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REGION

Feasibility and effectiveness of an ‘uptime’ participation intervention (U-PART) in girls and women with Rett syndrome

Michelle Stahlhut, PT, PhD student
Center for Rett syndrome

LUNDS UNIVERSITET
Medicinska fakulteten

Danish center for Rett syndrome

- 118 diagnosed with Rett syndrome, age 3-63 years
- ~ 80% annual follow-up
- 100 have a *MECP2* mutation, ♀= 98, ♂= 2

Background

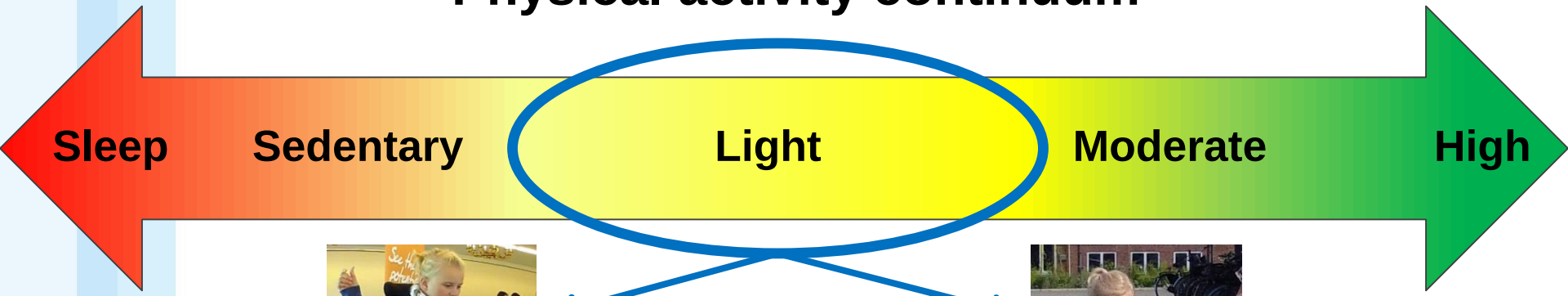
Girls and women with RTT experience:

- High dependency on caregivers in all areas of daily life
- Limited gross motor skills and high levels of sedentary time (>80% of awake hours) and low levels of physical activity (≤ 5,000 steps/day)
- Rescricted participation in everyday and community activities but they enjoy physical and social activities

Background

- Interventions to promote health throughout the lifespan in girls and women with RTT are lacking

Physical activity continuum



Background

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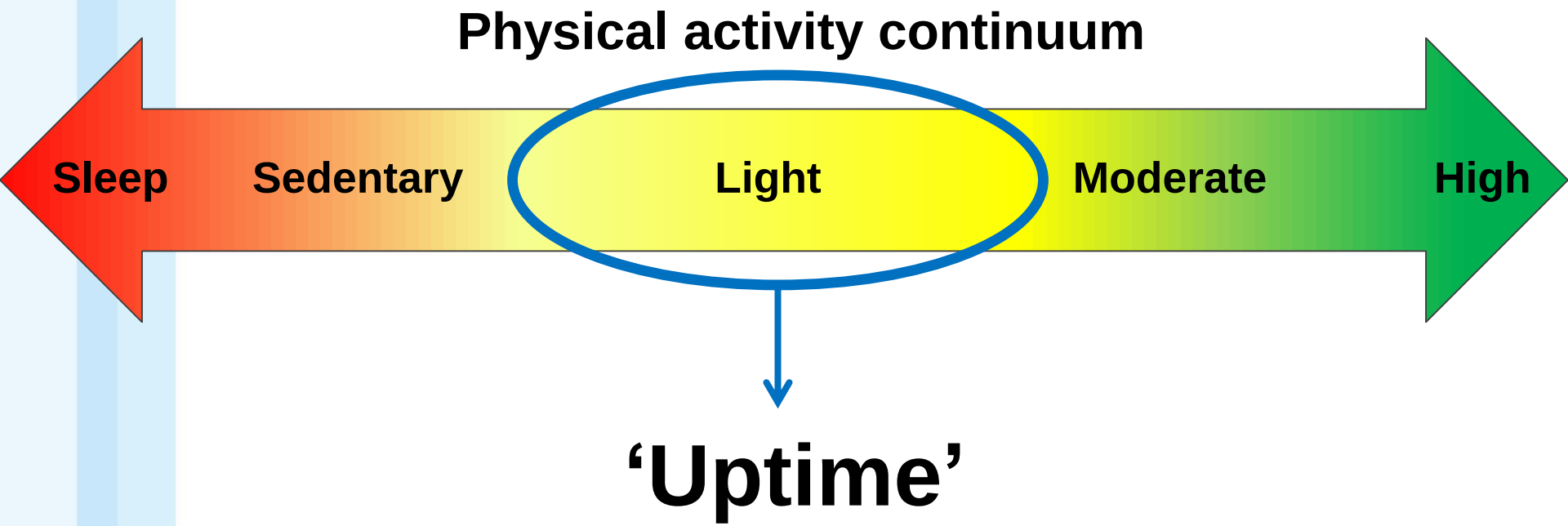
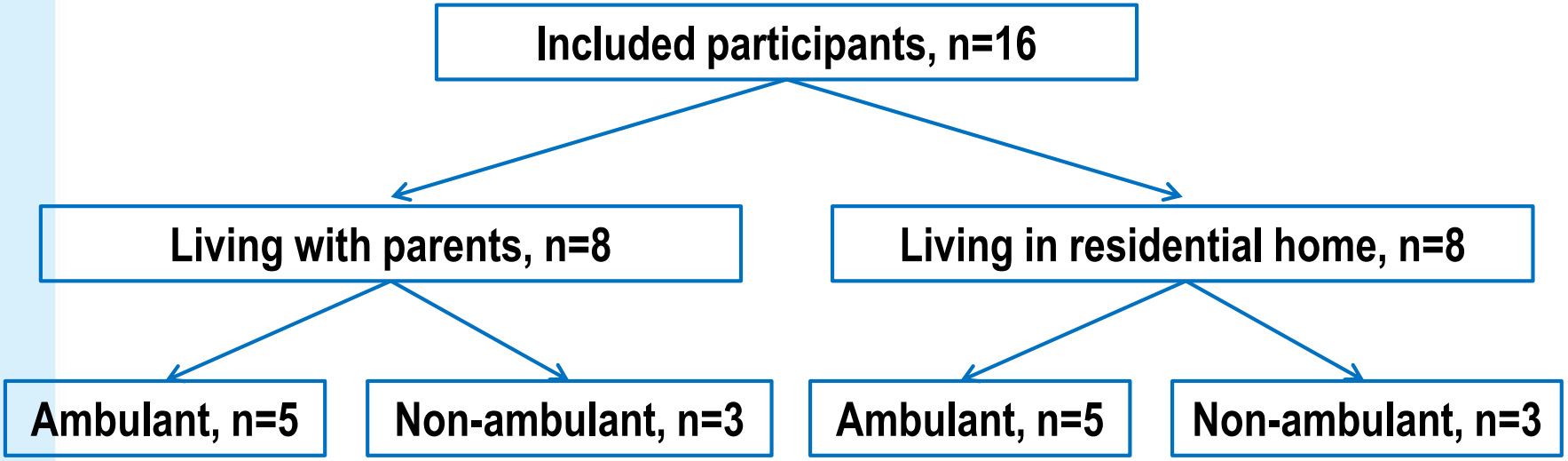


Aim and participants

To evaluate the feasibility and health-related effects of an 'Uptime' Participation (U-PART) intervention in girls/women with Rett syndrome



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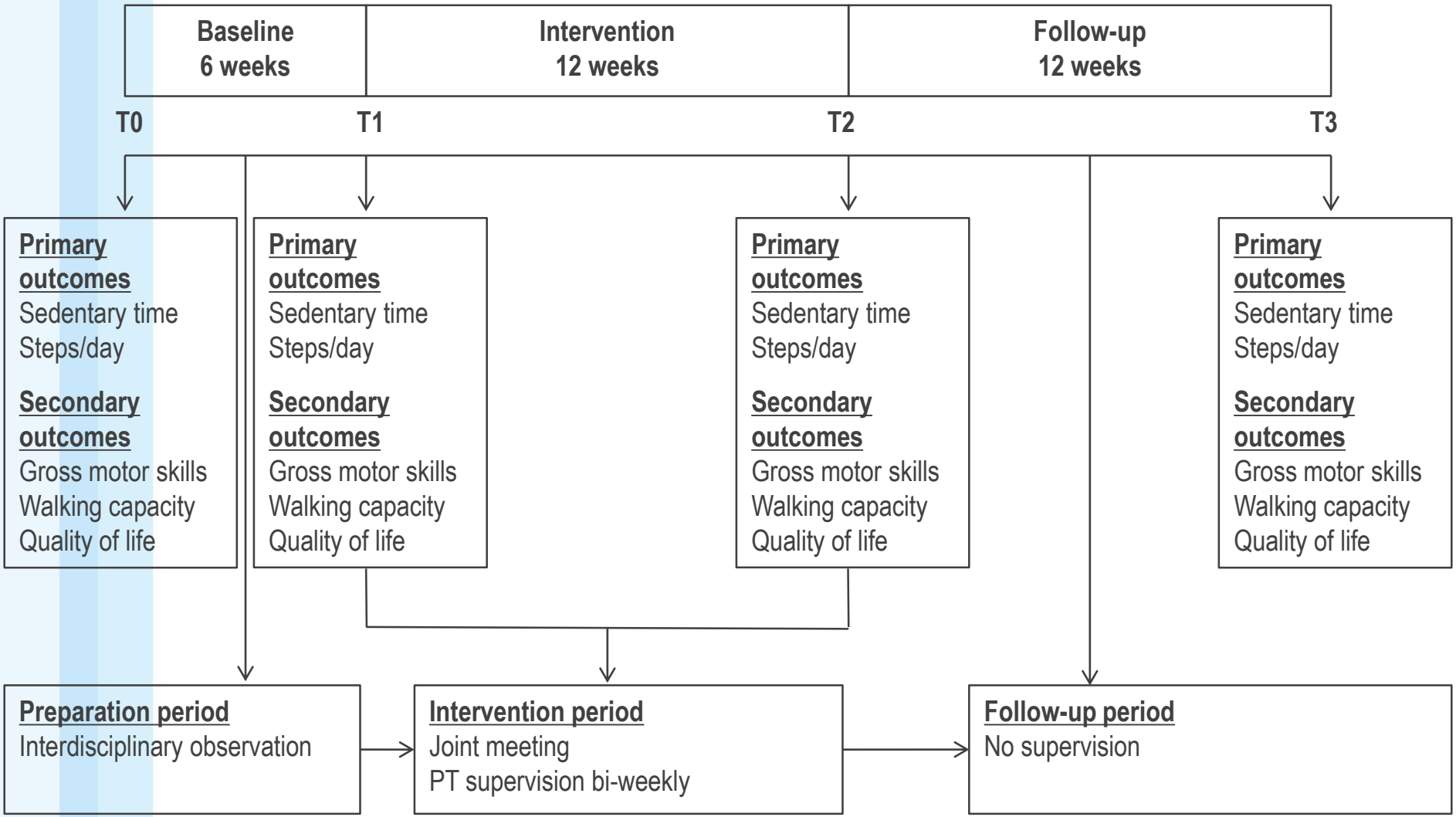
Methods



Data analysis

- Linear mixed-effects models with random intercepts
  - Random-effects variable: Unique participant ID
  - Fixed-effects variables: Intervention
    Age at assessment
    Residence
    Ambulation level

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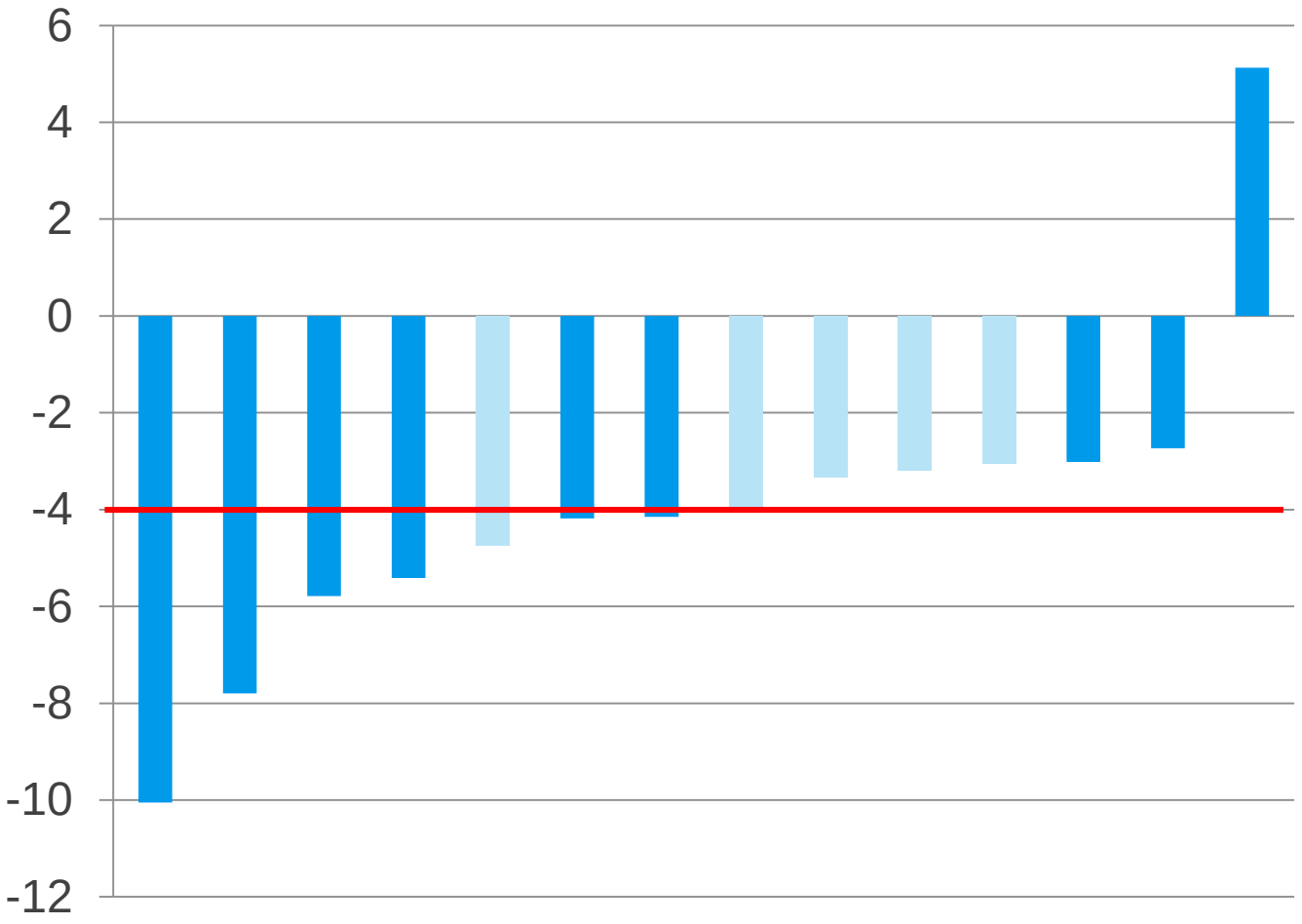
Results

Primary outcomes	T0	T1	T2	T3
Daily sedentary time				
Sitting time (h)	9.79 (1.6)	9.67 (1.6)	9.13 (1.56)	9.21 (1.45)
Relative to waking h (%)	83.5 (10.7)	84.8 (10.5)	80.1 (10.2)*	80.9 (9.6)*
Daily physical activity				
Step count	4291 (2650)	4522 (2642)	5096 (2546)*	4700 (2665)
Secondary outcomes	**T0**	**T1**	**T2**	**T3**
Gross Motor Skills (/45)	21.6 (8.6)	21.3 (8.4)	21.5 (8.3)	21.1 (8.3)
Walking capacity (m)	81.9 (35.4)	93.6 (35.3)	106.5 (33.7)*	99.9 (34)*
Quality of life (/100)	78.3 (7.9)	78.6 (7.8)	81.4 (8.8)*	79.8 (10.6)

After intervention: -4.09% ([95%CI -5.87,-2.32], $p<0.001$)
After follow-up: -3.36% ([95%CI -5.15,-1.58], $p<0.001$)

Individual changes in sedentary time



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After intervention: +708 steps/day ([95%CI 126,1290], $p<0.019$)

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After intervention: +18.94m ([95%CI 7.45,30.42], *p*=0.002)
After follow-up: +12.40m ([95%CI 0.87,23.29], *p*=0.036)

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After intervention: +2.81 points ([95%CI 0.5,5.11], *p*=0.018)

Subgroup analysis

- Ambulant participants (n=9)
  - After intervention: -4.30% ([95%CI -6.78,-1.82], p=0.001)
  - After follow-up: -2.28% ([95%CI -4.77,0.21], p=0.071)

- Non-ambulant participants (n=5)
  - After intervention: -3.71% ([95%CI -5.65,-1.76], p=0.001)
  - After follow-up: -5.28% ([95%CI -7.23,-3.34], p<0.001)

Take home messages

- Following a 12-wk U-PART intervention positive health-related effects were seen in the outcomes of sedentary time, daily step count, walking capacity and quality of life
- At short-term follow-up positive effects were maintained in sedentary time and walking capacity
- Outcome trajectories may differ according to the ambulation level of the participant

Key references

- Leonard H, Cobb S, Downs J. Clinical and biological progress over 50 years in Rett syndrome. Nat Rev Neurol 2017 Jan;13(1):37-51.
- Manns PJ, Dunstan DW, Owen N, Healy GN. Addressing the nonexercise part of the activity continuum: a more realistic and achievable approach to activity programming for adults with mobility disability? Phys Ther 2012;92(4):614-25.
- Downs J, Leonard H, Wong K, Newton N, Hill K. Quantification of walking-based physical activity and sedentary time in individuals with Rett syndrome. Dev Med Child Neurol 2017.
- Stahlhut M, Downs J, Aadahl M, Leonard H, Bisgaard AM, Nordmark E. Patterns of sedentary time and ambulatory physical activity in a Danish population of girls and women with Rett syndrome. Disabil Rehabil 2017 Oct 2;1-9.