Functional and Neuropsychological Abilities in Adults with Fetal Alcohol Spectrum Disorders

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Abstract: *Objective*: This study investigated functional and neuropsychological abilities in adults with mild to moderate fetal alcohol spectrum disorder (FASD).

Method: Sixty-eight participants completed various neuropsychological tests such as the California Verbal Learning Test – 2nd edition (CVLT-2), Trail Making Test parts A and B (TMTA/TMTB), Stroop Color and Word Test (Stroop), Grooved Pegboard Test (GPT), Finger Tapping Test (FTT), and Independent Living Scales (ILS). Independent samples *t*-tests were used to compare performance between brain scores 2 and 3. Hierarchical logistic regression was used to identify predictors for brain scores.

Results: Adults with severe impairment (i.e., a brain score of 3) performed significantly worse than those with milder brain dysfunction (i.e., a brain score of 2) on TMTB, Stroop, CVLT-2, and ILS (money management, and managing home and transportation subscales). Test scores from the CVLT-2 (long-delay cued recall) and ILS (money management subscale) predicted brain scores.

Conclusions: Psychosocial ability remained low in all participants and should be assessed as a discrete factor for guiding interventions in adults. The results provided an important piece of reference in support of the inclusion of psycho-emotional elements in the new diagnostic guidelines for FASD.

Keywords: Fetal alcohol spectrum disorder, neuropsychological assessment, functional assessment, cross-sectional study.

INTRODUCTION

Fetal alcohol spectrum disorder (FASD) is an umbrella term that encompasses many diagnoses. These diagnoses are characterized by the presence and severity of maternal alcohol exposure, facial dysmorphia, growth deficiency, and neurocognitive impairment [1,2]. Individuals demonstrate cognitive and affective impairments that disrupt mental processes such as attention, memory, planning inhibition, impulsivity, and judgment [3]. The impairments cause debilitating effects that emerge through secondary disabilities and progress across the life span. Some adverse effects are trouble with the law, inappropriate sexual behaviors, alcohol or drug addiction, employment difficulties, and problems with parenting [4]. Such effects can have profound implications for affected individuals, their families, support systems, and economic environment at a macro level [2,5]. Although early intervention has been described as a protective factor for promoting positive outcomes in adulthood, behavioral problems persist in adults with FASD [6]. Hence, there is a need for understanding the patterns of functional and neuropsychological performance in adults with FASD. The information could help predict which adults might be at risk of developing more significant functional deficits or struggle the most in everyday life.

The 4-Digit Diagnostic Code has been used to index the severity of FASD [1,2]. The brain score embedded in the 4-digit code reflects the functioning of the central nervous system (CNS) and provides information regarding neurological and functional deficits. The level of severity progresses from a brain score of 1 (no impairment) to 4 (severe impairment). Among them, brain scores 2 and 3 are the most commonly found diagnostic categories in clinics [7]. The former indicates possible evidence of CNS damage and describes the damage as mild to moderate [1]. The latter indicates probable evidence of CNS damage or significant global

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dysfunction as measured by standardized neuro-behavioral tests [2].

While the 4-digit-code has provided a framework to inform practitioners of the process of diagnosis, several noteworthy concerns remain [8]. One of the major criticisms is that the neurobehavioral assessments used to determine deficits do not include psycho-emotional elements that are essential indicators of psychosocial dysfunctions in FASD [2,8]. A new Canadian diagnostic guideline, soon to be released, will add recommendations for the diagnosis and significantly revise the neurobehavioural assessment components by adding psycho-emotional elements into its diagnostic criteria [9].

Most of the research on FASD has focused on behavioral problems and rehabilitative interventions for children (age below 13 years old) and adolescents. For example, Olson, Feldman, Streissguth, Sampson and Bookstein [10] found that children with FASD have impaired memory, attention, visual-spatial abilities, processing speed and declarative memory. Others found reduced cognitive flexibility such as planning, reasoning, inhibition, and working memory in children with brain scores of 2 or 3 [11,12]. Some research showed that executive function predicts levels of social skills in children with prenatal alcohol exposure [13]. Nevertheless, research in the adult population is rare.

This study is a secondary analysis of data from research on offenders suspected of FASD [14]. Participants were clients referred to a Canadian university-based service unit (UBSU) from a community organization that was part of the FASD provincial network. Investigated were the neuropsychological and functional characteristics in adults with FASD whose brain scores are within the range of 2 and 3. Two specific objectives were: 1) to compare performance between brain scores 2 and 3 on neuropsychological and functional performance; and 2) to explore factors (cognitive, psychosocial and functional) that could predict brain scores 2 and 3. To our knowledge, this is the first examination of functional and neuropsycho-logical profiles in the adult population. The identification of predictors for brain scores is expected to assist in clarifying how functional and neuropsychological deficits are related to the severity of FASD in adults.

METHOD

Participants

Sixty-eight adults with FASD were included in the study. Forty-nine of them were in contact with the justice system, and the remaining 19 were from the UBSU diagnostic services group. All participants had a diagnosis of FASD and a brain score of 2 or 3.

Procedures

The data were manually extracted from the UBSU data bank and the accompanying final reports from the FASD diagnostic formulation meeting. Any questions about the content were checked against the record in the original neuropsychological and functional occupational therapy reports. Once the data set for the present study was compiled, it was confirmed for accuracy and conceptual integrity by the contributing professionals. Participants at the time of their assessment gave written consent and agreed that their data could be used for future analysis. The study, as well as the secondary analysis of data, was approved by the Human Ethics Board of the host university and the executive committee of the community-based organization in the FASD provincial network.

Instruments - Neuropsychological Assessment

California Verbal Learning Test - 2nd Edition (CVLT-2)

The CVLT-2 is a widely used clinical instrument for assessing memory and learning abilities [15]. Participants were read a list of words and then asked to recall word lists orally. Participants completed trials 1 to 5, an interference trial, and free and cued recall trials (including short-delay recall, i.e., immediately after the interference trial, and long-delay recall, i.e., after 20 minutes of alternate task engagement). The number of words recalled in each trial was recorded.

Trail Making Test Parts A and B (TMTA / TMTB)

The Trail Making Test consists of parts A and B [16]. TMTA requires basic processes of visual attention and sequencing; TMTB demands higher-level cognitive skills such as executive control, divided attention, and cognitive flexibility [17]. Participants were required to connect numbers consecutively for TMTA and connect numbers and letters in alternating order for TMTB. Time (in seconds) taken to complete each part was recorded.

Stroop Color and Word Test (Stroop)

The Stroop test measures a person's ability to inhibit their automatic responses [18]. Participants were asked to read a list of words and state the color of the ink of specific words.The number of correct responses in each task was recorded.

Grooved Pegboard Test (GPT)

The GPT measures motor control and characteristics of movement [19]. Participants were asked to place 25 pegs, one at a time in a prescribed order. Pegs had to be placed from left to right on the pegboard when using right hand (GPT right hand), and in the opposite direction when using left hand (GPT left hand). Time (in seconds) required to complete the task in each hand (dominant or non-dominant hand) was recorded.

Finger Tapping Test (FTT)

The FTT measures the psychomotor speed of task performance [20]. Participants were asked to tap his/her index finger on a button for 10 seconds as quickly as possible. A total of 10 trials, five on the right and five on the left hand, were conducted. An average number of taps in each hand (dominant or non-dominant hand) was recorded.

Instruments - Functional Assessment

Independent Living Scales (ILS)

The ILS is a functional assessment that measures adults' competence in instrumental activities of daily living (IADL) [21,22]. There are 70 items distributed in five subscales: memory and orientation, managing money, managing home and transportation, health and safety, and social adjustment. Two factors derived from the subscales are problem-solving, and performance and information processing. The former refers to abstract reasoning and processing of factual knowledge; the latter denotes short-term memory and the ability to execute everyday tasks. The full-scale, individual subscale, and factor scores were converted to standard scores with a mean of 100 and standard deviation of 15. Scores on the ILS indicate low (55 – 85), moderate (86 – 100), and high (101 – 121) likelihood of living independently [21].

Data Analysis

Demographic information such as age, education level, employment status, and marital status was presented using descriptive statistics. Independent samples *t*-test (for age) and Chi-square (for education level, employment status, and marital status) were used to detect differences in demographic characteristics between groups. All neuropsychological and functional (ILS) test scores were converted to standard scores. For objective 1, independent samples *t*-tests were used to compare the test scores between brain scores 2 and 3. Bonferroni adjustment was applied, and statistical significance was adjusted to $p \leq 0.003$ (i.e., 0.05/19) to control for multiple comparisons. For objective 2, a hierarchical logistic regression was performed with brain score as the outcome variable and the neuropsychological test scores and ILS subscale scores (five subscales) as predictors. A forward stepwise (conditional) method was used to determine which of the predictors would contribute significantly to the brain score. Results were reported with the Cox & Snell R^2 and the Nagelkerke R^2 to reflect the percentage of variance explained by predictors. The significance of the change of each additional step was reported. All statistical analyzes were performed with IBMSPSS version 22.0. Power analysis on a stepwise logistic regression of a binary outcome variable was performed using NCSS/PASS 2002 [23]. A sample size of 68 participants would achieve 80% of power at a 0.05 significance level to detect a change in the outcome variable at an odds ratio of 4.75.

RESULTS

Sixty-eight participants, ten females and fifty-eight males, aged between 18 and 52 with a mean age of 28.5 years, were evenly distributed between brain score 2 and 3. Most of the participants were never married, had continuous employment for six weeks, and were educated at high school level. There was no significant difference in the demographic characteristics (Table **1**).

Comparison of the neuropsychological and functional test scores found that individuals with a brain score of 3 performed significantly worse than those with a brain score of 2 on the TMTB, Stroop, CVLT-2, and ILS (money management, problem-solving, and managing home and transportation subscales, and the performance and information processing factor) (Table **2**).

Results of the logistic regression showed that the money management subscale of the ILS and the long-delay cued recall subtest of the CVLT-2 significantly predicted brain score, with a Cox and Snell R^2 and Nagelkerke R^2 of 51.5% and 68.6% respectively. That is, the lower the scores on the money management subscale of the ILS and the long-delay cued recall subtest of the CVLT-2, the greater the likelihood of having a brain score of 3 instead of 2 (Table **3**).

Table 1: Descriptive Statistics of Demographic Information

	Brain score 2	Brain score 3	*p**
	34 (female = 4)	34 (female = 6)	---
Mean age (years)	30.3 ± 8.3	26.8 ± 8.5	0.095
Education level (%)			0.757
≤ Grade 8	11.8	23.5	
Grade 9	23.5	23.5	
Grade 10	20.6	26.5	
Grade 11	20.6	11.8	
Grade 12	11.8	5.9	
Post-secondary	5.9	2.9	
High school equivalent	5.9	5.9	
Employment status (%)			0.793
No employment	20.6	11.8	
< 2 weeks (limited period)	29.4	35.3	
3 – 6 weeks (steady period)	41.2	44.1	
Self-supported	8.8	8.8	
Marital status (%)			0.242
Married	16.7	9.5	
Divorced	16.7	0	
Common law	16.7	14.3	
Never married	50.0	76.2	

*significant at $p \leq 0.05$; independent samples *t*-test for age; Chi-square for all other variables.

Table 2: Mean and Standard Deviation (SD) of Standard Scores of the Neuropsychological Tests and the ILS, and the Results of Independent Samples *t*-Tests

	Brain score 2		Brain score 3		*t*	*p**
	Mean	SD	Mean	SD		
TMTA	90.29	22.15	84.32	22.51	1.103	0.274
TMTB	90.53	22.43	69.41	28.28	3.412	0.001*
Stroop	101.35	22.15	83.12	17.43	3.773	0.000*
FTT dominant hand	102.18	19.86	89.21	22.59	2.515	0.014
FTT non-dominant hand	99.24	17.25	88.53	15.51	2.691	0.009
GPT dominanthand	97.29	13.25	88.82	19.44	2.100	0.040
GPT non-dominant hand	96.59	17.99	86.44	19.91	2.205	0.031
CVLT-2						
Short delay free recall	90.18	12.16	80.38	13.90	3.092	0.003*
Short delay cued recall	92.56	12.28	76.50	15.69	4.700	<0.001*
Long delay free recall	92.24	12.38	76.09	12.45	5.361	<0.001*
Long delay cued recall	92.29	10.77	76.50	13.29	5.384	<0.001*
ILS						
Full Scale	91.44	14.14	81.85	12.69	2.943	0.004
Memory and Orientation[a]	52.29	8.74	49.50	6.78	1.473	0.146
Money Management[a]	42.88	7.04	31.59	8.83	5.835	<0.001*
Managing Home and Transportation[a]	51.15	6.22	45.24	9.11	3.125	0.003*
Health and Safety[a]	48.38	10.78	40.74	13.82	2.544	0.013
Social Adjustment[a]	35.09	12.31	36.19	12.93	-0.346	0.730
Problem Solving[b]	45.62	10.77	35.35	11.77	3.753	<0.001*
Performance and Information processing[b]	48.74	5.31	41.68	8.14	4.234	<0.001*

*Significant at $p \leq 0.003$; Adjusted *p*-level was based on Bonferroni adjustment = 0.05/19.
[a]ILS Subscale item.
[b]ILS Factor item.
All scores reported were standard scores.

Table 3: Results of Logistic Regression

Step	Cox & Snell R^2 (%)	Nagelkerke R^2 (%)	Variable	B	S.E.	*p*	Change in -2 Log Likelihood	*p* change
1	32.6	43.5	ILS MM[1]	-0.167	0.041	<0.001*	26.944	<0.001*
			Constant	6.278	1.586	<0.001*		
2	51.5	68.6	ILS MM[1]	-0.211	0.057	<0.001*	27.052	<0.001*
			CVLT-2 LC[2]	-0.149	0.041	<0.001*	25.176	<0.001*
			Constant	20.828	5.037	<0.001*		

[1]MM = Money management; [2]LC = Long delay cued recall; *significant at $p \leq 0.05$.

DISCUSSION

The present study used secondary data analysis to investigate functional and neuropsychological abilities on sixty-eight adults with FASD. Overall, individuals with a brain score of 2 performed better than individuals with a brain score of 3 on functional tasks, such as money management, managing home and transportation, problem-solving, and information processing, and on neuropsychological tests such as the CVLT-2, TMTB, and Stroop. Of all variables, the money management subscale of the ILS and the long-delay cued recall subtest of the CVLT-2 predicted the brain score. However, variables related to psychosocial and psychomotor abilities, such as the social adjustment subscale of the ILS, the FTT, and the GPT, were not significant in the between-group comparison and the regression analysis.

This study uses a social-ecological view that suggests that individuals with FASD are subject to environmental influences [24]. These influences modify individuals' behavior through reinforcing consequences, thereby developing adaptive or maladaptive responses [25]. In the context of this study, the social-ecological theory purports that FASD diagnosis and related codes influence the provision and nature of therapies available to affected individuals. Understanding the differences and similarities of performance between brain scores may facilitate the development of more sensitive therapeutic interventions, enhance the discrimination in the allocation of resources and the structuring of the environment, and maximize long-term outcomes.

Individuals with brain scores of 2 and 3 did not differ in basic cognitive processing measured by TMTA and psychomotor skills measured by FTT and GPT. These tasks required attention and visual-motor integration [26]. Comparable performance between the two groups suggests that basic cognitive processes are retained in mild to moderate severity of FASD, indicating the potential of using basic cognitive processing tasks as remedial training to strengthen functional skills. However, the range of mean scores for people in our study is still below the norms average (i.e., a score of 100). Therefore, the implication of the scores should be interpreted with caution.

Superior performance on TMTB, Stroop, and CVLT-2 (all subtests) was found for individuals with brain score 2 compared to brain score 3. Conversely, this means that higher order cognitive processes such as executive function, impulse control, and verbal learning are more significantly impaired in adults with more severe FASD. This information is valuable as secondary conditions, such as trouble with the law, inappropriate sexual behavior, and alcohol or drug problems, are the most debilitating factors that correlate with poor long-term outcomes for individuals with more severe FASD [4]. High rates of reconnecting with the justice system for "breaches" and the presence of affective disorders are also confirmed in this group [14]. It is also worth noting that the CVLT-2 (long-delay cued recall) was also a significant predictor of brain score. This suggests that those with more severe FASD have a greater degree of neurological damage that weakened the ability to learn and process information [27]. In essence, even with memory cues to help these individuals in our study, they rapidly forgot information and were not able to remember what they had previously learned. These individuals therefore likely need extensive reviews and repeat instruction of information they are to learn.

Functional assessment using the ILS showed that those with more severe FASD performed worse in general problem solving and information processing, and specifically at home, transportation, and money management items. For many individuals who have early histories of abuse, disadvantaged social and supportive learning experiences (social determinants of

health) adds support of environmental shaping of behavior [28]. In this study, the histories of adults with the more severe FASD support the view that the environment negatively impacted functional skills development. Another possibility for poor self-management in the home and society might be the lack of mentors, the misunderstanding of the presence of the brain impairments, and social stigma in the public. Dej [29] identified a developmental shift in the perception of FASD from child to adulthood. The former is perceived as a victim in need of medical attention, and the latter is seen as deviant, potentially dangerous, and hopeless. Such discrepancy can influence the way FASD adults are treated and respected in their home environments and society.

The ILS findings revealed comparable results for brain scores 2 and 3 on the memory and orientation, health and safety, and social adjustment subscales. Among these three subscales, the memory and orientation subscale showed the highest score in both brain score groups, with the score approaching the cutoff score between moderate and high functioning. These items are highly contextual with internal daily task associations, which might explain the differences in performance on the CVLT-2 items. The results suggest that embedding memory and orientation abilities within lifestyle situations are strengths for cognitive intervention in adults with FASD. Both brain score groups scored in the low functioning range on the social adjustment subscale which is sensitive to self-perception and networking. Social adjustment is related to long-term outcomes and the development of secondary conditions [4]. Social skills such as connecting with people, expressing ideas rationally, and maintaining healthy relationships with others are critical in forming positive relationships with employers and landlords and are major determinants of health in promoting and maintaining stability in one's life [30]. The recommendations for rehabilitation professionals are to address psychosocial and interpersonal factors in their treatment. This can be done by enhancing documented strengths and effective lifestyle strategies with the individual. Community caregivers and other influential people in clients' lives will benefit from ongoing education and supports. Individualized community programming is cost effective when compared to FASD's high cost to society through the inappropriate use and over-utilization of institutional resources [31]. Continued advocacy for ongoing community programming is a crucial programming component.

The brain score has been used as one of the diagnostic criteria for determining the presence and severity of FASD [2]. However, from our results, the brain scores do not seem to differentiate between adults with higher and lower levels of psychosocial abilities and do not appear to be predicted by psychological variables. Our results show that, among the many tests administered, only the CVLT-2 and the money management subscale of the ILS were significant predictors of brain score. It raises a question of whether a brain score, which is a gross measure of dysfunction, can reflect a full spectrum of deficits in adults with FASD. Our results further support the incorporation of a psychosocial element in any new diagnostic criteria of FASD. Canada's revisions are to be published and implemented in the very near future.

This study is subject to limitations. 1) The research is based on a secondary analysis of data gathered for clinical purposes. This methodology, though, is also known to provide opportunities for a deeper analysis of selected factors from the original data [32]; 2) As the analysis is limited to the data originally collected, it restricts expanding any factor considerations of more recent developments; 3) The included cohort is made up of approximately 60% of individuals of aboriginal origins and, therefore, may portray a bias in the variation of functional abilities. Environmental factors, especially social factors and adjustment, might be a dynamic of relationships with family members, support workers, and the participants' socioeconomic status [33]; and 4) Since some of the participants have received intervention services from the FASD network, it is unclear how far the previous interventions affected the scores in the study. Nevertheless, this research provides a fundamental understanding of the functional performance and its underlying neuropsychological performance in adults with FASD. Further studies are recommended to perform more in-depth quantitative and qualitative analyses.

Future studies need to consider participants with broader demographic profiles, including various ethnicities, different social backgrounds, and from multiple sites. More research is required to assess the construct of social adjustment among individuals with FASD and understand how environmental factors interact with functional performance.

CONCLUSION

The present study investigated the functional and neuropsychological abilities of adults with FASD. We found that individuals with brain score 2 performed

better than brain score 3 on nearly all neuropsychological and functional subtests except for basic cognitive processing and psychomotor tasks. Furthermore, the money management subscale of the ILS and the CVLT-2 test scores predicted the brain score. Psychosocial issues rated the lowest among all functional performance and did not differ between groups. Hence, psychosocial elements should be individually assessed to determine appropriate therapeutic interventions for maximizing functional independence supports for adults with FASD.

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