

Measuring Meaningful Change in Individuals with Intellectual Disability

Audrey Thurm¹

1. Neurodevelopmental and Behavioral Phenotyping Service, Office of the Clinical Director, NIMH, 10 Center Dr. Rm 1C250, MSC 1255, Bethesda, MD 20892

This talk will focus on measurement of functioning in Angelman and Dup15q Syndromes, and in populations with Intellectual Disability more generally. This topic is of particular importance, given that cognitive and behavioral measurements are necessary for describing the natural history of such conditions, as well as for treatment trials that require adequate outcome measures. For treatment trials, such measures will need to adequately show improvement when treatments are actually effective, so they must be sensitive to change. Many measures of cognition, including basic Intelligence Quotient (IQ) tests, are not adequately developed or validated for individuals with severe-to-profound Intellectual Disability. This means that they do not have the ability to show minimal clinically important differences. Potential methods for addressing these gaps will be discussed, as well as use of other proxies and associated measures, including those that measure functioning in the real world. Measurements of adaptive behavior, for instance, provide evidence for how individuals function in key domains, and can be measured over time and across ages. Examples will be given from related research where modifications to existing measures, novel measures and different approaches have allowed for detection of change in applicable populations.