

Designing measures of communication ability in individuals with Angelman syndrome

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INTRODUCTION

- Angelman syndrome (AS) is a rare neurodevelopmental disorder associated with communication impairment.
- Existing communication measures are unable to properly scale individuals with AS on a metric that allows for evaluation of changes in communication ability from baseline to post-treatment.
- To support the evaluation of new therapeutics for AS, we identified important domains of communication in this population and explored how caregivers defined meaningful change.

METHODS

- Concept elicitation interviews were conducted with experts in communication and caregivers of individuals with AS to understand the key concepts that are relevant for assessing communication ability in this population.
- Enrollment of caregivers was stratified by child age and AS genotype.
- Caregivers were asked to describe how their child communicates, including use of gestures, vocalizations and Augmentative and Alternative Communication (AAC) devices. Communication experts characterized communication ability in individuals with AS.
- After interviews concluded, data was organized, and interviewer notes and interview transcripts were coded for content and themes.

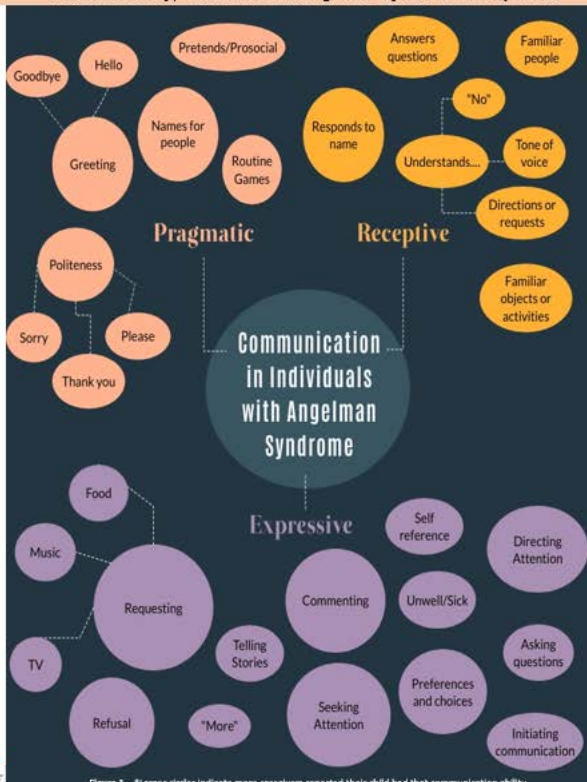


Figure 2. *Larger circles indicate more caregivers reported their child had that communication ability.

RESULTS

- Twenty-two caregivers (91% female) of individuals with AS and six communication experts (83% female) participated in interviews.
- Important communication concepts identified by caregivers and communication experts included expressive, receptive and pragmatic communication (Figure 1).
- Individuals with AS showed variability in communicative skills based on their primary communication modality (i.e. words, signs/gestures and AAC devices).
- Individuals with rare genotypes, such as Uniparental Disomy, had more advanced communication abilities than individuals with the Deletion Positive genotype.
- Caregivers defined meaningful change in communication as being able to express feelings, comment, express an opinion, or tell about an event.

DISCUSSION

- Information gathered in these interviews will be used to develop items for a novel caregiver-reported communication measure.
- Future work will include cognitive interviews to refine and revise content of the measure and a cross-sectional psychometric study.