deficit in phonological awareness, a key pre-function for reading function, may be a primary cognitive phenotypic characteristic in children with FASD.

Consistent with other findings in disadvantaged South African populations, the non alcoholexposed children performed below average for normed tests. In the same general scholastic tests, the LLT group did not make significant gains over exposed controls and the observed gains remained specific to the area of intervention. A possible explanation for the lack of improvement in reading and spelling in the UCT tests may be due to the tests themselves. The sharp incremental grading in the UCT tests may have discriminated against weaker participants and failed to reflect gains of the LLT group, in spite of the use of raw scores to optimize measurement of gains. Not surprisingly, there were no LLT gains over controls in mathematics, although Gathercole, 2006, has described deficits in phonological awareness as a contributing factor to poor mathematics in children with reading disabilities. This underscores the need for provision of comprehensive programs addressing general learning deficits and strengths in children with FASD, in addition to targeted interventions.

As expected, the FASD groups exhibited worse classroom behaviors than the non-exposed children. There was an association between behavior at time of diagnosis and limitations in language and early literacy gains. Adaptive and social skill deficits in FASD have been shown to be independent of IQ to some degree (Riley & McGee, 2005) and the early detection of problems in this area provides an opportunity for intervention to prevent or minimize secondary disabilities associated with impairments in social function and learning.

A limitation of this research was that cognitive mechanisms were not adequately assessed prior to the intervention. There were no specific measurements of other mediators of literacy, such as working memory, short term memory and visuo-spatial short term memory. The normed scholastic tests proved difficult for some children in the study and failed to accurately measure changes in function at the low end of the test. This detracted from the interpretability of the scholastic outcome measures. Ideally an intervention should also target younger children in order to maximize the intervention effect before formal schooling commences. Inclusion of a non alcohol-exposed intervention group would have allowed comparison of LLT outcomes in children with FASD with a treatment response in normally developing children. This would have contributed information on how children with FASD learn.

The outcome in the LLT group following one hour per week of language and literacy intervention for 9 school months, reflected an improvement from pre-literacy skill levels averaging 2 or more years of delay for chronological age, to that of some functional early literacy. Gains towards this functionality, if maintained, could have implications for greater personal autonomy at a later stage. Although the gains demonstrated are described for children with FASD in mainstream education in the context of a developing country, the outcomes will aim to contribute to information on appropriate and evidence-based interventions for all children with FASD.

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Table 1	
Subtests of the Phonological Awareness and Early Literacy Test (PA	ELT)

Phonological Awareness and Early Literacy Test		SUBTESTS	Number of items
A: Rhyme	A1	Rhyme awareness	10
-	A2	Rhyme production	
B: Segmentation	B1	First sounds	8
-	B2	Last sounds	8
	B3	All sounds	22
C: Blending	C1	Blending syllables	21
e	C2	Blending phonemes	13
D. Manipulation	D1	Manipulation syllables	12
I.	D2	Manipulation phonemes	36
E: Letter Knowledge	E1	Letter sound	42
U	E2	Writing of letters	42
F: Reading	F1	Reading Real words	18
5	F2	Reading Non-words	16
G: Spelling	G1	Spell Real words	15
	G2	Spelling Non-words	15
H: Reading Paragraphs	H1-5	Connected sentences	5 Passages

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		LLT (n - 18)			FASD-C (n - 18)			NONEXP-C $(n - 33)$	
Variable	%	Mean	SD	%	Mean	SD	%	Mean	SD
Child age (yrs) Child gender		9.52	(0.56)		9.63	(0.65)		9.42	(0.25)
Boys Girls	50.0 50.0			55.6 44.4			47.8 52.2		
Dysmorphology score at diagnosis ^a		17.78	(3.57)		18.55	(3.63)		7.65	(4.33)
Head circumference at diagnosis (cm)		48.82	(1.15)		48.43	(1.59)		50.93	(1.28)
Verbal score at diagnosis ^b (percentile)		11.45	(18.19)		9.62	(11.94)		26.09	(16.13)
Nonverbal score at diagnosis ^c									
(percentile)		11.28	(12.28)		9.06	(7.28)		16.62	(11.86)
Behavior at diagnosis ^d Classroom Behaviors		11.67	(8.18)		9.72	(7.58)		1.96	(2.58)
Inattention ^e		6.47	(3.79)		5.00	(3.46)		0.86	(1.25)
Hyperactivity ^e		4.59	(4.69)		3.40	(3.76)		1.05	(1.76)
Positive behaviors ^f		52.22	(17.68)		55.56	(20.14)		71.74	(17.03)
Self efficacy ^g		87.89	(36.16)		95.00	(31.79)		126.26	(28.97)

 $a_{\rm I}$ Institute of Medicine Revised Diagnostic Criteria for fetal alcohol syndrome

 b Test of Reception of Grammar

Alcohol. Author manuscript; available in PMC 2008 September 1.

^cRaven Colored Progressive Matrices

d Problem Behaviors Checklist PBCL -36

 e Achenbach Teacher Rating Scales

fwoods Classroom Behavior Score

 $^{\mathcal{B}}$ Children's Perceived Self Efficacy Rating Scale

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Table 2

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Table 3

Language and Literacy Training: Principles, Method and Materials Used in the Intervention Program

 Eungauge	and Enteracy Training. I	miciples, Method and Materials Used in the fi	iter vention i rogram
Literacy component	Principles of training	Method:	Materials
Phoneme Identification and Letter Knowledge	-Multi-sensory -Repetition Short vowels Long vowels Diphthongs	Multi-sensory stimulation exercises in the writing and reading of letters (consonant and vowel). The trainer articulated the sounds and paired the sound with a picture explaining the connection between the sound and the written letter. The learner was asked to look at flash cards, then try to write the letters, first by tracing sandpaper letters, then by completing dotted lines and lastly the children attempted to write the letters on their own.	 Sandpaper letters, silicone, pipe cleaners Writing letter in the air Writing letter in icing sugar Write the letter (paper) Plastic letters Lettercards
Segmentation and Blending	-Normal sequence of development Order of training: 1. compound words 2. syllables 3. phonemes 4. clusters e.g." spr ing"	Plastic or cardboard letters were used to teach segmentation and blending of single phonemes. For segmentation children were expected to sound out phonemes of short words and later more difficult words with diphthongs and finally multisyllabic words e.g.k-a-t k-l-a-p = "klap" The following order was followed in phoneme segmentation tasks: initial sounds, final sounds, medial sounds, and finally phoneme clusters. Real words were used for written spelling tasks in writing tasks	 Pictures cut to represent syllables Picture cards of whole words that could be combined to make compound words were used to teach segmentation and blending of words e.g. "blom" and "tuin" = "blomtuin". Auditory input only (without pictures)
Manipulation of syllables Manipulation of phonemes		The trainer would say compound words and ask the child to delete one of the words forming the compound word by firstly using picture card cues and then only producing auditory stimuli alone e.g. "skoolskoen" without "skoen" = "skool". Syllable deletion tasks were done using auditory stimuli alone, e.g. "vergaan" without "ver" = "gaan". Adding and exchanging phonemes. The most complex tasks are at the phoneme level e.g. say "Brush" without the /b/ = rush or say "splash" without the /p/=slash	 Picture card clues Auditory stimulus alone Plastic letters Coloured tokens for consonants and vowel Auditory stimulus only
Reading Real words Reading Non-words	-Vowel Consonant CV, VC VCV CVC, CVCC, CCVC	Word cards Short stories were used for reading practice. Simple story-books at the child's reading level were provided for group and individual practice Reading of Non-word practise sheets for initial medial and final sounds	 Individual real words printed or cards were given to each child at their level of progress. Non-words printed on cards
Spelling real word Non-word		Real words and non-words were used for spelling tasks at the child's level	- Worksheets, Games - Blackboard / whiteboard writing - Write the word (paper)
Language (semantic training)	- Increase semantic networks	Naming, tasks; semantic fields, category naming (superordinate/subordinate relations) Rapid automatic naming tasks (objects and pictures) Story comprehension tasks, Story retell (picture sequences)	 Objects and pictures Matching games FAS stories

There was a wide range of within group variability in performance, so it was not possible to use the same tasks for all children at the same time. Generally, the concepts were taught in this framework but the level was modified according to the strengths and weaknesses of individual learners in the groups. Language sessions alternated with literacy sessions in the first half of the intervention programme where after, the literacy training was emphasised. Each group was seen for 30 minutes twice weekly for a total of 76 sessions over a 2 year period. Verbal and material reinforcement (raisons, stickers, stars) were used.

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Descriptive Statistics of Baseline Scores and Baseline to Post-intervention Change in General Scholastic Test Scores for Language and Literacy Training

Table 4

(LLT), FASD Control (FASD-C) and Non-exposed (NONEXP-C) Control Groups

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T1 - T2

NONEXP-C

FASD-C

LLT

NONEXP-C

FASD-C

LLT

E

8.61 (10.71)

7.22 (7.90)

8.88 (7.36)

32.48 (11.85) 8.0-8.5 yr

17.55 12.17) 7.0-7.5 yr

11.44 (12.82) 7.0-7.5 yr

8.00 (3.90)

5.50 (6.59)

4.50 (4.35)

23.65 (11.01) 8.0 - 8.5 yr

13.55 (8.75) 7.0 - 7.5 yr

9.11 (9.74) 7.0 - 7.5 yr

3.30(3.18)

2.72 (3.95)

3.05 (2.67)

13.74 (5.07) 7.6 - 7.11 yr

6.50 (3.60) < 6 yr

5.78 (5.66) < 6yr

کیں SD Age Equivalent Ballard Subtraction Test Mean

SD Age Equivalent

Age Equivalent Ballard Addition Test Mean Aor

Age Equivalent UCT Spelling Test Agen Mean

Measure UCT Reading Test Mean

Scholastic Outcome

3.48 (3.63)

2.11 (2.80)

1.67 (2.11)

9.87 (4.81) 7.6 -7.11 yr

3.11 (3.66) < 6 yr

3.22 (3.50) < 6yr

T1 = baseline

T1-T2 = baseline to post-intervention change (increase) in test scores

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Table 5 Descriptive Statistics of Language and Literacy Training (LLT) with FASD (FASD-C) and Non-exposed (NONEXP-C) Controls: Assessment with the Phonological Awareness and Early Literacy Test (PAELT)

		T1 Score (percentage)			T2 Score (percentage)	
PAELT TEST						
Measure	LLT Mean (SD)	FASD-C Mean (SD)	NONEXP-C Mean (SD)	LLT Mean (SD)	FASD-C Mean (SD)	NONEXP-C Mean (SD)
First sounds segmentation	79.86 (35.65)	87.5 (26.43)	100.00 (0.0)	97.62 (6.43)	93.06 (23.57)	100.00 (0.0)
Last sounds segmentation	72.22 (36.27)	81.25 (37.44)	100.00(0.0)	95.14 (17.75)	94.44 (23.57)	100.00(0.0)
All sounds segmentation	43.89 (34.41)	56.67 (29.05)	73.26 (17.36)	65.00 (32.90)	68.33 (28.49)	92.61 (10.21)
Blending syllables	58.99 (27.91)	63.49 (28.85)	84.68 (15.19)	88.37 (9.12)	83.59 (20.35)	95.65 (7.97)
Blending phonemes	49.11 (32.75)	58.54 (38.06)	78.91 (15.77)	79.06 (26.87)	78.20 (28.56)	93.64 (8.26)
Manipulation syllables	54.63 (40.17)	69.44 (38.04)	93.48 (12.80)	86.12 (25.24)	79.62 (34.33)	100.00(0.0)
Manipulation phonemes	18.21 (27.93)	27.16 (28.40)	62.43 (25.44)	43.82 (30.11)	41.21 (34.41)	89.61 (16.90)
Letter sounds	62.84 (22.49)	67.46 (29.27)	88.52 (11.23)	84.52 (19.12)	76.44 (29.59)	91.62 (6.83)
Writing letters	56.23 (25.56)	65.74 (28.81)	83.85 (10.63)	70.89 (24.82)	70.24 (31.16)	90.07 (9.80)
Reading words	39.51 (36.14)	64.20 (39.11)	86.96 (20.28)	66.04 (32.43)	69.44 (39.52)	92.84 (12.39)
Reading non-words	42.73 (33.85)	60.09 (37.14)	81.00 (16.37)	72.59 (29.17)	65.99 (39.07)	92.13 (16.00)
Spelling	34.07 (30.16)	49.99 (35.37)	72.75 (17.86)	54.08 (31.09)	60.74 (37.29)	86.66 (18.53)
Spelling, non-word T1 = baseline	27.04 (25.40)	38.52 (27.30)	55.07 (18.01)	55.18 (27.80)	44.44 (34.83)	68.68 (16.14)
T2 = post-intervention of 9 school months	months					
Maximum score for each test = 100%	%(

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Comparison between Language and Literacy Intervention and Non-Intervention Group (FASD vs. FASD-C) and between LLT and Non-exposed Control Group (LLT vs. NONEXP-C) for Baseline to Post-Intervention Change in Scores on the Phonological Awareness and Early Literacy Test (PAELT). LLT vs. FASD - C Table 6

			LLI VS. FADU - U						LLI VS. NUNEAF - U	EAF-C	
Outcome Measure	LLT % (n = 18)	FASD - C % (n = 18)	đf	+	Significance <i>p</i>	Effect size d	NFASD - C % (n = 23)	đ	t	Significance <i>p</i>	Effect size d
1^{st} sounds ^{<i>a</i>} Mean (SD)	18.06 (30.98)	5.55 (12.29)	34	1.59	NS	0.52^{**}	0.0 (0.0)	17	2.47	0.024	0.81^{***}
Last sounds ² Mean (SD) All sounds ²	22.92 (29.78) Mean (SD)	13.19 (31.64) 21.11 (19.22)	34 11.66 (14.35)	0.949 34 1.67	NS NS	$0.31^{*}_{0.54}$	0.0 (0.0) 19.35 (13.67)	17 39	3.26 0.34	0.005 NS	1.06^{***} 0.13
Blending Syllables Mean (SD)	29.37 (22.71)	20.10 (19.22)	34	1.32	NS	0.43^{*}	10.97 (8.78)	21	3.25	0.004	1.04^{***}
Blending Phonemes Mean (SD)	29.91 (19.89)	19.66 (23.78)	34	1.40	NS	0.46^*	14.73 (12.26)	39	3.00	0.005	0.90^{***}
Manipulating Syllables Mean (SD)	31.49 (36.00)	10.18 (18.42)	25	2.23	0.034	0.73^{**}	6.52 (12.80)	20	2.81	0.011	0.90^{***}
Manpulating Phonemes Mean (SD)	25.61 (22.45)	14.05 (27.04)	34	1.40	NS	0.45*	27.18 (13.63)	39	2.61	NS	0.08
Letter Sounds Mean (SD)	21.68 (12.66)	8.99 (7.24)	27	3.70	0.001	1.20^{***}	3.10 (10.37)	39	5.17	0.000	1.57^{***}
Written Letters Mean (SD)	14.66 (10.74)	4.51 (8.56)	34	3.14	0.004	1.02^{***}	6.22 (6.42)	39	3.13	0.003	0.93^{***}
Keading Mean (SD)	26.53 (22.97)	5.24 (7.95)	21	3.72	0.001	1.21^{***}	5.88~(10.88)	23	3.52	0.002	1.12^{***}
Reading Non-words Mean (SD)	29.87 (23.53)	5.90 (14.93)	34	3.65	0.001	1.19^{***}	11.13 (15.06)	39	3.01	0.004	0.93^{***}
Spelling Mean (SD)	20.06 (18.45)	10.76 (13.93)	34	1.70	NS	0.55^{**}	13.91 (11.01)	39	1.32	NS	0.39^*
Spelling Non-Words Mean (SD)	28.14 (13.61)	5.92 (18.84)	34	3.44	0.002	1.12^{***}	13.61 (16.26)	39	2.56	0.014	0.78^{**}
Standardized Mean Difference Effect Size d: The test scores reflect the percentage change (%) from the baseline assessment to the post-intervention assessment	iterence Effect Size the percentage chang	d: ge (%) from the base	eline assessment to	the post-inter	vention assessment						

 $^{*}_{0.2 = \text{small}}$

** 0.5 =medium

*** 0.8 = large

^aFirst Sounds segmentation

b Last Sounds Segmentation

 c All Sounds Segmentation