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GoFAR: Improving Attention, Behavior and Adaptive Functioning in Children with Fetal Alcohol Spectrum Disorders (FASD): Brief Report

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Abstract

Objective: This brief report describes the GoFAR intervention designed to improve attention, behaviour and adaptive functioning in children with FASD, ages 5 to 10 years.

Methods: 30 children were randomized to one of 3 conditions: GoFAR; FACELAND and CONTROL; 25 completed the interventions. Over 10 sessions children and caregivers learned a metacognitive strategy (FAR) designed to improve cognitive control of behaviour and adaptive functioning and practiced it during behaviour analog therapy. Attention, behaviour problems and adaptive skills were measured pre and post intervention.

Results: From pre to post testing the GoFAR intervention group improved on the Test of Variables of Attention (TOVA). Both intervention groups improved in Daily Living Skills.

Conclusion: This pilot study demonstrated that children with FASD and their caregivers benefit from a focused intervention designed to improve effortful control of behaviour. The study suggests the need for a larger clinical trial to evaluate the intervention's effectiveness.

Keywords

FASD; metacognitive strategies; parent training; computer game

Introduction

Since fetal alcohol syndrome (FAS) was identified more than 40 years ago [1], it has been acknowledged that Fetal Alcohol Spectrum Disorders (FASD) [2] are among the most common developmental disabilities with a population prevalence higher than initially imagined[3]. Still, despite the greater awareness of FASD in the last decades, most affected

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Declaration of Interest.

individuals are not receiving the intervention and treatment their caregivers feel is needed [4]. One basis for this discrepancy is lack of empirically validated interventions specific to this population [5]. While focused interventions can be effective for children with FASD [6] there are a number of areas in which more research is needed [5, 7].

In clinical practice, caregivers of children with FASD most often complain of disruptive behaviour and adaptive dysfunction while few interventions focus specifically on these issues [7]. GoFAR is an intervention for children, ages 5 to 10, who are alcohol affected. The focus of the intervention is the impairment in effortful control of arousal and behaviour that is associated with the effect of prenatal alcohol exposure (PAE) on frontal lobe functioning [8]. Deficits in behaviour control and executive functioning are often observed in children with PAE as well as the resulting deficits in self-regulation and adaptive functioning. GoFAR, a manualized intervention, provides children and caregivers with a metacognitive strategy for problem solving and assumes that effective use of this strategy will be associated with improvement in behavioural control, attention and adaptive function.

The GoFAR program, uses the FAR metacognitive technique [9, 10] to support behaviour change. FAR teaches children to Focus (F) their attention appropriately and make a plan to reach a goal or solve a problem. Then they learn to Act (A) on that plan. In the last step, children must Reflect (R) on what worked or did not work so that learning is consolidated. When these “rules” are internalized, they help the child structure attention and reduce impulsive responding. In addition to more positive behavioural outcomes, improved problem solving can lead to more positive social interactions as well as less frustration and destructive behaviour. The GoFAR program goal was to improve ability to regulate behaviour, control impulsive responding, demonstrate sustained attention and improve adaptive functioning at home.

The program has three elements: 1) “GoFAR”, a computer game played by the child to teach the FAR metacognitive control strategy; 2) individual parent behavioural regulation training; and 3) behavioural analog therapy sessions (BATS) during which the parent and child are supervised in implementing the FAR metacognitive strategy in the context of learning adaptive living skills. Program efficacy was measured by assessing three areas often deficit in FASD: sustained attention, behaviour/emotional problems and adaptive functioning. We compared those receiving the complete GoFAR intervention to two contrast groups: 1) a control group who received no intervention, and 2) a second intervention group who received a different computer game to evaluate the relative contribution of the computer game play. Those who received the full GoFAR intervention were predicted to show greater improvements in target outcomes as compared to those who received a different computer game for the child instructional component of the intervention and those who were time-elapsd Controls. Those who receive the parent instruction and conjoint parent and child training sessions but a different computer game were predicted to do better than those who were time-lapsd Controls.

Methods

Participants:

Thirty families with children ages 5 to 10 years of age who were prenatally affected by alcohol were recruited and randomly assigned to one of three intervention groups: 1) GoFAR; 2) FACELAND; or 3) CONTROL. Families were recruited from a multidisciplinary diagnostic clinic where a pediatric geneticist with specialized training in assessing alcohol-related dysmorphic features used a standardized dysmorphology checklist for diagnosis [11]. All study participants were alcohol exposed with significant levels of alcohol-related physical features or had a clinical diagnosis of FAS or partial FAS using the Institute of Medicine criteria [12]. Families with children qualified for the study were identified by clinicians from medical records under a Health Insurance Portability and Accountability Act (HIPAA) partial waiver and recruited by mail or in person. Caregivers attended a group workshop on the impact of PAE on neurodevelopmental functioning before being allowed to enroll in the study. Parents or guardians then completed the consent procedure approved by the Human Subjects Committee of the Emory University School of Medicine. Following consent, they were randomized into experimental groups.

Procedures:

Caregivers in both the GoFAR and the FACELAND groups received 5 parent training therapy sessions focusing on building behavioural regulation skills [10]. The intervention provided to children differed, with those in the FACELAND group playing a computer game to learn to recognize facial expressions associated with different emotions (e.g., fear, joy, anger) (FACELAND®) within an exploratory context while those in the GoFAR group receiving computer instruction that was developed specifically to teach the child to employ the FAR learning strategy that emphasized the need to control impulsive responding and plan future actions (GoFAR®) [9]. Subsequently, both groups of caregivers and children participated together in five behaviour analog therapy sessions (BATS) that provided direct instruction in using FAR in the context of learning the domestic adaptive skills required for daily living (e.g., picking up toys after play; dressing for school). CONTROL group participants served as time-elapsed controls and were yoked to a participant in one of the treatment groups to minimize group differences in time between assessments.

Outcomes:

Three areas, neurocognition, behaviour, and adaptive function, were used to measure effectiveness of GoFAR in improving child outcomes associated with FASD and to identify outcomes responsive to the intervention. The following tests were used: 1) Neurocognitive: Test of Variables of Attention (TOVA) [13], a computerized performance task (CPT) that measures sustained attention, ability to inhibit responding and attentional efficiency; and three NEPSY subtests, Inhibition Speeded Naming and Word Generation. These individually administered subtests measure ability to withhold inappropriate responses, efficiency in identification, rapid naming of perceptual items and ability to generate appropriate items in specific categories [14]; 2) Behaviour, measured with the following parent report questionnaires: Child Behaviour Questionnaire (CBQ), a measure of child temperament [15], Achenbach Child Behaviour Checklist (CBCL), a standard measure of behavior

problems in school-aged children [16], Behaviour Rating Scale of Executive Function (BRIEF), that measures factors associated with executive functioning and impact on behaviour [17]; and 3) Adaptive Functioning: Vineland Adaptive Behaviour Scales (VABS), 2nd Edition, a measure of everyday coping skills (e.g., ties shoes, picks up toys) necessary of efficient adaptive functioning [18]. Participants (parents and children) were evaluated before randomization and group assignment and again after the completion of the program. One of the focuses of this pilot project was the identification of appropriate outcome measures and of effect sizes to be used in planning future studies. Caregivers also were asked to rate their satisfaction with the program on a 5 point Likert scale. Finally, caregiver fidelity to treatment protocol was measured over the 5 BAT sessions.

Results and Conclusion

Twenty five of 30 families completed the study. See Table 1 for sample characteristics. One control family did not complete post testing and one child's comparatively more significant intellectual disability (I.Q. <60 vs the means in the 80's among the total group) prevented her from completing the neurocognitive measures. Others completed some but not all of the sessions.

Behavioural outcomes are shown in Table 2. In analyzing the behavioural outcomes for those who completed the study per protocol, a repeated measures analysis of covariance was used with the child's age at pretest as the covariate. Age was used as a covariate as it was significantly related to the outcomes of the TOVA and the CBQ as well as some of the subdomains of the VABS. Treatment group (GoFAR, FACELAND, CONTROL) was the between subject independent variable and time of assessment (Pretest/Post-test) was the repeated measure.

A significant difference was found on the summary score of the TOVA, the Attention Performance Index (API) that measures efficiency in sustaining attention and inhibiting impulsive responding. On this measure, children who received the GoFAR intervention, including the GoFAR game showed significant improvement at Post Test while the other two groups did not (Wald $\chi^2(2) = 6.09$, $p < .05$). In contrast, on the Vineland Daily Living Skills, Domestic subscale, which reflects the adaptive functioning in the home that was the focus of therapy, both intervention groups showed significant improvement relative to the performance of the Control group (Wald $\chi^2(1) = 5.39$, $p < .02$). Finally, on the CBQ, which measures Temperamental Functioning, Fear, one of the elements of Negative Affect was significantly reduced both when the three groups are compared (Wald $\chi^2(2) = 8.59$, $p < .01$) and when both intervention groups were combined (Wald $\chi^2(1) = 7.91$, $p < .005$). Other measures with substantial effect sizes did not achieve significance in this small sample.

When parent fidelity over the BATS sessions was rated, there was significant improvement in parent fidelity in carrying out the FAR methodology ($F_{(4, 13)} = 8.0$, $p < .002$, $\eta^2 = .71$) over the five sessions. (See Figure 1). There were no differences between the two Intervention groups which is to be expected as the caregivers did not experience different interventions. When parents were asked to rate their satisfaction with the GoFAR program, an important element in assuring program adherence, all responses were in the positive range with the

majority agreeing that the program was helpful and that they would recommend the program to others. There were no differences between the two groups of caregivers who received intervention in ratings of satisfaction.

These results suggest that even in this small pilot sample, it is possible to see positive effects of intervention on children with FASD. The goals of the GoFAR intervention were specific, to improve focus and reduce impulsivity and to improve adaptive functioning in children whose prenatal exposure has made the development of self-regulation challenging. In alcohol-affected children some of the effortful control skills that develop without specific instruction in typical children require more direct instruction to implement. Of course, interpretation of results is limited by the sample size and by the clinical nature of the participants. In addition, two of the measures, the VABS and the CBQ, are parent questionnaires and caregivers may be motivated to respond more positively after participating in 10 weeks of therapy although the TOVA is a computerized measure and cannot have a motivation of this kind. Future studies would benefit from independent observations of child outcomes, like teacher reports. It is also important to consider the characteristics of the sample used in this pilot. For instance, almost all of the children are in adoptive homes and many have had multiple placements. More than 50% are taking medications for behavioural problems. These environmental challenges may have influenced the results of this pilot study; future research with larger samples will allow assessment of the effects of such factors on outcomes.

In summary, the GoFAR program is a manualized intervention for children with FASD that was well accepted by children and caregivers. Previously reported results suggested that children's behaviour is improved by exposure to the FAR metacognitive strategy in that disruptive behaviour was significantly reduced [9]. In this brief report, we report that there is evidence also for improvement in sustained attention as well as in the practical adaptive outcomes that were the focus of the clinical intervention. Given these findings, future research should be carried out with larger and more diverse groups of children to demonstrate the effectiveness of these methods in supporting neurorehabilitation in this high risk group.

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Figure 1.
Mean group parent fidelity rating over the course of 5 Behavior Analog Therapy Sessions (BATS). There is significant improvement with experience but no group differences.

Table 1.

Characteristics of participants in GoFAR intervention pilot study.

Characteristic	GoFAR (n=10)	FACELAND (n=10)	CONTROL (n=10)	Statistic
Caregivers				
Age (yrs) M (SD)	49.6 (7.0)	46.8 (10.0)	51.5 (11.1)	$F_{(2,27)} < 1$, NS
Sex (% female)	90%	90%	90%	$X^2_{(2)} < 1$, NS
Education in Years M (SD)	14.5 (1.8)	13.8 (5.1)	13.9 (1.7)	$F_{(2,27)} < 1$, NS
Household Income Category (6=\$35,000–49,999/yr) M (SD)	7.4 (1.7)	6.0 (2.4)	5.8 (1.9)	$F_{92,23} = 1.83$, NS
Children				
Age (yrs) M (SD)	7.5 (1.4)	6.8 (1.3)	7.0 (1.6)	$F_{(2,27)} < 1$, NS
% Adopted or Legal Guardianship	100%	90%	100%	$X^2_{(8)} = 9.53$, NS
Sex (% male)	70%	40%	70%	$X^2_{(2)} = 3.73$, $p = .16$
Race: %Caucasian/African-American/Mixed Race	40%/10%/40%	50%/40%/10%	50%/40%/Other	$X^2_{(6)} = 8.34$, NS
Number of Custody Placements M (SD)	3.3 (2.7)	2.0 (1.1)	1.9 (1.1)	$F_{(2,27)} = 1.85$, NS
% Protective Services Involvement	80%	40%	54.5%	$X^2_{(2)} = 5.46$, $p = .07$
Birth Head Circumference/cm M (SD) (N=26)	32.3 (4.9)	30.4 (3.4)	32.1 (2.3)	$F_{(2,24)} < 1$, NS
Birthweight (gm) M (SD) (N=27)	2837.0 (1308.8)	2268.3 (638.8)	2444.5 (790.5)	$F_{(2,24)} < 1$, NS
Dysmorphology Score Total ¹ M (SD)	20.0 (3.6)	17.0 (4.1)	15.8 (7.0)	$F_{(2,28)} < 1$, NS
General Conceptual Ability ²	89.3 (11.3)	81.6 (19.5)	87.7 (11.3)	$F_{(2,27)} < 1$, NS

¹ This is the sum of the 30 weighted items on the Pediatric Checklist used for diagnosis of alcohol-related physical features. See Coles, et al. [11] for description.

² As measured by the Differential Ability Scales, 2nd Edition, Elliott, CD, 2007. This is a standard score with a mean of 100 and a standard deviation of 15.

Table 2.

Significant outcomes by group. Note that these results are based on the 25 participants completing the protocol.

Least square means and standard error by group status; per protocol (based on those completing protocol)									
	GOFAR		FACELAND		COMBINED TREATMENT		CONTROLS		Statistics (3-Group *Time, 2-Group *Time)
	Pretest	Post-test	Pretest	Post-test	Pretest	Post-test	Pretest	Post-test	
Attention Regulation									
TOVA API	-3.6 (1.0)	-0.8 (1.0)	-4.4 (1.1)	-3.2 (1.0)	-4.0 (0.79)	-2.1 (0.77)	-2.9 (1.2)	-2.9 (1.1)	Wald χ^2 (2) = 6.092, p < 0.048 Wald χ^2 (1) = 2.54, p < 0.111
TOVA Response Time	-81 (35)	-42 (59)	-1.3 (48)	-1.1 (58)	-1.1 (31)	-78 (42)	-47 (0.60)	-58 (45)	Wald χ^2 (2) = 5.506, p < 0.064 Wald χ^2 (1) = 2.021, p < 0.155
TOVA Resp. Time Variability	-2.5 (40)	-1.6 (51)	-2.8 (48)	-3.2 (88)	-2.6 (32)	-2.4 (56)	-2.1 (75)	-2.3 (53)	Wald χ^2 (2) = 4.81, p < 0.090 Wald χ^2 (1) = 0.319, p < 0.573
TOVA Errors of Omission	-7.7 (3.1)	-3.5 (1.3)	-10.9 (3.6)	-6.0 (1.9)	-9.4 (2.4)	-4.8 (1.2)	-14.1 (6.7)	-4.6 (7.7)	Wald χ^2 (2) = 0.519, p < 0.471 Wald χ^2 (1) = 0.537, p < 0.765
TOVA Errors of Commission	-1.0 (40)	-0.29 (40)	-2.0 (73)	-1.7 (55)	-1.5 (45)	-1.0 (39)	-1.6 (67)	-2.2 (75)	Wald χ^2 (2) = 5.018, p < .081 Wald χ^2 (1) = 3.677, p < .055
TOVA D-Prime	-2.0 (24)	-1.5 (16)	-2.8 (32)	-2.3 (27)	-3.0 (32)	-2.2 (19)	-3.0 (32)	-2.2 (19)	Wald χ^2 (2) = 0.741, p < 0.691 Wald χ^2 (1) = 0.734, p < 0.392
NEPSY-Auditory Attention-SS*	8.7 (1.2)	7.0 (1.3)	8.1 (1.4)	8.8 (1.4)	8.4 (0.96)	8.0 (0.98)	6.8 (1.0)	9.1 (1.6)	Wald χ^2 (2) = 8.568, p < 0.014 Wald χ^2 (1) = 4.780, p < 0.028
Cognitive Inhibition and Planning									
NEPSY-Inhibition/ Naming Combined	9.0 (1.8)	10.4 (1.4)	7.6 (1.5)	6.4 (1.3)	8.0 (1.2)	8.2 (1.1)	6.5 (2.0)	7.6 (1.2)	Wald χ^2 (2) = 2.111, p < 0.348 Wald χ^2 (1) = 0.341, p < 0.559
NEPSY-Inhibition/Inhibition	6.8 (1.4)	7.5 (1.4)	6.0 (1.0)	7.2 (1.3)	6.3 (0.83)	7.3 (0.95)	7.8 (1.6)	7.5 (1.7)	Wald χ^2 (2) = 1.375, p < 0.503 Wald χ^2 (1) = 1.226, p < 0.268
NEPSY Speeded Naming-Combined	7.8 (1.6)	9.0 (1.4)	6.8 (1.0)	5.7 (0.90)	7.3 (.94)	7.1 (0.89)	7.4 (1.3)	8.1 (1.3)	Wald χ^2 (2) = 2.542, p < 0.281 Wald χ^2 (1) = 0.304, p < 0.581
NEPSY Visuomotor Precision-Combined	9.6 (1.2)	7.8 (0.73)	6.7 (0.67)	7.4 (1.3)	8.0 (0.74)	7.6 (0.81)	6.3 (0.55)	6.9 (1.5)	Wald χ^2 (2) = 3.782, p < 0.152 Wald χ^2 (1) = 0.466, p < 0.495
NEPSY Word Generation-Semantic	9.1 (1.6)	9.4 (1.0)	8.0 (1.4)	8.0 (1.6)	8.4 (1.1)	8.6 (1.0)	8.7 (1.4)	9.4 (1.3)	Wald χ^2 (2) = 0.194, p < 0.907 Wald χ^2 (1) = 0.139, p < 0.710
Parent Perception of Child Behavior									
CBQ: Negative Affect	3.9 (0.36)	3.4 (0.19)	3.6 (0.26)	3.2 (0.22)	3.7 (0.22)	3.3 (0.15)	3.6 (0.19)	3.6 (0.20)	Wald χ^2 (2) = 2.403, p < 0.301 Wald χ^2 (1) = 2.372, p < 0.124

Least square means and standard error by group status; per protocol (based on those completing protocol)									
	GOFAR		FACELAND		COMBINED TREATMENT		CONTROLS		Statistics (3-Group*Time, 2-Group*Time)
	Pretest	Post-test	Pretest	Post-test	Pretest	Post-test	Pretest	Post-test	
CBQ: Fear	3.9 (0.55)	3.2 (0.20)	4.0 (0.26)	3.2 (0.30)	3.9 (0.28)	3.1 (0.20)	3.4 (0.34)	3.6 (0.27)	Wald χ^2 (2) = 8.590, $p < 0.014$ Wald χ^2 (1) = 7.907, $p < 0.005$
CBQ: Anger Frustration	4.7 (0.45)	4.0 (0.32)	3.9 (0.42)	3.6 (0.42)	4.2 (0.32)	3.8 (0.27)	4.1 (0.42)	4.3 (0.26)	Wald χ^2 (2) = 1.683, $p < 0.431$ Wald χ^2 (1) = 1.469, $p < 0.225$
CBQ: Discomfort	3.7 (0.55)	3.1 (0.27)	3.5 (0.24)	3.2 (0.17)	3.6 (0.27)	3.2 (0.16)	3.0 (0.23)	3.3 (0.19)	Wald χ^2 (2) = 2.888, $p < 0.236$ Wald χ^2 (1) = 2.875, $p < 0.090$
CBQ: Sadness	3.5 (0.28)	3.0 (0.23)	3.4 (0.28)	2.7 (0.20)	3.5 (0.20)	2.9 (0.15)	3.7 (0.15)	3.4 (0.23)	Wald χ^2 (2) = 0.991, $p < 0.609$ Wald χ^2 (1) = 0.744, $p < 0.388$
CBQ: Soothability	4.3 (0.26)	4.4 (0.20)	4.7 (0.28)	4.6 (0.31)	4.5 (0.20)	4.5 (0.20)	4.3 (0.24)	4.7 (0.27)	Wald χ^2 (2) = 2.469, $p < 0.291$ Wald χ^2 (1) = 1.348, $p < 0.246$
Effortful Control	4.1 (0.29)	3.7 (0.17)	3.7 (0.12)	3.7 (0.18)	3.9 (0.15)	3.7 (0.13)	3.8 (0.20)	3.8 (0.17)	Wald χ^2 (2) = 1.175, $p < 0.556$ Wald χ^2 (1) = 0.201, $p < 0.654$
Surgency	4.4 (0.28)	4.3 (0.23)	4.8 (0.27)	4.6 (0.20)	4.6 (0.20)	4.4 (0.16)	4.5 (0.23)	4.3 (0.11)	Wald χ^2 (2) = 0.132, $p < 0.936$ Wald χ^2 (1) = 0.004, $p < 0.950$
BRIEF: Executive Function Composite	74.8 (3.3)	68.3 (2.2)	75.6 (3.4)	71.1 (3.3)	75.3 (2.4)	69.9 (2.2)	73.0 (2.7)	69.0 (3.2)	Wald χ^2 (2) = 0.807, $p < 0.668$ Wald χ^2 (1) = 0.358, $p < 0.549$
BRIEF: Behavioral Regulation Index	71.8 (3.3)	66.2 (1.7)	71.4 (4.3)	67.2 (4.1)	71.6 (2.8)	66.8 (2.4)	73.7 (2.6)	68.4 (2.9)	Wald χ^2 (2) = 2.267, $p < 0.322$ Wald χ^2 (1) = 0.026, $p < 0.871$
BRIEF: Metacognitive Index	73.8 (3.9)	66.8 (2.9)	75.3 (3.1)	71.0 (2.8)	74.6 (2.4)	69.3 (2.2)	69.9 (3.0)	66.9 (3.5)	Wald χ^2 (2) = 2.202, $p < 0.333$ Wald χ^2 (1) = 1.131, $p < 0.288$
VABS Composite Scale	87.2 (3.6)	85.5 (3.1)	81.7 (3.6)	82.7 (3.0)	84.5 (2.6)	83.7 (2.2)	98.0 (9.4)	92.2 (2.9)	Wald χ^2 (2) = .873, $p < 0.646$ Wald χ^2 (1) = .584, $p < 0.445$
VABS Daily Living Skills	91.4 (4.1)	98.3 (6.8)	85.0 (4.2)	89.7 (4.1)	88.0 (3.0)	93.3 (4.3)	96.6 (4.5)	97.3 (4.6)	Wald χ^2 (2) = 1.399, $p < 0.497$ Wald χ^2 (1) = 1.147, $p < 0.284$
VABS DLS Domestic	12.9 (0.80)	15.3 (.91)	12.4 (1.0)	14.7 (0.51)	12.7 (0.66)	15.0 (0.53)	14.9 (0.83)	14.0 (0.94)	Wald χ^2 (2) = 5.637, $p < 0.060$ Wald χ^2 (1) = 5.386, $p < 0.020$
CBCL-Internalizing	61.4 (2.7)	61.1 (3.1)	59.0 (4.3)	55.5 (3.1)	60.0 (2.7)	57.6 (2.3)	62.5 (2.7)	60.7 (3.9)	Wald χ^2 (2) = 0.358, $p < 0.836$ Wald χ^2 (1) = 0.019, $p < 0.890$
CBCL-Externalizing	69.0 (2.7)	65.3 (2.3)	63.8 (1.9)	62.7 (2.4)	66.0 (1.7)	64.0 (1.8)	63.5 (2.8)	64.9 (2.4)	Wald χ^2 (2) = 3.445, $p < 0.179$ Wald χ^2 (1) = 1.783, $p < 0.182$
CBCL-Total	69.0 (1.9)	68.5 (1.3)	66.4 (2.3)	63.1 (2.3)	67.5 (1.6)	65.4 (1.7)	66.1 (3.0)	67.1 (3.0)	Wald χ^2 (2) = 1.041, $p < 0.594$ Wald χ^2 (1) = 0.665, $p < 0.415$

*These outcomes are significant but the control group is performing better!