Remote Assessment: Promises & Challenges

HEATHER HAZLETT, PHD

DEPT PSYCHIATRY & THE CAROLINA INSTITUTE FOR DEVELOPMENTAL DISABILITIES

UNIVERSITY OF NORTH CAROLINA AT CHAPEL HILL

Challenges in Pediatric Neuroscience   May 12, 2023

Remote phenotyping

Remote phenotyping can include
- Paper or online questionnaires
- Unsupervised assessments (e.g., home recordings)
- Phone interviews
- Home visits
- Virtual assessments and observations (e.g., telehealth)

Remote Assessment

Pros

- Cost efficient
- Increased sample size
- Familiar environment
- Reduced burden

Cons

- Tele/video requires tech
- Internet connection
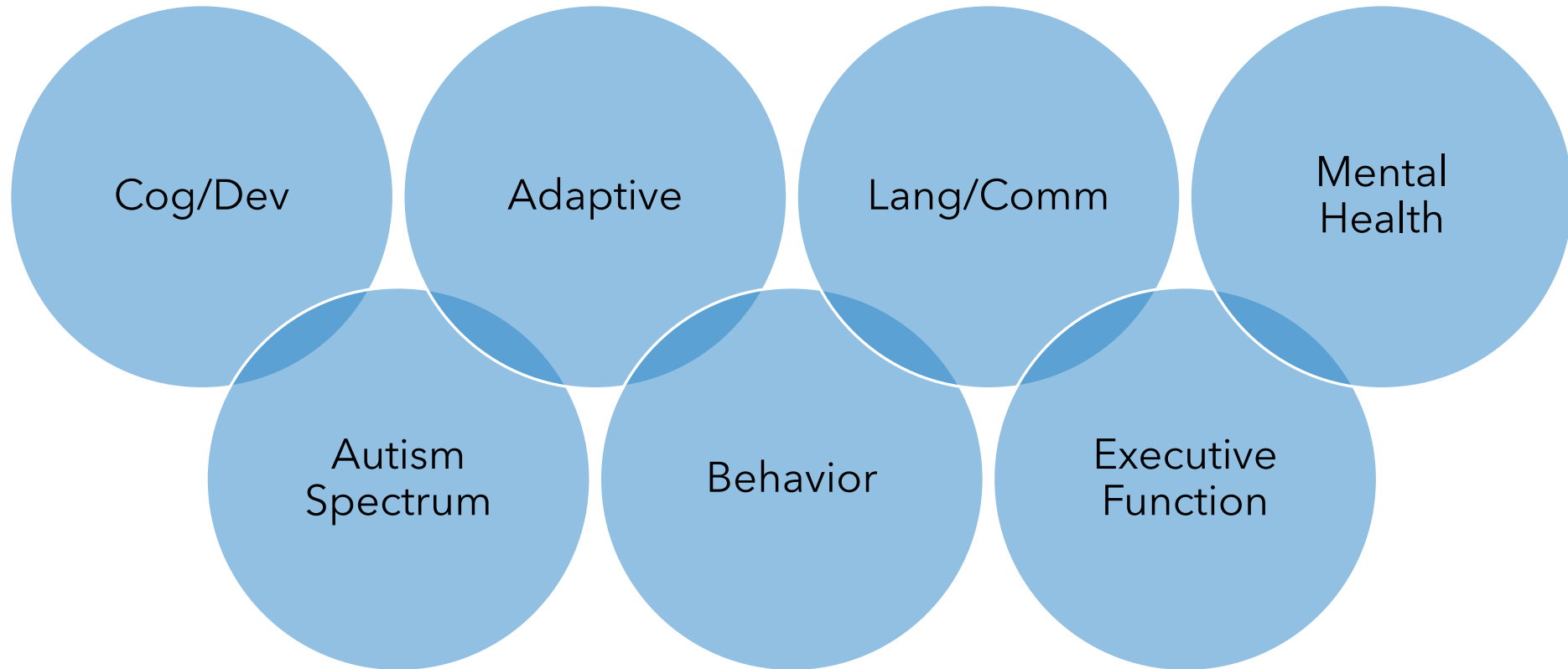- Limitations to measures
- Uncontrolled space

Need for remote assessments

- Research visits during COVID pandemic paused at most institutions and investigators needed to pivot to other means of data collection

- Limited number of tools validated for remote (telehealth) administration, especially cognitive, and even less for special populations (IDD, rare diseases, etc.)
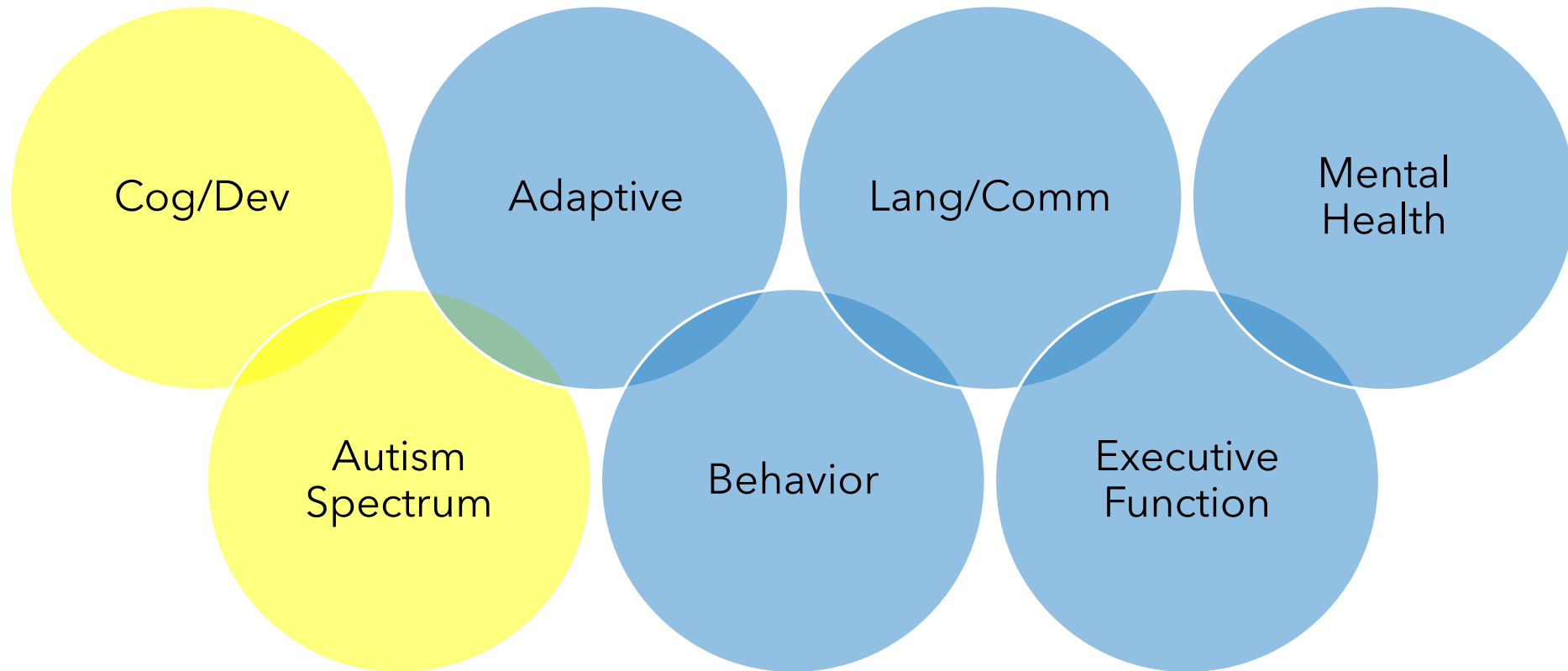
Informed by Literature/Studies

- Remote use of NIH Toolbox and use with IDD (Rebchuk et al, 2019; Shields et al. 2020)

- Review existing remote measures currently in use or under development
  - PANDAbox, Tele-ASD-PEDS, BOSA, SORF, CARS observer, etc.

- JADD special issue (Dec 2022, v51,12): Expanding Telehealth Opportunities in Neurodevelopmental Disorders (editors Rachel Shaffer & Craig Erickson

Domains of interest

Cog/Dev

Adaptive

Lang/Comm

Mental Health

Autism Spectrum

Behavior

Executive Function

Some domains better suited than others to telehealth

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Adaptive

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Remote Assessment & Validation Studies

IDDRC-CTSA - National Brain Gene Registry

IDDRC-CTSA
Brain Gene Registry
(U01)

Collaboration of 13 IDDRCs
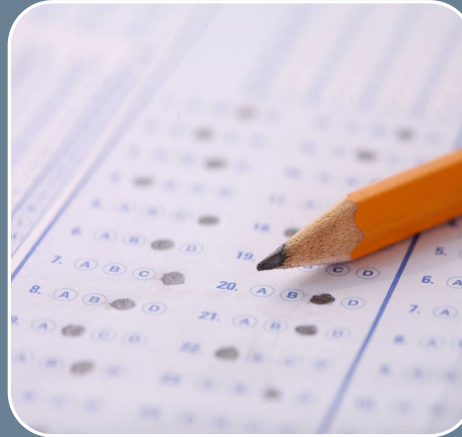
Aim1

Standardized Phenotyping

University of North Carolina
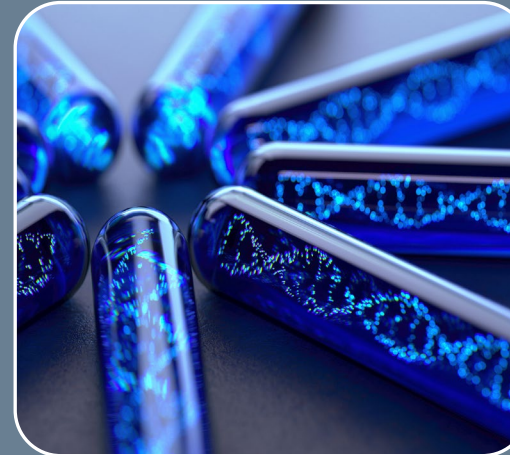
Aim 2

Gene Curation

Boston Children's/ Harvard

Aim 3

Build the Registry

Washington University at St Louis

Rapid Neurobehavioral Assessment Protocol (RNAP)

- Standardized phenotyping battery for domains relevant to IDD
- Flexible for a broad developmental age range
- Consists of existing, normed tools available at no or low cost
- Can be administered by a variety of clinicians
- Remote/telehealth administration

What domains are assessed in the RNAP?

Cognitive

Adaptive

Neurologic

Physical features

Autistic symptoms

Motor/ Sensory

Psychiatric symptoms
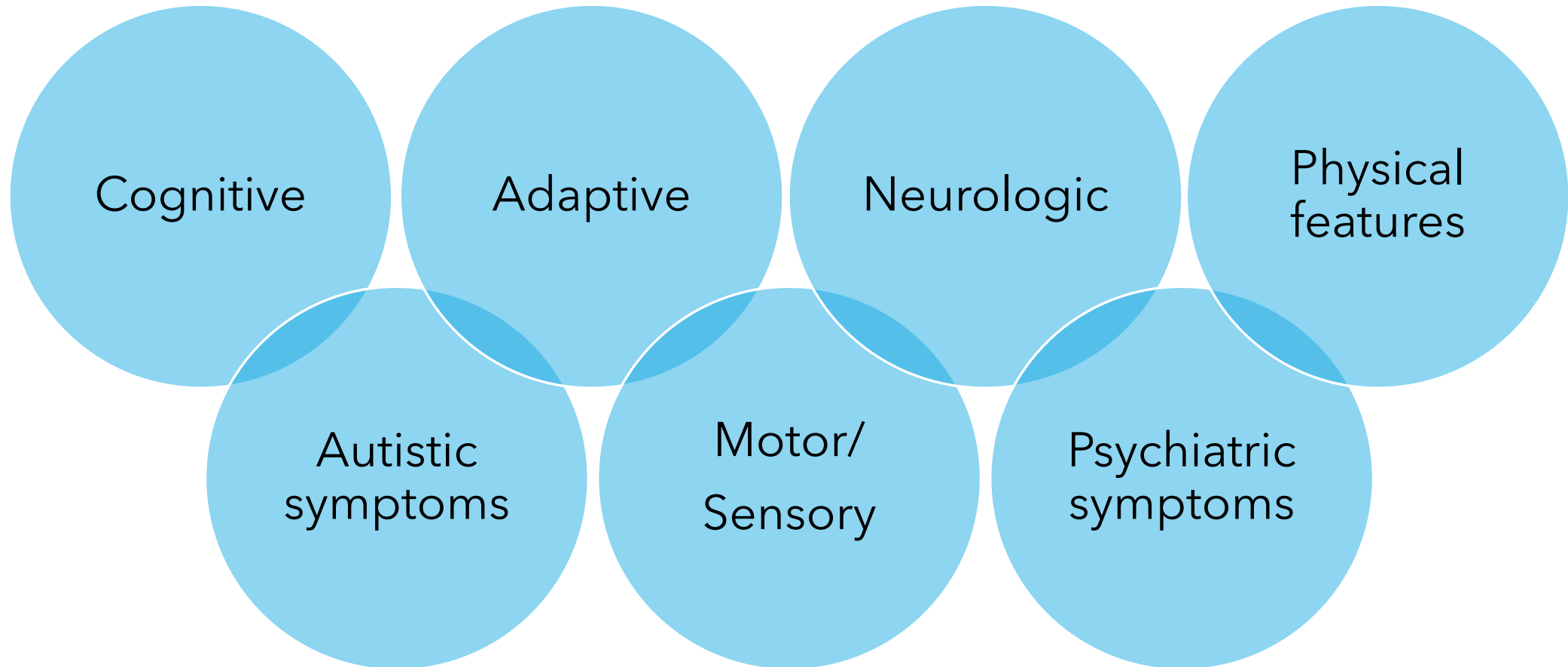
RNAP battery

Domains	Measures	Assessment
Cognitive ability	Shipley or Dev-Profile 4	Direct assessment/report
Adaptive functioning	Vineland Behavior Scales-3	Caregiver report
Autism symptoms	SCQ, CARS-2, SRS	Ratings/Observation
Motor/Sensory	RBS-R, SEQ, DCDQ, GMFCS	Caregiver report
Psychiatric symptoms	Vanderbilt ADHD, CBCL, ABC-2	Caregiver report
Neurologic concerns	Seizure Hx, Neuro screen	Ratings/Observation
Physical features	Dysmorphology screen	EHR/Observation

Measures Legend: RBS-R (Repetitive Behavior Scale, Revised); SEQ (Sensory Experiences Questionnaire 3.0); DCDQ (Developmental Coordination Disorder Questionnaire); GMFCS (Gross Motor Function Classification System, Family & Self Report); SRS-2 (Social Responsiveness Scale, 2nd Ed); CARS-2 (Childhood Autism Rating Scale, 2nd ed); Vanderbilt ADHD (NICHQ Vanderbilt Assessment Scale, 3rd ed); ASEBA CBCL (Achenbach Child Behavior Checklist scales); ABC-2 (Aberrant Behavior Checklist, 2nd ed.)

Two main components of the RNAP

- **Questionnaires and/or caregiver reports**
    - Approximately 90-120 minutes (depending on number of forms needed)
    - All delivered via email to the subject/caregiver (from the BGR REDCap portal)
    - Caregivers can complete at their own pace (e.g., convenient times, done in batches, etc.)
    - Completion of forms is tracked in the BGR REDCap database

- **Direct assessment, conducted via telehealth**
    - Approximately 30-45 minutes (depending on need for ASD assessment)
    - Direct assessment includes: cognitive testing, autism screening (if necessary), neurologic screener, and the dysmorphology screener

RNAP Validation

Grant proposed 'passive' validation of RNAP
- Establish concurrent validity (e.g., results of RNAP classifications align with clinical 'gold standard' diagnostic data

Added an 'active' validation cohort at UNC site (N=40)
- Pediatric or adult subjects with diagnosed neurodevelopmental disorder by clinical DX evaluation (do not need BGR genes)
- Enrollment for purpose of completing RNAP for 'active' validation

  ✓UNC IDDRC Research Registry eligible subjects
  ✓Subject enrollment/data collection - ongoing

RNAP-Spanish data collection for BGR

- Spanish enrollment needed for sites with identified subjects

- Required translation of all BGR materials (e.g., consents, recruitment) and RNAP into Spanish

- RNAP Spanish data collection (*started 2023*)

Remote is here to stay….

- New measures under development
- Validation efforts underway by several groups
- Clinical care will continue to offer telehealth
- Test publisher offering remote/virtual measures and platforms
- Many positives and potential…

Test developer platforms

Remote phenotyping brings equity

- Expands potential geographic range/location
- Reduces need for travel/cost for research
- Inclusion of rare disease and medically-involved cohorts
- Use of Spanish and other languages
- Well suited for clinical trials (less travel burden on families)

Opportunities for young investigators

- Extends research dollars

- Allows for cross-site collaborations, mentoring, and consultation

- New lines of scientific inquiry for junior investigators, fellows, and trainees

Broadens the scope and scale

- Remote phenotyping can be well suited for implementing on large scale

- Application to phenotyping for genetics studies, subject registries, rare disease networks, clinical trials, biospecimen repositories

Next steps and actions

- Validation efforts for measures, special populations

- Expert curation, white papers, reviews, etc.

- Development of new measures, practices, and tools tailored for telehealth

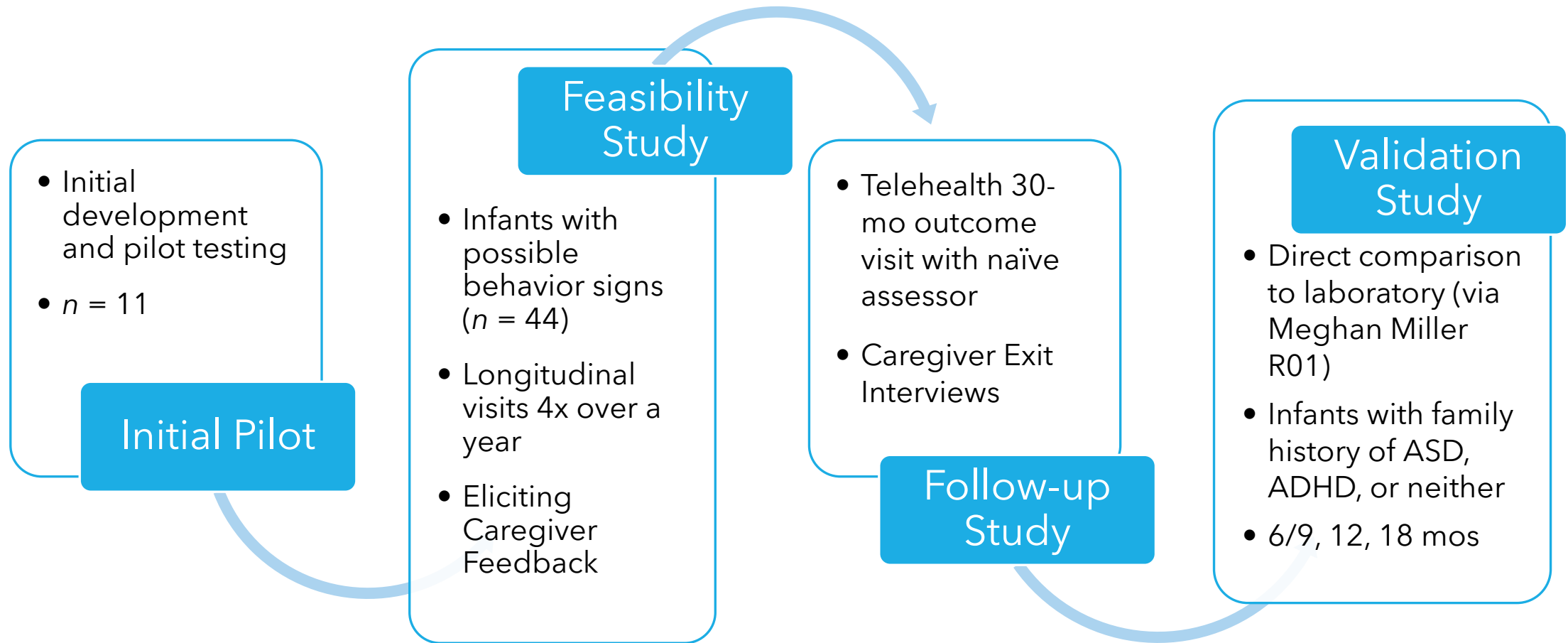# The TEDI: Telehealth Evaluation of Development for Infants

PI Meagan Talbott with Sarah Dufek, Greg Young, Sally Rogers and Meghan Miller

R21 & KL2 (Talbott)

Goals:

1. Develop and validate a social-communication focused telehealth evaluation for 6-30 months

2. Use the TEDI to understand developmental trajectories and outcomes of infants with concerns in first year of life

- TEDI components: families sent small kit and activity descriptions, 1hr live telehealth session coaching parents through 12-15 play activities

- Measures: AOSI, P-ESDM Curriculum Checklist (Developmental), Early Communication Indicator (ECI), Alberta Infant Motor Scales (AIMS), caregiver questionnaires, behavioral coding.

The TEDI: Telehealth Evaluation of Development for Infants

Initial Pilot

- Initial development and pilot testing
- *n* = 11

Feasibility Study

- Infants with possible behavior signs (*n* = 44)
- Longitudinal visits 4x over a year
- Eliciting Caregiver Feedback

Follow-up Study

- Telehealth 30-mo outcome visit with naïve assessor
- Caregiver Exit Interviews

Validation Study

- Direct comparison to laboratory (via Meghan Miller R01)
- Infants with family history of ASD, ADHD, or neither
- 6/9, 12, 18 mos

IDDRC Clinical Translational Core Remote Cog/Bx Workgroup

Baylor – Holly Harris

Boston Children's/Harvard – Ellen Hanson

CHOP – Juhi Pandey

Children's National – Andrea Gropman, Youssef Kousa, Sen Kuntal, Madison Berl, Nikkia Zarabian

Einstein –Catherine Sancimino,

NIH – Alice Kau, Tracy King

UCLA – Amanda Gulsrud

UC-Davis – Sally Ozonoff, Meagan Talbott

UNC – Heather Hazlett, Jessica Kinard

University of Rochester – Heather Adams, Leona Oakes

University of Washington – Eva Kurtz-Nelson, Emily Neuhaus, Sara Jane Webb

Wash Univ St Louis – Christopher Smyser, Anna Abbacchi

Wisconsin – Hayley Crain

Measure curation & evaluation

Curate
- Name, Publisher, Domain, Modality, Age Range, Admin Time, Materials needed, Examiner Qualifications

Evaluate
- Valid for telehealth, Appropriateness for IDD, Nonverbal, Other Languages

IDDRC Remote Phenotyping

"Brown Bag" Invited Talks

Motor Phenotyping : Monday July 18, 12-1pm EST/9-10am PST

- Bhooma Aravamuthan, MD, Dphil, Washington Univ in St. Louis
- Amena Smith Fine, MD, PhD Kennedy Krieger Institute
- Rujuta B. Wilson MD, MS UCLA

EEG "at home": Monday October 17, 12-1pm EST/9-10am PST

- Kate Nooner PhD, UNC Wilmington
- Caitlin Hudac, University of South Carlina

Thank You!