Development, validation and use of the Quality of Life Inventory-Disability (QI-Disability)

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Intellectual disability

- Children may experience
  - Poorer physical and mental health
  - Activity limitations and participation restriction
  - Social isolation and disconnectedness from peers
- Convention on the Rights of Persons with Disabilities – article 24
  - *“Persons with disabilities have the right to the enjoyment of the highest attainable standard of health without discrimination on the basis of disability.”*
- The words of a Mother
  - *“The most important thing to remember is that when a child is born, that child is a child first to his or her family. It is his well-being, not his disability, that affects all of our everyday lives.”*

Toolkit of QOL measures

Modified Auto Questionnaire Enfant Image (AUQEI) (Cui 2008)	27 items	Based on QOL domains for children in general population
Quality of life – Profound Multiple Disabilities (QoL-PMD) (Petry 2009)	55 items	Scale limited by group and item structure, e.g. … *mental health status is good* …. *receives tasty and balanced nutrition*
KidsLife scale (Gomez 2016)	96 items	Based on an eight-domain QOL model for adults

The development of QI-Disability

1. To describe consumer perspectives on QOL in children with intellectual disability

2. To illustrate development processes to ensure content validity of QI-Disability

3. To present initial validation data and use of QI-Disability

Consumer perspectives

77 qualitative interviews

Cerebral palsy (n=18)	**Autism spectrum disorder (n=21)**
Victorian Cerebral Palsy Registry est. 1987 7-18 years 9 males and 9 females 5 (28%) GMFCS IV and 6 (33%) GMFCS V 14 (77%) non verbal 9 (50%) with behaviour problems	**WA Autism Biological Registry est. 1990** 6-17 years 13 males and 8 females 7 (33%) with health comorbidities (e.g. obstructive sleep apnoea, epilepsy, otitis media)
Rett syndrome (n=21)	**Down syndrome (n=17)**
Australian Rett Syndrome Database est. 1993 7-18 years All females 12 (57%) walked independently 4 (19%) with gastrostomy 5 (24%) with spinal fusion	**Down Syndrome NOW Database est. 1997** 6-18 years 8 males and 9 females 6 (35%) with complex health problems 13 (76%) with some intelligible speech 6 (35%) dependent for personal care

Interview schedule and coding

- Semi-structured interviews
  - e.g. *When everything is going great, what sorts of things are happening?*
  - *When things are not so good, what sorts of things are happening?*
  - Probe: *"How do you know?"* or *"What does that look like?"*
- Recordings transcribed and reviewed by parents

- Qualitative analyses
  - Grounded theory
- Coded observable behaviour, e.g.

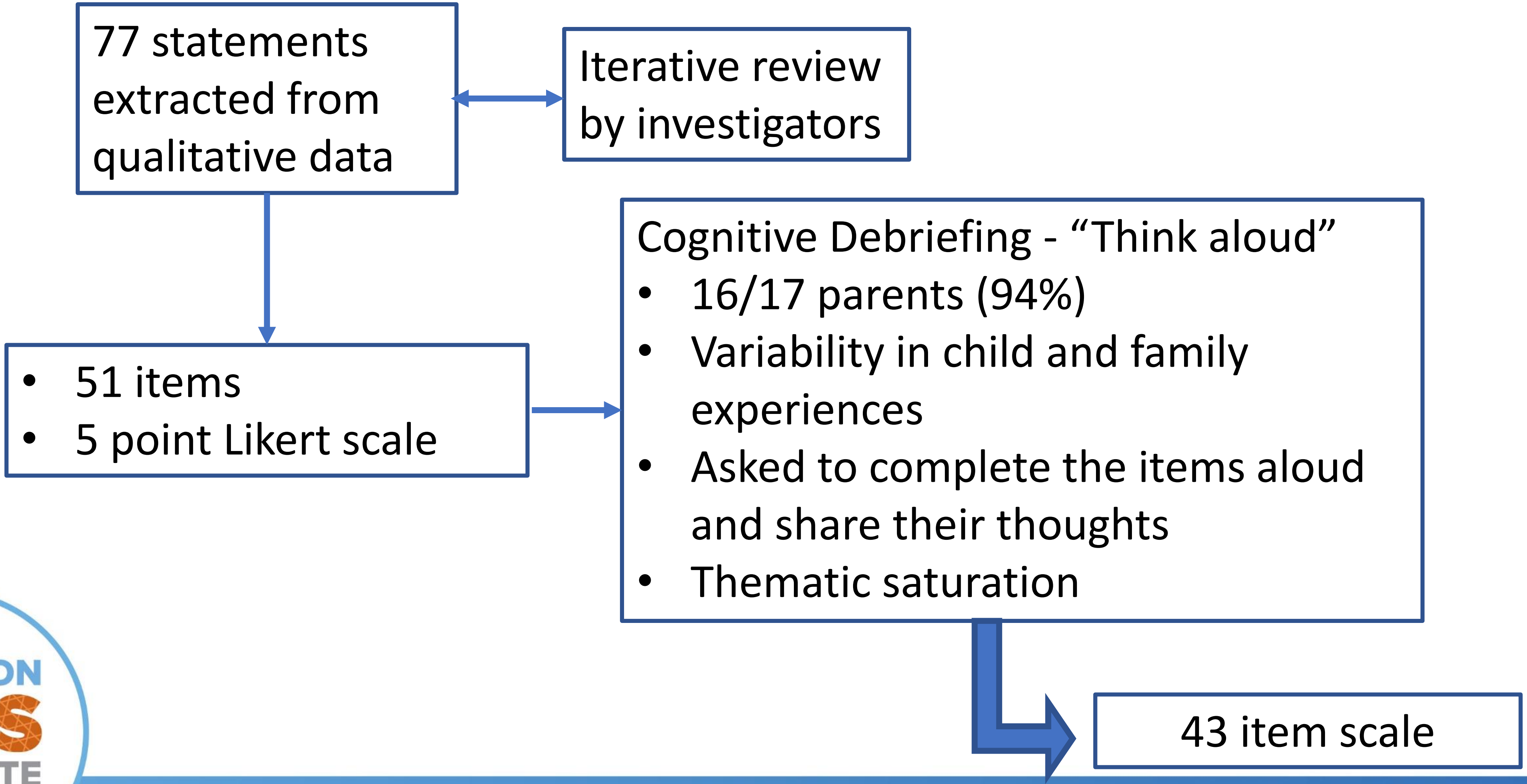
Emotion
"She's feeling happy"
Observable behaviour
"She will smile and laugh."

Categories	11 Domains
Health and wellbeing	• Physical health • Behaviour and emotion • Body pain and discomfort • Personal value
Daily activities	• Communication • Movement and physical activity • Stability in daily routines ← • Independence and autonomy
Community immersion	• Social connectedness and relationships • The natural environment ← • Variety of activities

Epstein 2016, Murphy 2017, Davis 2017, Epstein 2019

Content validation

Candidate items to a draft measure



77 statements extracted from qualitative data

Iterative review by investigators

- 51 items
- 5 point Likert scale

Cognitive Debriefing - “Think aloud”

- 16/17 parents (94%)
- Variability in child and family experiences
- Asked to complete the items aloud and share their thoughts
- Thematic saturation

43 item scale

Epstein 2019

Initial validation

Pilot study (n=253)

Down syndrome (n=61)	Autism spectrum disorder (n=62)
The Down Syndrome NOW Database est. 1997	The WA Autism Biological Registry est. 1990 & WA Autism Registry
Rett syndrome (n=66)	**Cerebral palsy (n=64)**
The Australian Rett Syndrome Database est. 1993	The Victorian Cerebral Palsy Registry est. 1987

Downs 2019

Child and family characteristics (n=253)

- Children
  - 5-12 y – 45.4%
  - Female – 62.1%
  - Use words – 27.7%
  - Walk independently – 58.5%
  - Could look after personal needs – 23.1%
  - Gastrostomy – 17.0%
  - Epilepsy – 35.6%
  - Vision problems – 35.6%

- Families
  - Natural mother – 89.7%
  - F/T or P/T work – 53.6%
  - Urban living – 82.6%

Factor analysis

Exploratory factor analysis
6 items dropped

Confirmatory factor analysis
2 items dropped

Questionnaire
32 generic items

- Six-factor structure (item loading >0.4)
- Convergent and divergent validity
- Factor structure held when the sample was split
  - Independent walking
  - Ability to talk
- Factor scores modestly correlated
- Cronbach alpha values >0.72

Six-factor structure

“Social interaction” (7 items)	**“Negative emotions” (7 items)**
Expressed happiness when they were understood Enjoyed the social experiences of meal times Appeared relaxed when making eye contact	Showed aggression (e.g. hitting, kicking, using offensive language, being destructive) Appeared upset or angry (e.g. crying, screaming, moving or stiffening the body)
“Leisure and the outdoors” (5 items)	**“Independence” (5 items)**
Enjoyed moving their body (e.g. crawling, walking, swinging, swimming) Enjoyed spending time outdoors (e.g. contact with water, grass, wind, sunshine)	Expressed their needs (e.g. hunger, thirst, toileting) Helped to complete routine activities (e.g. dressing, feeding)
“Physical health” (4 items)	**“Positive emotions” (4 items)**
Had enough energy to participate in daily routines and activities Been alert and aware during the day	Showed happiness through body language (e.g. making eye contact, body facing others) Showed cheeky or comical mannerisms (e.g. laughed, giggled)

Factor scores

	Mean (SD)	Range
Total score	67.9 (14.3)	25.6 - 94.8
Social interaction	70.6 (19.8)	17.8 - 100
Negative emotions	63.4 (18.7)	7.1 - 100
Leisure and the outdoors	71.3 (21.0)	0 - 100
Independence	60.4 (24.0)	0 - 100
Physical health	66.2 (19.1)	12.5 – 95.8
Positive emotions	74.1 (18.6)	18.8 – 93.8

Comparison of scores by diagnosis (coefficient, 95%CI)

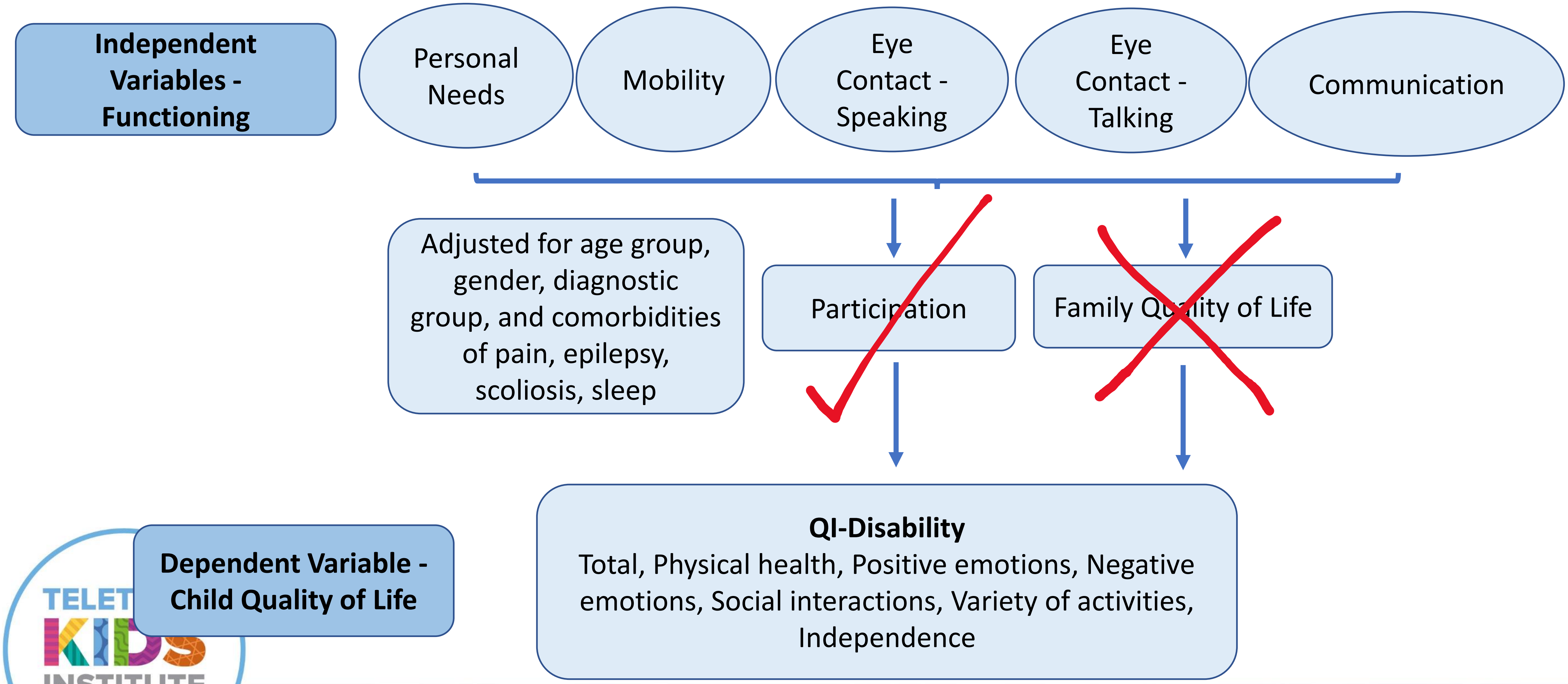
	Social interaction	**Negative emotions**	**Leisure and the outdoors**	**Independence**	**Physical health**	**Positive emotions**
Rett syndrome	REF	REF	REF	REF	REF	REF
Cerebral palsy	-6.52 (-12.96, 0.08)	2.94 (-3.47, 9.34)	-1.54 (-8.71, 5.64)	-3.38 (-10.12, 3.35)	2.67 (-3.95, 9.29)	0.67 (-5.64, 6.98)
Down syndrome	7.13 (0.61, 13.65)	1.87 (-4.56, 8.30)	10.60 (3.36, 17.83)	29.70 (22.88, 36.52)	9.10 (2.42, 15.78)	4.78 (-1.59, 11.14)
Autism spectrum disorder	-12.81 (-19.28, -6.35)	-8.07 (-14.47, -1.67)	0.27 (-6.97, 7.50)	23.31 (16.55, 30.07)	4.12 (-2.53, 10.77)	-7.09 (-13.42, -0.75)

Determinants of child QOL

Is functioning associated with child QOL?

- Methods
  - Recruited from 4 diagnostic groups
  - Online (paper) questionnaire
  - Independent variables
    - Functioning
    - Comorbidities
    - Participation
    - Family QI-Disability
  - Dependent variable
    - Child QOL – QI-Disability

- Sample
  - Completed questionnaires for 425 5 to 19 year children
    - ASD – 133 (31.3%)
    - CP – 151 (34.7%)
    - DS – 89 (20.9%)
    - RTT – 62 (14.6%)
  - 50.8% primary school
  - 51.5% male
  - 404 in final model



Independent Variables - Functioning

Personal Needs

Mobility

Eye Contact - Speaking

Eye Contact - Talking

Communication

Adjusted for age group, gender, diagnostic group, and comorbidities of pain, epilepsy, scoliosis, sleep

Participation ✓

Family Quality of Life ✗

Dependent Variable - Child Quality of Life

QI-Disability
Total, Physical health, Positive emotions, Negative emotions, Social interactions, Variety of activities, Independence

Independent variables		Coefficient (95% CI)	P value
Personal Needs	Independent	-	--
	Independent with checking	-3.98 (-9.28, 1.31)	0.140
	Needs assistance	-6.82 (-12.29, -1.35)	0.015
	Dependent	-6.57 (-12.56, -0.58)	0.030
Mobility	No difficulty	-	-
	Walks some distances	-1.85 (-4.34, 0.67)	0.149
	Walks with assistance	1.88 (-2.90, 6.65)	0.440
	Unable to walk	-0.67 (-4.97, 3.63)	0.759
Eye contact	Speaking (/12)	0.81 (0.18, 1.43)	0.011
	Listening (/12)	0.24 (-0.40, 0.89)	0.456
Communication	Speaks well	-	-
	Some difficulty speaking	-0.06 (-3.12, 3.25)	0.969
	Difficulty speaking	0.75 (-3.00, 4.51)	0.694
	Nonverbal communication	1.88 (-2.30, 6.07)	0.337
	Unable to communicate	-0.68 (-5.40, 4.03)	0.775
Participation (PEM-CY)	Frequency of participation (/7)	2.84 (1.74, 3.93)	<0.001

Discussion

- Best practice development process
- 32-item scale with a six-factor structure
  - Consistencies with models of ICF and definitions of QOL used for other measures
  - Some concepts and items are unique
- Factors held across levels of functioning
- Differences by diagnosis and age consistent with theory
- Advocacy for more participation opportunities - NDIS

What next?

- Immediate opportunities to measure QOL
  - Possibilities ahead to identify more modifiable determinants of child QOL
  - Exploratory measure in clinical trials, evaluating
    - Ganaxolone for CDKL5 and another for PCDH19
    - An “uptime” intervention for Rett syndrome
- Afford greater capacity
  - To clearly identify support needs
  - As an outcome measure for evaluation studies
  - To guide the development of a self-report QOL measure for children with intellectual disability

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