Signals of CNS damage in the discourse behavior of school-aged children with prenatal alcohol exposure during a narrative generation task.

Rationale: Research on narrative performance in school-aged children indicates that impaired populations perform differently than typically developing (TD) children on measures of productivity and cohesion [e.g., 1, 2]. If these measures are to be turned into clinically useful tools, it is imperative that performance standards which discriminate between typically developing and impaired populations be established for particular narrative tasks. The current research examines potential performance standards for a common narrative task: story generation using *Frog, Where Are You[3]*.

Primary Questions:
1) Does performance on *Frog* narratives (as measured by a TREIN analysis [4]) become more impaired with increasing evidence of structural CNS abnormality (as measured by the FASD CNS Rank 1-4 [5])?
2) Do significant differences (i.e., > 2 SD) in narrative performance discriminate between children with FASD that have *Static Encephalopathy* and their TD peers in this sample of 32 children?

Secondary Question: What is the relationship between severity of FAS diagnosis and narrative performance in the 32 school-age children in this sample when additional narrative measures are also included in analysis?

Subjects: 32 school-age children (8;5 to 11;7 years):

16 diagnosed with prenatal alcohol exposure (PAE):

- PAE was assessed by an experienced interdisciplinary team using the *4-Digit Diagnostic Code* (Astley, 2004).
- Team ranked cases for evidence of CNS damage [5]:

1= unlikely	(n= 0)
2 = possible	(n=8)
3 = probable	(n=4)
4 = definite	(n=4)

16 typically developing (TD) matched on age:

- Considered CNS-rank 1 (i.e., unlikely CNS damage).
- School records screened
- No interdisciplinary assessment

Methods:
To answer the primary questions, children were grouped into four CNS Ranks defined using criteria from the FASD *4-Digit Diagnostic Code* [5]. FASD CNS Ranks are based on evidence of underlying structural CNS abnormality found during a comprehensive interdisciplinary diagnostic evaluation. CNS Rank 4 indicates “definite” structural CNS abnormality based on direct structural/neurological evidence. CNS Rank 3 is assigned when functional evidence indicates “significant” impairment in three or more domains of brain function (i.e., a performance deficit equivalent to 2 SD or more from the mean on a standardized test). A child with CNS Rank of 3 or 4 is diagnosed with “*Static Encephalopathy*.” CNS Rank 2 is assigned when the child exhibits at least mild to moderate delay or impairment in some domain of functioning, but does not qualify for a

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CNS Rank of 3. CNS Rank 1 indicates that the individual does not meet criteria for Ranks 2 through 4. Although this system is an indirect indicator or CNS impairment, Astley et al. [6] confirms that as CNS Rank moves from 1 to 4 there is increasing probability of underlying structural CNS abnormality. All children in the TD group were assigned CNS Rank 1, while children in the FASD group were CNS RANK 2-4.

Narratives came from two databases [7, 8].

- Elicited using *Frog Where Are You*.
- Child previewed storybook
- Asked to tell "best story possible," using the picture book as a visual prompt
- Examiners sat with storybook out of their view.

Narratives recorded and transcribed using SALT [9].

- 26 standard SALT measures generated in three domains
  - *Productivity*: NDW, MLU, Total Words/Utterances, etc.
  - *Mazes*: Total Maze Words, Mazes per Utterance, etc.
  - *Standard Words*: Personal Pronouns, conjunctions, quantifiers, etc.
- 9 Cohesion measures generated using TREIN protocols [4].
  - Coding conducted blind to storyteller characteristics.
  - Interrater reliability (25% of transcripts); Kappa = 0.90

Analysis:

- Correlations for each measure to CNS-rank calculated (Kendall's tau b; $p<0.05$).
- All TREIN measures as well as SALT measures that were significantly correlated were subject to One-way ANOVA with CNS-rank as the grouping factor.
- Sensitivity/specificity for classifying "probable/definite" CNS damage at 1.25, 1.5, and 2.0 standard deviations from the mean of the TD group were calculated for each measure.

Results:

Primary Questions

One-way ANOVA

- Significant effects of CNS-rank on
  - INI (F=5.9, p= 0.003)
  - rNRE (F= 15.8, p= 0.0001)
- Pairwise Comparisons (Scheffe, $p<0.05$)
  - CNS-rank 4 produced fewer INI with higher rNRE than ranks 1 or 2.
  - CNS-rank 3 had higher rNRE than rank 1.

Mean (SD) & planned contrasts (Scheffe's Multiple Comparison Test) by CNS Rank for INI and rNRE

	INI		**rNRE**	
CNS RANK	**mean** (SD)	**Significant Contrasts**	**rNRE** (SD)	**Significant Contrasts**
1-*unlikely (n=16)*	16.7 (3.6)	1>4 (p=0.003)	1.57% (0.7)	1<4 (p<0.0001); 1<3 (p=0.006)
2-*possible (n=8)*	15.4 (3.5)	2>4 (p=0.03)	2.59% (0.9)	2<4 (p=0.01)
3-*probable (n=4)*	14.0 (2.4)	ns	3.40% (1.2)	3>1 (p=0.006)
4-*definite (n=4)*	8.8 (2.8)	4<2(p=0.03);4<1(p=0.003)	4.48% (0.9)	4>1 (p<0.0001); 4>2 (p=0.01)

Sensitivity/Specificity
Only rNRE had clinically important ability [10] to classify cases with "probable/definite CNS damage"

- 2 sd cut-off
  - sensitivity 88%,
  - specificity 92%
    - ➢ False-positives CNS-rank 2
- 1.5sd and 1.25 sd
  - sensitivity 88%
  - specificity (79% & 71% respectively)
    - ➢ False-positives included TD children.

Secondary Question:

Significant Correlations to CNS-rank (Kendall's tau b, $p < 0.05$)

Measure	**Correlation**	**p-value**
Rate of Nominal Reference Error (rNRE)	0.63	<0.0001
Indefinite nominal introduction (INI)	-0.40	=0.006
Cohesive pronoun ties	-0.37	=0.008
Total Utterances	0.37	=0.010
Number of Different Words	-0.31	=0.025
Number of Words	-0.31	=0.028
Personal pronouns	-0.28	=0.043

Children diagnosed with FAS had the highest rNRE (4.36%, SD 0.8), children with TD had the lowest (1.57%, SD 0.7), and children with FASD without FAS facial features (other FASD) fell in between (2.77%, SD 0.9) with little overlap between groups ($F = 23.0$, $p < 0.0001$; Scheffe's contrasts: FAS>TD, $p<0.0001$, FAS>other FASD, $p=0.005$, other FASD>TD, $p=0.004$).

All 5 children with FAS and an additional 4 with other FASD had rNRE more than 2 SD from the mean of the TD group (1.57%, SD 0.7).

The group "*Static Encephalopathy*" had significantly smaller NDW (mean 92.1, SD 25.8) than the group "*CNS Rank 1 or 2*" (mean 125.8, SD 38.6; t-test -2.29, $p=0.03$). They also produced fewer cohesive pronoun ties (PNTIE=16, SD 8.6) than the "*CNS Rank 1 or 2*" group (mean 24.4, SD 11.6; t-test -2.18, $p=0.04$).

No cases produced NDW or PNTIE more than 2 SD below the mean of the TD group (NDW=130, SD 43.1; PNTIE=27.4, SD 12).

No other measures analyzed produced important contrast between groups.

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Discussion: Using a narrative task, story generation using *Frog Where Are You*, we found discourse behaviors significantly correlated with CNS damage. One, rNRE, provided a clinically significant ability to classify cases into those with *FASD and Static Encephalopathy*, indicating that CNS damage may leave identifiable signals in performance on this task. These results support further research and development of this task for use in diagnostic contexts involving school-aged children with PAE. Results support research in other complex clinical populations (e.g., ADHD) to determine the utility of narrative analysis for differential diagnosis of underlying CNS impairments.

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