

PARENTAL RATINGS OF CHILDREN WITH FETAL ALCOHOL SPECTRUM DISORDER ON THE BEHAVIOR RATING INVENTORY OF EXECUTIVE FUNCTION (BRIEF)

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ABSTRACT

Children with FASD typically display deficits on cognitive measures of executive functioning, but the goal of this study was to examine performance of children with FASD on a behavioral measure of executive function using the Behavior Rating Inventory of Executive Function (BRIEF). Participants were 64 children (aged 5 to 16 years) with FASD. These children showed significant deficits on the BRIEF, with mean scores on all scales reaching clinical significance. A distinctive pattern was also found, with the children showing most difficulty on Inhibit, Working Memory, and Initiate scales and least difficulty on Organization of Materials. Females demonstrated substantial difficulty with inhibition and older children had more difficulty (relative to the norm) than younger children on scales measuring working memory and initiation. The BRIEF appears to be a useful tool for evaluating behavioral, social, and emotional aspects of executive functioning in children with FASD.

Fetal Alcohol Spectrum Disorder (FASD) is an umbrella term used to refer to individuals who may have physical, mental, behavioral, and learning disabilities as a result of maternal alcohol consumption.¹

Understanding the neurobehavioral deficits associated with FASD is particularly important for both diagnosis and treatment of FASD. Some of the primary neurobehavioral impairments associated with FASD involve deficits in memory, attention, visual-spatial abilities, declarative learning, planning, cognitive flexibility, processing speed² as well as language and motor delays.³ These children tend to score lower on measures of IQ and academic achievement⁴, specifically in mathematics.⁵ Executive functioning, which has been defined as higher-order psychological abilities involved in goal-oriented behavior under conscious control⁶, is a significant deficit in individuals with FASD.⁷ Executive functioning involves cognitive, emotional, and behavior components⁸ and refers to abilities such as planning, organized search, inhibition, working memory, set-shifting, strategy employment, and fluency.^{9,10} The executive function deficits in FASD have been documented on tests of cognitive flexibility, inhibition, planning and strategy use, concept formation and verbal reasoning, set shifting, working memory measures, and fluency.^{11,12,13,14} The bulk of this previous research has been on cognition-

based or 'cool' executive functioning tests. More recently researchers have started examining emotion-related or 'hot' executive function in FASD. Hot executive function is thought to be involved in response to reward and punishment stimuli and involves regulation of motivated and emotional behavior.^{7,15} Hot and cool executive functions are associated with different areas of the frontal cortex; the orbitofrontal cortex and the dorsolateral prefrontal cortex, respectively.⁷ In one study, children with FASD were found to be impaired on emotion-related or hot executive function,¹⁶ however, very few researchers have examined hot executive functioning in FASD and even less have looked at 'real-world' executive functioning *behaviors* in FASD.

One test that aims to evaluate a child's executive functioning behaviors in a real-world setting is the Behavior Rating Inventory of Executive Function, or BRIEF.¹⁷ The arrival of the BRIEF has provided a welcome alternative to prevailing neuropsychological tests that are largely modified versions of adult measures.⁸ Unlike these traditional tests, which are administered under regulated laboratory conditions, the BRIEF is a parental and teacher rating scale of a child's executive functioning behaviors in everyday situations and settings, measured across eight domains/subscales (Inhibit, Shift, Emotional

Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, and Monitor).

The BRIEF appears to uniquely evaluate a set of metacognitive, behavioral, and emotional abilities that go beyond common psychopathology and behavioral disturbances measured by other behavior rating scales.⁸ Both Baron¹⁸ and Donders⁸ attest to the BRIEF's sound psychometric properties and believe it has potential clinical utility for capturing areas of dysfunction not currently measured by alternative instruments. Some researchers have suggested that the behaviors measured on the BRIEF may be involved in the brain area that is important for emotional and behavior skills¹⁹ (i.e., hot executive functions) rather than areas more related to cognition-based or cool executive function.

Anderson et al.¹⁹ looked at the relationship between traditional cognitive executive functioning tests and the BRIEF among children with early treated Phenylketonuria (PKU), Hydrocephalus, and frontal focal lesions. All children showed significant dysfunction on the BRIEF, but the children with frontal lobe damage exhibited the greatest executive dysfunction. These results indicate that the BRIEF may tap into behavioral symptoms specific to frontal lobe systems,¹⁹ which are thought to be involved in executive functioning.^{20,21,9} Very low correlations between behavioral and cognitive instruments were also found, suggesting that cognitive and behavioral measures may be tapping into different dimensions of executive functioning. Similar to Zelazo and Muller,⁷ Anderson et al.¹⁹ suggest that cognitive aspects of executive functioning are associated with the dorsolateral prefrontal cortex, whereas the orbito-frontal regions of the cortex mediate the emotional and social aspects of executive functioning. Anderson et al.¹⁹ also suggest that the low correlations may be due to the different test administrations used.

For instance, cognitive tests are typically given in a structured laboratory setting, whereas the BRIEF involves naturalistic observations of the child's day-to-day behavior. Vriezen and Pigott²² found that among children with moderate to severe traumatic brain injury (TBI), BRIEF scores did not correlate with impairments on performance-based measures of executive functioning. More children were found to be impaired based on the BRIEF than on any other performance-based executive functioning tests indicating that children can perform relatively

well on highly structured neuropsychological tests and still have great difficulties in daily life.²² The authors suggest that the BRIEF measures a unique aspect of executive functioning.

The BRIEF has been validated as a multidimensional measure of executive functioning abilities for a mixed diagnosis clinical group, which allows for broad generalization.²³ However, Gioia, Isquith, Kenworthy and Barton²⁴ also believe that with further study of specific clinical populations the BRIEF may help achieve "disorder-specific executive profiles." The BRIEF has thus far been useful in identifying differences in disorders such as ADHD, autism spectrum disorder (ASD), reading disabilities, and TBI.²⁴ The BRIEF shows great promise in highlighting differences between different neurodevelopmental profiles without being a diagnostic tool per se.²⁴

Although there is ample research on the use of the BRIEF for children with other neurodevelopmental disorders, to our knowledge, there has only been one published study on the BRIEF and FASD.²⁵ In this previous study, children with FASD displayed considerable deficits on the BRIEF. The BRIEF was not the focus of this study (it was one of many neurobehavioral measures) and the sample size was relatively small (n=31), which did not allow for a detailed analysis of the data. Nevertheless, some interesting, albeit preliminary, patterns emerged with children showing most difficulty on the Plan/Organize and Working Memory scales on the BRIEF and least difficulty on Organization of Materials. Also, girls tended to have higher scores (indicating more difficulty relative to their peers) than boys. Age effects were not observed but this analysis may have been constrained by the small sample size over a large age range.

This research provided initial evidence regarding patterns of performance of children with FASD on the BRIEF and calls for a more detailed study specially examining the BRIEF among a larger sample of children with FASD. The goal of the current study was to conduct a more detailed examination of performance on the BRIEF among 64 children (aged 5 to 16 years) with FASD, who were a different sample of children as in Rasmussen et al.²⁵ First, we wanted to determine whether children with FASD showed deficits on the BRIEF, and more importantly, whether they showed a distinctive pattern of strengths and

weaknesses on scales of the BRIEF.²⁵ This is important for determining whether there are aspects of executive functioning that are particularly difficult for children with FASD, or conversely whether there are areas of strength within the domain of executive functioning.

Such information is important for developing instruction and remediation that can target specific areas of weakness or build upon areas of strength. Given that working memory is a significant area of difficulty for children with FASD and was one of the lowest scales on the BRIEF in Rasmussen et al.,²⁵ we hypothesized that the working memory subscale would be a significant area of deficit on the BRIEF among this sample. Second, we examined whether gender and age were related to performance on the BRIEF. We hypothesized that, similar to Rasmussen et al.,²⁵ girls, would have higher scores (indicating more difficulty) than boys, but in the current study we also examined the specific subscales of the BRIEF that are responsible for this gender effect. This comparison will help us determine which aspects of executive functioning are particularly difficult for girls with FASD. Although we did not observe age effect in Rasmussen et al.,²⁵ in a recent study¹³ we found that on a cognitive-based assessment of executive functioning, older children with FASD showed more difficulty (relative to the norm) than younger children on some tests, suggesting that some executive functioning deficits may become pronounced with age in FASD. Thus, we hypothesized that children with FASD will show more difficulty with age (relative to normative scores) on a behavioral measure of executive functioning, in accordance with the findings of Rasmussen and Bisanz¹³ with a cognition-based executive functioning test. This comparison is important for understanding the developmental trajectory of executive function deficits in FASD, which has strong implications for diagnosis and remediation.

METHOD

Participants

Parents/guardians completed the BRIEF on 64 children (37 males and 27 females) with an FASD. The mean age (in years; months) was 8;10 (range 5;1 to 16;2). All participating children had

a confirmed medical diagnosis of an alcohol-related disorder falling under the umbrella term FASD. Twenty-four of the children were diagnosed with Neurobehavioral Disorder, Alcohol Exposed; 18 with Static Encephalopathy, Alcohol Exposed; and 4 with Neurobehavioral Disorder, Alcohol Exposed with sentinel psychical findings (according to Diagnostic guide for Fetal Alcohol Spectrum Disorders: The 4-digit diagnostic code: 3rd edition, Astley, 2004).²⁶ The remaining 18 had a diagnosis of FASD, and although we acknowledge that the term 'FASD' itself is not a diagnostic term, it was the term used in these children's files and it is also the term used by the education system in the province in which these children resided. Thus some physicians use the term 'FASD' in the diagnostic reports. In order to be diagnosed with any of the classifications that fall under FASD, children would have had confirmed prenatal alcohol exposure and cognitive deficits suggesting some degree of brain dysfunction. The diagnostic information was obtained from the health records (with parental permission) of each child to confirm that a physician (or paediatrician) had diagnosed each child.

Children were recruited through medical FASD diagnostic clinic, FASD community agencies, and some data was obtained from an existing database at a hospital FASD clinic. IQ scores (WISC-III or WISC-IV) obtained from the children's medical files were available for 49 of the children, with the mean Full Scale IQ being 83.08 ($SD = 11.18$), range from 55 to 103. In terms of living arrangements (and who completed the BRIEF), 6 children were with their biological mother, 4 with their biological father, 26 with a foster parent or guardian, 18 with an adoptive parent, and 10 with a grandparent.

MATERIALS AND PROCEDURES

Behavioral Rating Inventory of Executive Function (BRIEF)

The BRIEF is an executive functioning rating scale designed for individuals between the ages of 5 and 18. The BRIEF consists of eight clinical scales: three scales (Inhibit, Shift, and Emotional Control) that comprise the Behavioral Regulation Index (BRI); and five scales (Initiate, Working Memory, Plan/Organize, Organization of

Materials, and Monitor) that comprise the Metacognition Index (MI). The Inhibit scale measures inhibitory control and the ability to control one’s behavior, and the Shift scale assesses the ability to move freely from one task or situation to another. The Emotional Control scale measures the ability to control emotional responses, whereas the Initiate Scale measures the ability to start a task and independently generate ideas, responses, or problem-solving strategies. Working Memory involves holding information in mind to complete a task, and the Plan/Organize scale measures the ability to manage current and future task demands. Lastly, the Organization of Materials scale assesses orderliness of work, storage, and play areas; and the Monitor scale assesses work-checking habits as well as behavioral monitoring. The BRI and MI combine to form the Global Executive Composite (GEC).

The BRIEF also includes two validity scales: the Inconsistency scale aims to determine if the respondent has answered in an especially conflicting manner, whereas the Negativity scale measures whether the respondent has answered in an unusually pessimistic manner. In this study the parent or guardian completed the BRIEF. The BRIEF yields T- scores which have a mean of 50 and standard deviation of 10, with higher scores indicating more difficulty. Abnormally elevated scores suggesting clinical significance are indicated by T-scores of 65 or greater which are at least 1.5 standard deviations above the mean. The BRIEF has high validity and reliability scores. For instance, the Cronbach alpha measure of internal consistency ranges from 0.80-0.98 for both clinical and normative samples on the parent version of the BRIEF. The parent form test-retest reliability correlation was $r = 0.81$ (range: 0.76-0.85) for an average two week interval (Gioia et al., 2000).

RESULTS

Children with FASD displayed profound deficits on all scales of the BRIEF as shown in Figure 1. All mean T scores were significantly higher than the normative mean of 50 (using 99% confidence intervals) and in the clinically significant range of 65 or above. The BRI ($M = 75.11, SD = 11.55$),

MI ($M = 73.69, SD = 9.44$), and GEC ($M = 76.10, SD = 11.16$) index scales also had means in the clinically significant range. Another way to capture difficulty on the BRIEF is to look at the percentage of children whose performance was in the clinically significant range.

TABLE 1 Percentage of Children with BRIEF Scores in the Clinically Significant Range

BRIEF Scale	% of children
Inhibit	75.0
Shift	71.7
Emotional Control	65.6
Behavioral Regulation Index: BRI	83.9
Initiate	79.4
Working Memory	78.1
Plan/Organize	59.6
Organization of Materials	66.1
Monitor	68.9
Metacognition Index: MI	84.5
Global Executive Composite: GEC	86.5

As evident in Table 1, the majority of children had BRIEF scores in the clinically significant range, with the highest number of children showing deficits on Initiate, Working Memory, and Inhibit scales and the lowest number on Organization of Materials. As evident in Figure 1 and Table 1, there appears to be a difference across scales of the BRIEF.

Next we conducted a repeated measures ANOVA to determine whether performance differed among the scales on the BRIEF. Performance varied considerably across scales of the BRIEF, $F(7, 392) = 7.25, p < .00$, with children showing the greatest difficulty on the Inhibit, Working Memory, and Initiate scales and the best performance on Organization of Materials.

FIG. 1 Performance of Children with FASD on Scales of the BRIEF

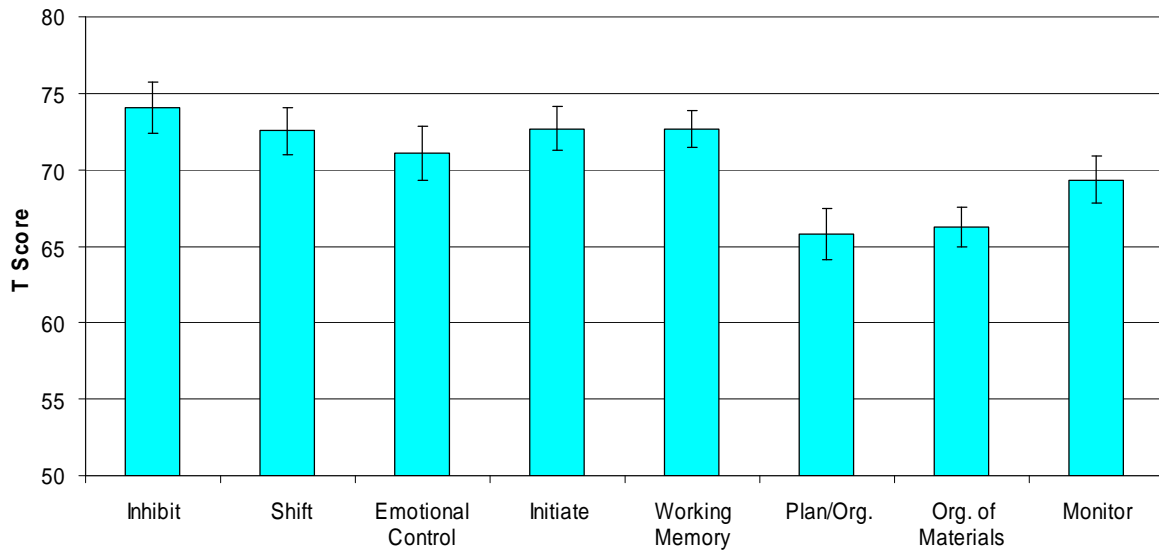
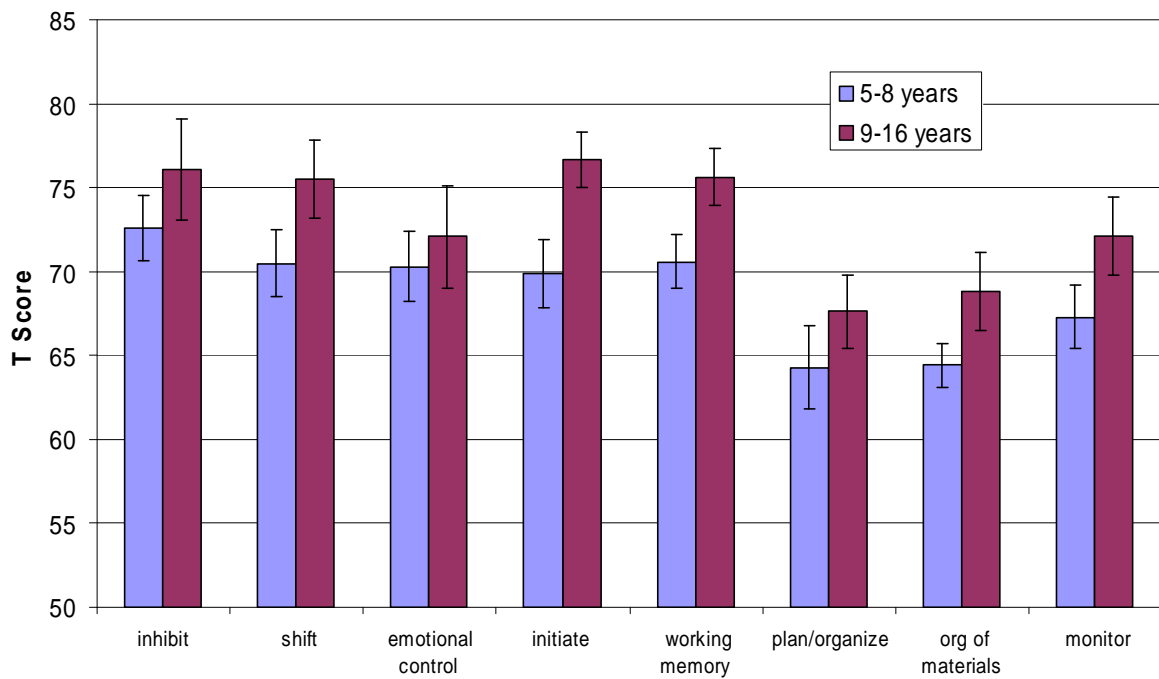


FIG. 2 Performance on the BRIEF as a Function of Age Group



Note: The bars represent standard errors

To test for gender effects, T scores of males and females were compared on the scales of the BRIEF. Females score significantly higher ($M = 78.81$, $SD = 12.55$) than males ($M = 70.54$, $SD = 13.18$) on the Inhibit scale, $F(1, 63) = 6.4$, $p < .05$, as well as on the BRI, $F(1, 61) = 7.25$, $p < .01$. No other gender differences were significant.

To examine age effects children were divided into a younger (5-8 years, $n=38$) and older (9-16 years, $n=26$) age group. The age range in the older group is larger than that of the younger group because we had more children in the younger half of our sample. This data is presented in Figure 2. On average older children tended to have higher scores (indicating more deficits) than younger children, but this difference was only significant on the Initiate, $F(1, 62) = 6.02$, $p < .05$, and Working Memory scales, $F(1, 63) = 4.35$, $p < .05$.

DISCUSSION

Children with FASD typically display deficits on cognitive measures of executive functioning, but the goal of this study was to examine performance of children with FASD on a behavioral measure of executive function using the BRIEF (based on parental ratings).

We wanted to determine whether children with FASD showed a distinctive pattern of strengths and weaknesses on the BRIEF and also whether gender and age related to performance on the BRIEF. Children with FASD demonstrated profound executive functioning deficits on the BRIEF (based on parental ratings). For all scales T scores differed significantly from the normative mean of 50 and all mean T scores were in the clinically significant range. These results cannot be attributed to a small number of children driving up scale means because the majority of the children showed this pattern of highly elevated T scores. When compared to other diagnostic groups (ASD, ADHD, TBI, PKU, hydrocephalus, and frontal focal lesion) also evaluated on the BRIEF, children with FASD show considerably higher scores on almost all scales.^{19,24} Thus, children with FASD display significant deficits on the BRIEF and these deficits may be more severe than in other neurodevelopmental disorders. A distinctive pattern of strengths and weaknesses also emerged, with scores being poorest on the Inhibit, Working

Memory, and Initiate and scales and best on Organization of Materials. Organization of Materials appears to be an area of relative strength for children with FASD (based on parental ratings).

However, this result may be partially confounded with the tendency of parents of children with FASD to maintain highly structured living and play areas, thus further research is warranted. Parental ratings indicate that inhibition, working memory, and initiation appear to be significant areas of weakness for children with FASD. Inhibition deficits have also been found on cognitive executive tests among individuals with FASD^{11,12,13} supporting the notion that inhibition is particular area of difficulty. The poor working memory scores on the BRIEF are consistent with the findings of Rasmussen et al.²⁵ and with previous research indicating working memory is a key deficit in children with FASD.^{13,6}

This study further demonstrates that children with FASD have particular difficulty on initiation of tasks and generating ideas, responses, and problem solving strategies. Such significant deficits in the ability to hold information in memory, control one's behavior, and initiate effective problem-solving strategies would present many daily challenges and difficulties adapting to society for children with FASD and thus represent some of the real-life consequences of prenatal alcohol exposure. Children with frontal focal lesions display a similar, though less severe, pattern of deficits, (poorest on Working Memory and highest on Organization of Materials)¹⁹ indicating a strong connection between specific/localized frontal impairment and FASD. Prenatal alcohol exposure has been implicated as a negative factor in the development of the frontal cortex^{27,28,29} indicating that frontal lobe impairments may account for similar profiles of executive functioning deficits in individuals with frontal lobe lesions and those with FASD. Females had higher T scores (indicating more difficulty) than males on the Inhibit scale and BRI of the BRIEF. It is important to note that higher scores of females over males does not mean that females with FASD have more severe inhibition deficits than males with FASD, but rather that as compared to other same-aged girls in the norm sample of the BRIEF, females with FASD have higher scores than males (as compared to same aged boys in the

norm sample of the BRIEF). The inhibition deficits in females with FASD are substantial (mean was over 2 standard deviations above the mean) indicating that special attention needs to be focused on strategies and resources to deal with and ameliorate these difficulties.

Overall, older children showed more difficulty (relative to the normative data of the BRIEF) than younger children on all scales of the BRIEF and this difference was significant on the Initiate and Working Memory scales. It appears that performance on some emotional and social executive functions are worse (relative to the norm) among older children with FASD. Perhaps adolescence places an extra demand on executive functions and in particular on working memory and initiation, which could result in more pronounced deficits in these areas during adolescence. These age effects must be interpreted with caution, as this was not a longitudinal study with the same children followed over time. Therefore it is possible the poorer scores among the older group could be because these children were diagnosed later, received fewer services, or had more severe brain dysfunction, all of which could lower their performance on the BRIEF. Longitudinal research is necessary to determine the developmental trajectory of executive functions in children with FASD.

Another factor worthy of consideration is the high number of children in this sample not living in their biological homes, but with foster or adoptive parents. Adoptive or foster parent homes are extremely common in the general population of children with FASD so generalizability is not an issue, however living situations may affect performance levels. Children who have an unstable home life, are frequently moved to different homes, or are lacking a permanent caregiver may be at increased risk for behavioral and emotional problems, areas that are tapped by the BRIEF. Lastly, the potential for rater bias should be mentioned, as certain caregivers may view their child's performance in an excessively negative light. However we do not believe rater bias to be at play in this study, as not only does the BRIEF itself include a negativity scale, but also the vast majority of children were given high scores (indicating deficits) by their caregivers, offering evidence that our results are not due to a small number of biased raters. This study has

provided new evidence that executive functioning deficits in FASD are not limited to only cognitive aspects. These children also show considerable deficits on parental ratings of social, emotional, and behavioral executive functioning and thus instruction and intervention should focus on both the cognitive and behavioral aspects of executive functioning. The distinctive pattern of performance on the BRIEF has significant implications for both diagnosis and treatment of children with FASD. Identifying weaknesses in areas such as inhibition, working memory, and initiation may help the diagnostic process by indicating key areas that might warrant further investigation and also informs intervention by identifying specific deficits that can be targeted with instruction and treatment. The substantial inhibition deficits faced by females with FASD has implications for tailoring some intervention techniques and resources specifically for females. Finally, the finding of lower scores (relative to the normative data on the BRIEF) with age may have implications for early intervention with children with FASD in hopes that if intervention is started early it may help reduce some of the gaps in functioning with age. Extra resources and support are needed during adolescence to address the significant deficits in social, emotional, and behavioral aspects of executive functioning to improve functioning and adapting in society.

In conclusion, the BRIEF appears to be a very important tool for documenting 'real-life' executive functioning behaviors in children and provides useful clinical data on the complexity of difficulties faced by children with FASD that may not be obtained from traditional cognitive tests.

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