OPTIMIZING A CLINICAL LANGUAGE **MEASURE FOR USE IN IDENTIFYING SIGNIFICANT** NEURODEVELOPMENTAL IMPAIRMENT IN DIAGNOSIS OF FETAL ALCOHOL SPECTRUM DISORDERS (FASD)

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FETAL ALCOHOL SYNDROME DIAGNOSTIC AND PREVENTION NETWORK

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EVIDENCE OF NEURODEVELOPMENTAL IMPAIRMENT CONTRIBUTES TO THE DIAGNOSIS OF FASD USING ANY OF THE CURRENT DIAGNOSTIC SYSTEMS

PUTS THE "D" IN FASD

DIFFERENT SYSTEMS MAKE DIFFERENT CHOICES,

BUT SEVERITY MATTERS IN ALL DIAGNOSTIC SYSTEMS.

CRITERIA ARE CHOSEN TO AVOID FALSE POSITIVE DIAGNOSIS: SPECIFICITY IS CRITICAL IN DIAGNOSTIC CONTEXTS

CRITERIA FOR University of Washington NEURODEVELOPMENTAL IMPAIRMENT O

4-DIGIT CODE

SIGNIFICANT IMPAIRMENT

2 STANDARD DEVIATIONS FROM THE MEAN OR MORE

3 OR MORE DOMAINS

DIAGNOSES: STATIC ENCEPHALOPATHY, FAS, & PFAS

MILD TO MODERATE

PERFORMANCE OUTSIDE AVERAGE RANGE IN AT LEAST ONE DOMAIN

DIAGNOSIS: NEUROBEHAVIORAL DISORDER

CANADIAN GUIDELINES

SIGNIFICANT IMPAIRMENT

2 STANDARD DEVIATIONS FROM THE MEAN OR MORE

3 OR MORE DOMAINS

DIAGNOSES: FASD WITH OR FASD WITHOUT SENTINEL FACIAL FEATURES

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SIGNIFICANT IMPAIRMENT

1.5 OR MORE STANDARD DEVIATIONS FROM THE MEAN ON SPECIFIED COGNITIVE OR NEUROBEHAVIORAL MEASURES

DIAGNOSES: ARND, FAS, & PARTIAL FAS

"ADAPTIVE" SKILLS DEFICITS

CAN "ASSIST" IN DIAGNOSIS, BUT CAN'T STAND ALONE AS EVIDENCE OF NEURODEVELOPMENTAL IMPAIRMENT

LANGUAGE AS POSSIBLE DOMAIN OF NEURODEVELOPMENTAL IMPAIRMENT

4-DIGIT CODE

CANADIAN GUIDELINES

LANGUAGE IS ONE EXAMPLE AMONG MANY

"MULTIPLE DOMAINS THAT MAY INCLUDE, BUT ARE NOT LIMITED TO, EXECUTIVE FUNCTION, MEMORY, COGNITION, SOCIAL/ADAPTIVE SKILLS, ACADEMIC ACHIEVEMENT, LANGUAGE, MOTOR, ATTENTION OR ACTIVITY LEVEL." NEURODEVELOPMENTAL DOMAINS: MOTOR SKILLS; NEUROANATOMY/NEUROPHYSIOLOGY ; COGNITION; **LANGUAGE**; ACADEMIC ACHIEVEMENT; MEMORY; ATTENTION; EXECUTIVE FUNCTION, INCLUDING IMPULSE CONTROL AND HYPERACTIVITY; AFFECT REGULATION; AND ADAPTIVE BEHAVIOUR, SOCIAL SKILLS OR <u>SOCIAL COMMUNICATION</u>.

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VERBAL IQ IS EXPLICITLY SANCTIONED AS PROVIDING EVIDENCE OF COGNITIVE IMPAIRMENT.

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"LANGUAGE" NOT EXPLICITLY MENTIONED AS ONE OF THE NEUROBEHAVIORAL DOMAINS SANCTIONED FOR DIAGNOSTIC PURPOSES

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- <u>CAN "ASSIST WITH DIAGNOSIS"</u> NOT DIAGNOSTIC ON THEIR OWN
 - DELAYED AUDITORY PROCESSING: CENTRAL AUDITORY DELAY
 - SPEECH AND LANGUAGE DEFICITS: DIFFICULTIES WITH LANGUAGE ACQUISITION
 - RECEPTIVE, EXPRESSIVE LANGUAGE DELAYS
 - DEFICITS IN WORD PROCESSING/WORD RECOGNITION
 - ARTICULATION ERRORS
 - DEFICITS IN HIGHER ORDER LANGUAGE PROCESSING
 - DEFICITS IN SOCIAL PRAGMATICS/PERCEPTION;
 - LANGUAGE PROCESSING: IMPAIRED GESTURAL COMMUNICATION
- "DEFICIT" AND "DELAY" ARE NOT BENCHMARKED TO SPECIFIC PERFORMANCE CRITERIA.
- <u>SEE TABLE 3, PAGE 10-11, HOYME ET AL 2016</u>



FOR OUR PURPOSES TODAY

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- TWO OUT OF THREE DIAGNOSTIC SYSTEMS DIRECTLY INCORPORATE EVIDENCE OF LANGUAGE IMPAIRMENT INTO THE DIAGNOSTIC PROCESS.
- IN THE OTHER, EVIDENCE OF LANGUAGE IMPAIRMENT CAN ASSIST IN DIAGNOSTIC DECISION MAKING AS AN EXAMPLE OF "ADAPTIVE SKILLS" DEFICITS.
- SO, EVALUATION OF LANGUAGE CAPACITIES IS AN IMPORTANT COMPONENT IN THE DIAGNOSTIC PROCESS USED TO DIAGNOSE FASD IN ALL SYSTEMS.

THAT MEANS WE NEED VALIDATED TOOLS TO IDENTIFY LANGUAGE IMPAIRMENT IN FASD.

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LANGUAGE IMPAIRMENT: HOW COMMON IN FASD?

BASIC LANGUAGE

ABOUT 40%: OF CHILDREN WITH FASD PERFORM 2 SD OR MORE FROM THE MEAN ON TRADITIONAL LANGUAGE TESTS (COGGINS, TIMLER, & OLSWANG, 2007)

EXPRESSIVELY, THESE TESTS TEND TO REQUIRE **RESPONSES AT OR BELOW THE LEVEL OF THE SENTENCE**. MORE COMPLEX, LATER DEVELOPING SKILLS

MORE THAN 60%: DIFFICULTY PRODUCING AGE-APPROPRIATE NARRATIVES BASED ON INFORMED, BUT SUBJECTIVE CLINICAL JUDGMENT

(COGGINS, TIMLER, & OLSWANG, 2007)

BUT THESE MEASURES DON'T MEET DIAGNOSTIC STANDARDS.

NEED: CAPTURE LATER DEVELOPING SKILLS WITH TOOLS VALIDATED FOR DIAGNOSTIC PURPOSES

IF WE MISS IMPORTANT IMPAIRMENT IN DIAGNOSTIC CONTEXTS, IT CAN LEAD TO FALSE NEGATIVE DIAGNOSES.

SENSITIVITY MATTERS, TOO.

CAPTURING IMPAIRMENT IN LATER DEVELOPING LANGUAGE SKILLS

TASK NEEDS TO BE COMPLEX

TASK NEEDS TO BE CLINICALLY EFFICIENT TASK NEEDS TO BE VALID FOR IDENTIFYING CNS IMPAIRMENT IN FASD

RESPONSES THAT ARE MORE COMPLEX THAN A SINGLE SENTENCE. LANGUAGE ANALYSIS CAN BE LABOR INTENSIVE, SO THE EFFORT REQUIRED TO EXTRACT THE INFORMATION IS AN IMPORTANT ELEMENT TO CONSIDER

THIS MEANS IT HAS BEEN EMPIRICALLY SHOWN TO BE SENSITIVE TO CNS IMPAIRMENT WHILE ALSO AVOIDING EXCESSIVE FALSE POSITIVE DIAGNOSES.

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ELICITING COMPLEX LANGUAGE FROM SCHOOL-AGED CHILDREN IN A CLINICAL SETTING

"FROG WHERE ARE YOU?" BY MERCER MAYER,

AN UNDERGROUND CLASSIC OF CHILD LANGUAGE RESEARCH

THE TASK

LOOK THROUGH THIS BOOK TO LEARN THE
 STORY...

THEN USE THE PICTURES TO TELL ME THE BEST VERSION OF THE STORY THAT YOU CAN...

Importantly, The listener cannot see the pictures.



LOTS OF BEHAVIORS CAN BE TRACKED AS THE STORY UNFOLDS

ELEMENTS THAT IMPROVE QUALITY:

- A BETTER PLOT/ STORY GRAMMAR*
- MORE EVALUATIVE LANGUAGE*
 - OPINIONS/PERSONAL REACTIONS
- MORE INTERESTING/VARIED VOCABULARY**
- MORE COMPLEX GRAMMATICAL STRUCTURES**
 - LONGER UTTERANCES (MLU)
 - MORE SUBORDINATION
- MORE SKILLED STORYTELLERS INCLUDE MORE OF
 THESE ELEMENTS.

*Skilled and labor intensive analysis required ** Quick and easy analysis captures these behaviors

- ERRORS THAT DEGRADE FIDELITY:
 - GRAMMATICAL ERRORS**
 - ORGANIZATIONAL ERRORS
 - MICROSTRUCTURAL "COHESION"
 - MACROSTRUCTURAL "COHERENCE"*
 - WE WILL FOCUS ON ERRORS OF GRAMMAR AND COHESION THAT O ARE EASY TO IDENTIFY



EXAMPLES OF ERRORS

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GRAMMATICAL ERRORS

"THE BOY'S LOOKING FOR <u>HIM</u> FROG"

"THE TWO FROGS IS SITTING"

"HE <u>RUNNED</u> THERE"

"WHERE <u>HE DID</u>GO?"

"FROG __ GONE"

ERRORS OF REFERENCE

"THERE WERE <u>TWO FROGS</u> ON THE LOG, AND <u>THE FROG</u> WAS HIS."

THE BOY AND THE DOG WERE LOOKING IN THE FOREST, AND <u>HE</u> STARTED CHASING THE BEES"

Errors of cohesion

ERRORS OF INTRODUCTION

ON FIRST MENTION:

"HE HAD A FROG"

"IT PICKED HIM UP"

"THE FROG ESCAPED FROM HIS JAR"

IN ENGLISH, THE FORMS IN BLUE ASSUME THAT THE OBJECT HAS BEEN PREVIOUSLY MENTIONED

CAN WE TELL WHO HAS A NEURODEVELOPMENTAL IMPAIRMENT BY COUNTING BEHAVIORS DURING NARRATIVE?

- IF THIS IS A VALID APPROACH,
 - THEN A TALLY OF ERRORS AND/OR POSITIVE BEHAVIORS SHOULD HELP US DISCRIMINATE BETWEEN
 - <u>STORYTELLERS WITH NEURODEVELOPMENTAL IMPAIRMENT</u>
 - MORE ERRORS AS A GROUP
 - FEWER POSITIVE BEHAVIORS
 - AND THOSE WITHOUT NEURODEVELOPMENTAL IMPAIRMENT
 - FEWER ERRORS AS A GROUP
 - MORE POSITIVE BEHAVIORS
 - IDEALLY -VERY LITTLE OVERLAP IN THE DISTRIBUTION OF BEHAVIORS.
 - TO AVOID FALSE POSITIVES AND FALSE NEGATIVES.

HYPOTHESIS: ERRORS WILL BE MORE CLINICALLY USEFUL THAN QUALITY MEASURES

• MEASURES OF QUALITY INVOLVE MOTIVATED CHOICES.

• A SKILLED STORYTELLER MAY CHOOSE NOT TO INCLUDE QUALITY ELEMENTS THEY ARE CAPABLE OF PRODUCING.

• ERROR MEASURES ARE UNLIKELY TO INVOLVE MOTIVATED CHOICES

- SKILLED STORYTELLERS ARE UNLIKELY TO CHOOSE TO MAKE ERRORS.
- ERRORS, INSTEAD, INDICATE THAT THE TASK IS CURRENTLY EXCEEDING AVAILABLE CAPACITY.

PARTICIPANTS: 78 CHILDREN AGES 9 TO 12 YEARS



- DIAGNOSES:
 - NEUROBEHAVIORAL DISORDER,
 - STATIC ENCEPHALOPATHY,
 - FAS, OR PARTIAL FAS.
- INTERDISCIPLINARY ASSESSMENT
 - <u>13 (29.5%) WITH CLINICALLY</u> <u>IDENTIFIED "SIGNIFICANT"</u> <u>LANGUAGE IMPAIRMENT</u>
 - BASED ON 2 SD DEFICIT ON
 STANDARDIZED TESTING
 - SENTENCE-LEVEL TASKS

- CONTROL SAMPLE OF 34 CHILDREN
 SCREENED TO ELIMINATE THOSE
 WITH DEVELOPMENTAL CONCERNS
 - NO ACADEMIC, SOCIAL, COGNITIVE, OR BEHAVIOR PROBLEMS
 - ALCOHOL EXPOSURE UNKNOWN,
 - BUT NOT SUSPECTED

All participated in our narrative task during their clinical visit or as part of previous research

PREPARING THE NARRATIVES

• TWO TRANSCRIBERS, **BLIND TO PURPOSE OF THE STUDY** FOLLOWING PROTOCOLS FOR SYSTEMATIC ANALYSIS OF LANGUAGE TRANSCRIPTS (SALT) SOFTWARE.

- ALL TRANSCRIPTS COMPARED AND DISAGREEMENTS WERE RESOLVED THROUGH CONSENSUS TO ENSURE TRANSCRIPTS CAPTURED WHAT WAS SAID ACCURATELY.
- NEXT, <u>SEGMENTATION AND GRAMMATICAL ERROR CODING</u> BY TRAINED CODERS FOLLOWING SALT CODING PROTOCOLS, **BLIND TO DIAGNOSIS**, **AGE**, **GENDER**
 - 20% RANDOMLY SELECTED AND RECODED INTERRATER AGREEMENT GREATER THAN 90%
- COHESION ERRORS IDENTIFIED WITH TALLYING NARRATIVE REFERENCE ERRORS IN NARRATIVE (TREIN) BY TRAINED CODER BLIND TO DIAGNOSIS, AGE, GENDER.
 - 20% RANDOMLY SELECTED RECODED, 95% OVERALL POINT-BY-POINT AGREEMENT

DISCRIMINATING BETWEEN GROUPS? University of Washington QUALITY MEASURES







Children without CNS impairment maintain 95% accuracy or greater.

Children with CNS impairment make more errors as a group.

This suggests potential as a behavioral marker of impaired capacity

100-Specificity

Potential as a behavioral marker of impairment <u>AUCroc:</u> The more of the top left of the square filled, the better.

Larger number indicates more potential 1.0 = perfect >0.9 = good potential

Total Errors AUCroc 0.905

significantly better than <u>MLUm</u> (p = 0.0012) <u>NDW</u> (p = 0.0001)

<u>HYPOTHESIS:</u> ERRORS WILL BE MORE CLINICALLY USEFUL THAN QUALITY MEASURES

• MEASURES OF QUALITY INVOLVE MOTIVATED CHOICES.

RESULTS IN SIGNIFICANT OVERLAP BETWEEN DISTRIBUTIONS OF CHILDREN
 WITH AND WITHOUT IMPAIRMENT

• ERROR MEASURES ARE UNLIKELY TO INVOLVE MOTIVATED CHOICES

SHOWS SIGNIFICANT POTENTIAL TO BE USED AS A DIAGNOSTIC TOOL

AUCROC ONLY GAUGES POTENTIAL

• TO DEMONSTRATE CLINICAL UTILITY:

- IDENTIFY PERFORMANCE AT RELEVANT CUT-POINTS
 - 4-DIGIT CODE & CANADIAN SYSTEM "SIGNIFICANT IMPAIRMENT"
 - 2 SD FROM THE MEAN OF CONTROLS
 - IOM GUIDELINES : EXPLORE TO FIND VALUES THAT CAN "ASSIST" IN DIAGNOSIS.
 - 1.5 SD FROM MEAN OF CONTROLS = THE NEUROBEHAVIORAL CUT-OFF

- THE MOST ACCURATE CUT-OFF
- THE HIGHEST SPECIFICITY

Total Errors Divided by Length

Total Errors Divided by Length

PREDICT CNS IMPAIRMENT WITH TOTAL ERRORS

• +2 SD ABOVE THE MEAN OF CONTROL CHILDREN

- SENSITIVITY OF 52% (23 TRUE POSITIVES OUT OF 44 CASES)
- SPECIFICITY OF 94% (ONLY 2 FALSE POSITIVES OUT OF 34 CASES)

+1.5 SD CUT-POINT – IOM

- SENSITIVITY TO 66% (29 TRUE POSITIVES OUT OF 44 CASES)
- REDUCES SPECIFICITY TO 91 % (3 FALSE POSITIVES OUT OF 34 CASES)

• ACCURACY OPTIMIZED THROUGH EMPIRICAL METHODS,

- AT +1.0016 SD ABOVE THE MEAN OVERALL ACCURACY WAS 83%
- SENSITIVITY OF 82% (36 TRUE POSITIVES OUT OF 44 CASES)
- SPECIFICITY OF 85% (5 FALSE POSITIVES OUT OF 34 CASES)
- MORE THAN DOUBLES THE BURDEN OF FALSE POSITIVES COMPARED TO A +2 SD CUT POINT

Recall that 13 cases of severe language impairment were identified clinically using standard tools at 2 SD cut-off

CONCLUSIONS

 • TALLYING ERRORS DURING THIS NARRATIVE TASK APPEARS TO HAVE SUBSTANTIAL POTENTIAL TO IDENTIFY IMPAIRMENT IN FASD

<u>CLINICAL UTILITY COMES FROM</u>

- REASONABLE ACCURACY, SENSITIVITY, AND SPECIFICITY AT CUT-POINTS IDENTIFIED BY THE MAJOR DIAGNOSTIC SYSTEMS
- A STRONG ABILITY TO "ASSIST" IN DIAGNOSIS BY CAPTURING SUBTLE NEURODEVELOPMENTAL IMPAIRMENTS THAT MIGHT BE MISSED BY LESS COMPLEX TASKS – IMPROVES OVER CURRENT PRACTICE WHEN ADDED TO ASSESSMENT.

MOVING FORWARD

- REPLICATE THESE RESULTS IN A LARGER SAMPLE
 - CURRENT WORKING TO DO THIS WITH 250 CHILDREN WITH FASD.

• REFINE THE SYSTEM TO MAKE IT MORE CLINICALLY EFFICIENT

• WORK WITH CODING AUDIO DIRECTLY, RATHER THAN RELYING ON WRITTEN TRANSCRIPTS

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CREATE VALID NORMATIVE DATABASE

• TO ESTABLISH EXPECTED PERFORMANCE ACROSS A RELEVANT RANGE OF AGES.

MOVING FORWARD

- WHY DO CHILDREN WITH FASD MAKE MORE ERRORS?
- WHAT ARE THE UNDERLYING NEURAL MECHANISMS THAT SUPPORT FIDELITY IN NARRATIVES, AND HOW DOES PRENATAL ALCOHOL EXPOSURE DAMAGE THOSE SYSTEMS.
- SPECULATION:
 - EXECUTIVE FUNCTION/COGNITIVE CONTROL SYSTEMS ARE IMPAIRMENT
 - SUBTLE PERCEPTION PROBLEMS MAKE LEARNING SUBTLE ASPECTS OF LANGUAGE MORE CHALLENGING (I.E. CENTRAL AUDITORY PROCESSING IMPAIRMENT)

QUESTIONS?