

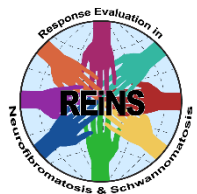


Response Evaluation In Neurofibromatosis Schwannomatosis INTERNATIONAL COLLABORATION

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Neurocognitive Outcomes Working Group Update: Evaluation of Measures of Intellectual Ability, Memory, Language and Executive Functions

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Response Evaluation In Neurofibromatosis Schwannomatosis
INTERNATIONAL COLLABORATION

Mission of Neurocognitive Group

- Goal to identify measures of cognition and psychosocial functioning for use in clinical trials
 - Well-designed measures (good psychometrics)
 - Easily administered in clinical trials setting
 - Previous use in clinical trials
 - Previous use with NF1
 - Alternate forms
 - Availability in other languages



Measures Defined as Core, Supplemental, or Emerging

- Core
 - Established use in NF1
 - Good Psychometrics
 - Brevity and ease of administration
 - Multiple languages
- Supplemental
 - Specific topics or more in-depth assessment
- Emerging
 - Instruments under development, in the process of validation, or nearing point of published findings with NF1



Relevant Domains

- Cognition
- Language
- Visual-spatial
- Memory
- Attention
- Executive functions
- Academics
- Social skills
- Social cognition
- Computerized measures
- Global outcome measures



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- Cognition
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School age measures 2016 Neurology supplement

Preschool measures 2021 Neurology supplement

2021 Neurology supplement



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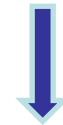
Inhibit

Self-Monitoring



Shifting

Emotional Control



Initiate

Working Memory

Task Monitoring

Plan/Organize

Flexibility



Measures of Executive Functions

STROOP PARADIGM TASKS

Stroop Match-to-Sample Task
ClinicaVR: Classroom-Stroop
Motor Stroop task
Emotional Stroop task
Number-quantity Stroop
Chimeric animal Stroop
Counting Stroop task
Stroop residual [interference] test
D-KEFS Color-Word Interference Test (CWIT)
Classical Stroop task
Stroop Color-Word Test (SCWT)

Total

TOWER PARADIGM

Tower of Coimbra
Tower of Hanoi (ToH)
Tower of London (ToL)
DKEFs Tower test

Total

TRAILS PARADIGM

Children's Color Trails Test (CCTT)
Comprehensive Trail Making Test (CTMT)
Color Trails Test (1 and/or 2)
TMT (Trail Making Tests, A and/or B)

GO/NO-GO PARADIGM

Affective Go/No-Go (AGN)
Classical Go/No-go

MAZE PARADIGM OF TASKS

Executive Maze Task [EM]
Virtual Water Maze
Reasoning and Problem-Solving mazes
Arena Maze
Porteus Mazes [Maze test]

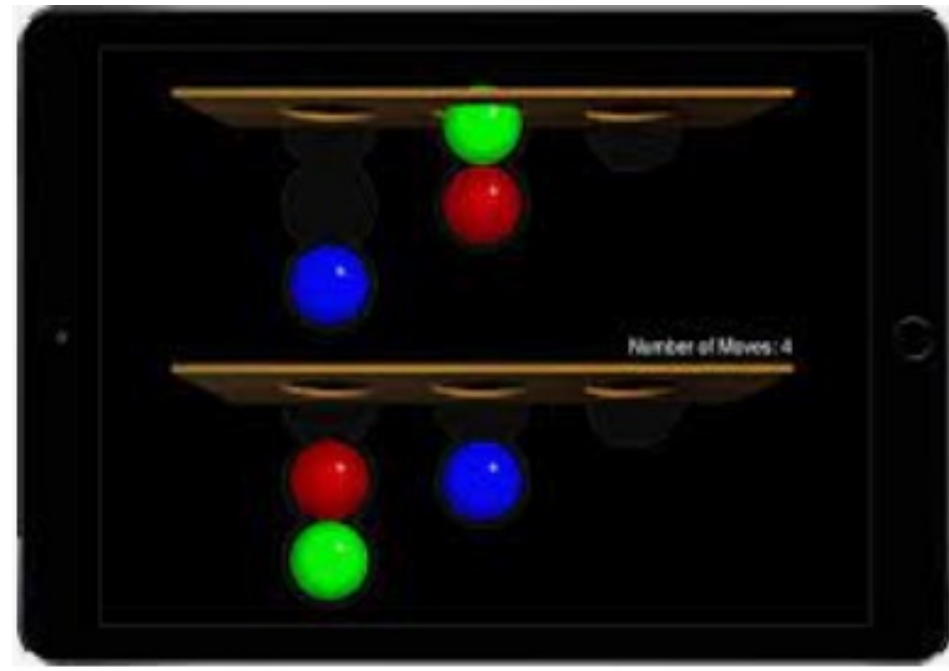
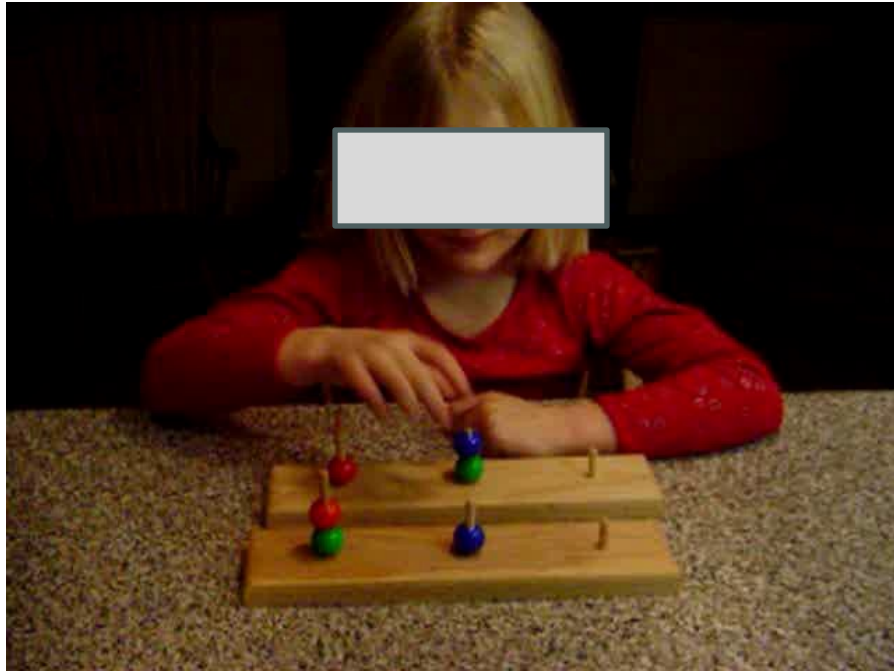
CARD SORTING PARADIGM

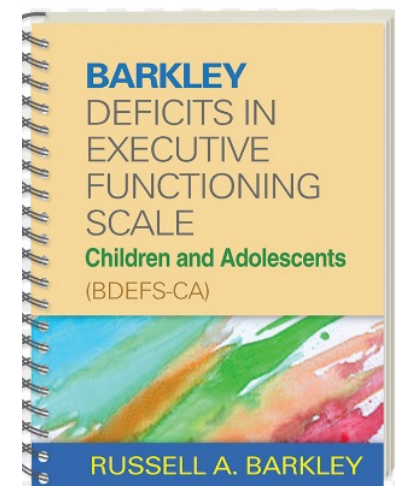
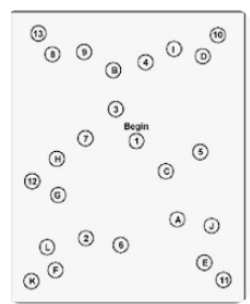
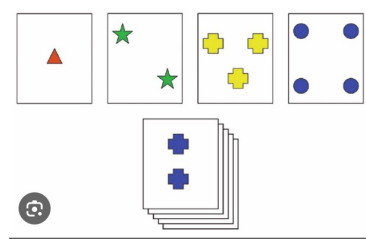
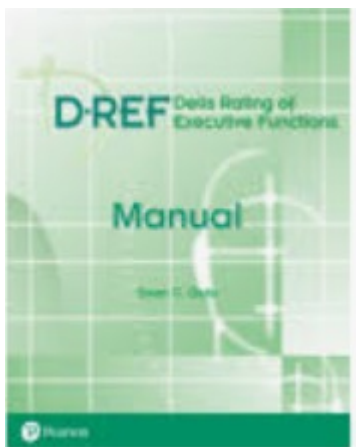
Dimensional Change Card Sort Test (DCCS)
Wisconsin Monster Sorting Test
Madrid Card-Sorting Test (MCST)
DKEFs Card sorting test
WCST (Wisconsin Card Sorting Test)

N-BACK PARADIGM

Penn Short Letter N-Back Test (SLNB)
2-n-back task
Spatial n-back
Letter N-back test
N-back test (verbal and/or visual)







COGRATE: COGNITIVE outcomes Rating Acceptance Tool for Endpoints

Measure:

Rater: _____ Date: ____/____/____

RATINGS

3=Solid data and published information supporting its use in clinical trials

2=Good preliminary data and relevant information but needs more work

1=Limited data but information suggests potential

0=No/poor data/information

*Half ratings (.5, 1.5, 2.5) can be used if needed



Rating Criteria	Rating (0-3): use in NF trials
1. Patient characteristics: Age range (e.g., child, adolescent, adult) Normative groups (e.g., general, NF, oncology, other, # subjects)	
2. Used in published studies: Number and types of studies (e.g., descriptive, clinical trials)	
3. Test appropriateness for clinical trials endpoint: Test specificity/purity (how pure is the measure to the domain/skill it is developed to measure?) Test targets one or more of the known or future endpoints for NF clinical trials directly	
4. Scores available: Types of scores available (e.g., raw, standardized, domain, total; gaps in normative data)	
5. Psychometric Data: Reliability (e.g., internal consistency, test/retest) Validity (e.g., construct, discriminative) Factor analysis Practice Effects/Availability of Alternate Forms/Time between testing information	

6. Feasibility: Cost (for test instrument, protocols, and scoring) Length (time to administer/complete) Ease of administration/challenges to administration Qualifications to administer test (level of training) Appropriateness for alternative testing settings (e.g., clinic, etc.) Other languages available	
Overall Impression for use in NF Clinical Trials (Pros/Cons) – Is the measure a critical Primary Outcome Measure?:	Total (mean):

Recommended as a:

- Core outcome measure
- Supplementary outcome measure
- Emerging outcome measure
- Not acceptable (no further review)

Committee Notes/Comments:

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Thank you

Chair

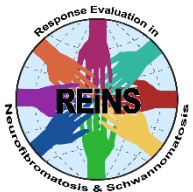
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- Maureen Hussey
- Sharon Loftspring
- Jessica Samblanet
- Herb Sarnoff
- Steven Sheard
- Connie Sorman
- Tracy Wirtanen



* Steering Committee