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**Disparity in child/parent reports of quality of life among children with cerebral palsy  
persists into adolescence**

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## **ABSTRACT**

**Short title:** Child/parent reports of quality of life

### **Aim**

To examine the evolution of child/parent discrepancy in reporting quality of life between childhood and adolescence in children with cerebral palsy (CP), and to investigate potential factors associated with the discrepancy.

### **Method**

We used data from the SPARCLE project, a population-based cohort study of children with CP, aged 8-12 years at baseline (in 2004-2005), in 9 European centers and followed-up at 13-17 years. KIDSCREEN-52 QoL questionnaire was used at baseline and follow-up. 354 child/parent dyads out of 500 eligible dyads were followed-up. We used intra-class correlation coefficients (ICC) to examine agreement between parent proxy-reports and self-reported QoL, and linear regression to examine factors associated with child/parent discrepancy.

### **Results**

Agreement was low to moderate (ICC from 0.16 to 0.48) in childhood and in adolescence across all domains of QoL. In four domains (*Moods and emotions*, *Self-perception*, *Relationship with parents and home life* and *Social support and peers*), the extent of the discrepancy (child>parent) was found to increase significantly between childhood and adolescence. Parenting stress, child pain, and child behaviour problems influenced parent proxy-reports in both childhood and adolescence.

### **Interpretation**

Child and parent points of view are complementary for better knowledge of the QoL of children and adolescents with CP.

## **WHAT THIS PAPER ADDS**

- There is child/parent discrepancy in quality of life reports in both children and adolescents with cerebral palsy
- Factors associated with child/parent discrepancy are similar in childhood and adolescence
- Child pain and parenting stress are consistently associated with child/parent discrepancy in childhood and adolescence

The World Health Organization considers quality of life (QoL) to be multidimensional, covering subjective appraisal of personal feelings, social relationships, local environment, societal values, and material conditions. Therefore, assessment of a child's perception of his/her QoL is important, and should be self-reported,<sup>1</sup> using appropriate instruments now available for children.<sup>2</sup> Parents often serve as proxy reporters where it is difficult to obtain information directly from children, but self- and proxy- reports are not interchangeable.<sup>3</sup> Children and parents differ in their views about the child's QoL, primarily because they base their judgment on different information.<sup>4</sup> The literature consistently reported that parents of children with early-onset chronic conditions rate their children's QoL lower than the children themselves,<sup>4,5</sup> even though discrepancy can occur in both directions.<sup>6,7</sup> Known factors associated with discrepancy depend on the domain being measured, and the characteristics of the child and parent. Parents' perceptions are mainly affected by their mental state, with depressive symptoms or parenting stress associated with greater discrepancy in child/parent dyads of healthy children<sup>8</sup> and children with cerebral palsy (CP).<sup>7</sup> Regarding children with early-onset neurological conditions, significant pain and emotional distress are more likely to be associated with discrepancy;<sup>4,9</sup> results with regard to the impact of severity of the condition or family burden on discrepancy are less consistent.<sup>4,7,9,10</sup>

Increased age was found to be associated with greater child/parent discrepancy in healthy children,<sup>11</sup> as well as in children with chronic health conditions.<sup>10</sup> However, little is known about such discrepancy in children with CP, or about factors that might influence such discrepancy. The study of factors influencing discrepancy is especially relevant to those who cannot self-report due to learning difficulty. For instance, if higher parenting stress was found to be associated with parents' underestimation of their child's QoL, then this may influence how to interpret a parent's account of the QoL of their child who cannot self-report. Such

knowledge should contribute to improve our understanding of the differences in child's and parents' perspectives.

Our aims were to examine child/parent discrepancy in reporting QoL in childhood and adolescence in a large European sample of children with CP, and to investigate potential factors associated with the discrepancy. We hypothesized that discrepancy persists at adolescence, that its magnitude varies according to the domain explored, with more discrepancy in more subjective domains, and that the factors associated with discrepancy differ between childhood and adolescence in terms of the strengths of the relationships.

## **METHOD**

### **Study design and participants**

Participants were recruited from the SPARCLE project, which is a nine-center prospective cohort of 818 families with children with CP of all severities aged 8-12 years at baseline in 2004/2005. Children were sampled from population-based registries of children with CP in eight European regions, sharing a standardised definition and classification of CP;<sup>12</sup> an additional sample was recruited from multiple sources in Northwest Germany.<sup>13</sup> The participants were visited again aged 13-17 years in 2009/2010. The protocols are reported elsewhere.<sup>13,14</sup>

Of the 500 children who self-reported QoL, 377 (75%) participated at follow-up. For the present study, the sample was restricted to 354 child/parent dyads with complete data on at least one QoL domain at baseline and follow-up (eFigure 1). Trained researchers undertook home visits to administer questionnaires to parents and children, using the same questionnaires at baseline and follow-up. The proxy-reports were completed mostly by mothers (90.1%) at both time points.

## Measures

QoL was assessed at baseline and follow-up using the self-report and parent-proxy versions of the generic KIDSCREEN-52 questionnaire, a European instrument with strong psychometric properties, designed for 8-18-year-olds.<sup>15</sup> Both versions have the same 52 items covering ten domains of QoL: *Physical well-being*, *Psychological well-being*, *Moods and emotions*, *Self-perception*, *Autonomy*, *Relationship with parents and home life*, *Social support and peers*, *School environment*, *Social acceptance* and *Financial resources*. Raw scores are obtained by summing items in each QoL domain. Rasch person parameters are then allocated to each score, and transformed to 0–100 scale (100 indicating best QoL), with a mean score of 50 and a standard deviation (SD) of 10 in the reference general population.<sup>15</sup> We amended one item of the *Physical well-being* domain from “able to run well” to “able to get about easily” to make it suitable for children with CP.<sup>16</sup>

Potential predictors of child/parent QoL discrepancy were selected on the basis of previous results.<sup>4,7</sup> Child and family characteristics and instruments used are described in eTable 1. Children and adolescents reported their pain in the last week (none; slight to moderate; severe to very severe). Parents provided information on : severity of motor impairment as captured by the Gross Motor Function Classification System (GMFCS), intellectual impairment (Intellectual quotient (IQ)  $\leq 70$  or  $> 70$ ), communication difficulties (normal; mild to profound), emotional and behavioral symptoms using the Strengths and Difficulties Questionnaire (normal; borderline; abnormal), child’s school type (mainstream school; special classroom in mainstream school; special school; not at school), marital status (married or living with a partner; other family structure), number of children at home (none; sibling(s) but none disabled; sibling(s) with at least one disabled), and their highest educational qualification (at follow-up) categorised for analysis as high, medium and low. Parenting stress was assessed with the Parenting Stress Index Short Form (abnormal; borderline; normal).

## **Statistical analysis**

Cronbach's alphas were used to assess the reliability of child and parent reports. Analyses were stratified by KIDSCREEN domain and were performed on complete cases because there were less than 5% of missing data on covariates (Table1), and up to 6% of missing data on raw child-parent difference scores. The *Financial resources* domain was not investigated due to high ceiling effects and the high rate of missing data (around 20%).<sup>17</sup>

### ***Level of discrepancy between self- and proxy- reports of child QoL at baseline and follow-up***

The differences between the scores obtained from the children's self-reports and the parents' proxy-reports were computed, and presented as a mean with their SDs. Bland-Altman plots were used to examine graphically whether the differences in scores between parent/child varied over the range of child/parent mean score. Spearman Rho coefficients were used to test potential patterns, and paired t-tests were used to compare the level of individual child/parent agreement. Agreement was further quantified using intra-class correlation coefficients (ICC), categorised as low  $\leq 0.40$ ; moderate 0.41-0.60; good 0.61-0.80; excellent 0.81-1.00)<sup>18</sup> and estimated from a one-way ANOVA model.

To investigate the direction of child/parent discrepancy, and in the absence of widely agreed-upon minimal clinically meaningful difference for the KIDSCREEN, we considered half SD (equivalent to 5 points) as a reasonable threshold for discriminating changes,<sup>19</sup> as recommended by the KIDSCREEN manual.<sup>15</sup> We defined three categories of discrepancy: i. '*(child < parent)*' when a child rated their QoL to be more than 5 points below their parents' rating; ii. '*(child > parent)*' when a child rated their QoL to be more than 5 points above their parents' rating; iii. 'Agreement (*child ~ parent*)' when the absolute difference was less than or equal to 5 points

### ***Potential predictors of child/parent QoL discrepancy***

Bivariate associations between child/parent discrepancy and child and family characteristics were studied separately for baseline and follow-up. Factors associated with discrepancy were investigated using linear mixed models with the raw difference between child and parent scores (ranging from -100 to 100) as the dependent variable using a random intercept to take into account the repeated measures. All models controlled for region of residence as a fixed effect. Models were adjusted for the child/parent mean score when patterns were identified in the Bland-Altman plots. Marginal means in child/parent discrepancy scores (i.e. average adjusted predictions of discrepancy) were estimated from the models by stratifying each covariable using the Stata ‘Margins’ command. A test of global effects was performed for each covariable using 2-sided p-values with significance at  $p < 0.05$ . The linearity of the link between age and child/parent discrepancy was checked graphically and tested with fractional polynomials. Age\*covariable interactions were tested to determine whether the associations were stable across the age range. Bootstrapping (1000 replications) was used to compute the standard errors of the regression parameters. Assumptions of the linear mixed models and goodness of fit were checked.

### ***Sensitivity analyses***

We investigated whether it made a difference if the proxy person was the child’s mother or father.<sup>4</sup> We re-ran the final models excluding outliers with standardised residuals less than -2 or greater than +2 (between 15 and 21 depending of the QoL domain). To handle incomplete information, we used multiple imputation by chained equations,<sup>20</sup> to impute missing values of covariates considered in the linear mixed effect model, as well as the missing values on the self-reports and on the proxy-reports of QoL at both time points and in the nine QoL domains. The raw difference between child and parent reported Rasch-scores were re-estimated at each time

point, and we ran the regression models on 10 imputed datasets (354 dyads), by applying Rubin's rules.<sup>21</sup> Statistical analyses were performed using Stata 11.0 (StataCorp, College Station, TX, USA). Due to the number of comparisons, the critical value was set of 1%.

### ***Ethics***

We obtained ethical approval as appropriate to each country. Written informed consents were obtained from all parents and from the adolescents themselves whenever possible.

## **RESULTS**

The mean age of children was 10.4 years (SD 1.5 years) at baseline and 15.1 years (SD 1.5 years) at follow-up. The standardized Cronbach's  $\alpha$  of the KIDSCREEN domains at follow-up ranged from 0.70 to 0.86 for adolescents and from 0.73 to 0.93 for their parents. Cronbach's  $\alpha$  at baseline are reported elsewhere.<sup>17,22,7</sup> Child/parent pairs included in the analyses (n=354) were not significantly different from the eligible sample at baseline regarding sociodemographic and family characteristics, and child impairment, with the exception of region of residence (Table 1). Children included in the follow-up sample were less likely to report severe pain during the last week than those at baseline. No difference was found regarding QoL, except for parent-reported QoL in the *School environment* domain where the mean score was higher in dyads available for analysis compared to those not available.

### ***Level of discrepancy between self- and proxy-reports of child QoL at baseline and follow-up***

On average, parents rated their child's QoL to be worse than their child did across all domains at baseline and follow-up (Table 2). Bland-Altman plots indicated that the scatter of the child/parent discrepancy increased as the child/parent mean score increased in two QoL domains at baseline (*Physical well-being* and *School environment*); no pattern was found at

follow-up (plots not shown). Spearman Rho coefficients showed a poor correlation ( $<0.25$ ) between child/parent discrepancy and child/parent mean scores; paired t-tests indicated that parents and children reported significantly different levels of QoL for all nine domains at baseline and follow-up ( $p<0.001$ ). The level of agreement (ICC) between QoL scores from self- and parent reports were low to moderate across all domains and at baseline and follow-up (ICC ranged from 0.16 in *Autonomy* to 0.41 in *Social acceptance* at baseline and from 0.20 in *Moods and emotions* to 0.47 in *School environment* at follow-up) (Table 2).

### ***Evolution of discrepancy between childhood and adolescence***

The percentage of dyads with the category ‘Agreement (*child~parent*)’ was 43% at baseline and 41% at follow-up for the *Social acceptance* domain. Percentages of dyads with discrepancy category ‘(*child>parent*)’ were highest in most of QoL domains at baseline and follow-up (except *Social acceptance* domain at baseline); rates ranged from 37% in *Social acceptance* at baseline to 53% in *Self-perception* at follow-up (Figure 1). To explore consistency in discrepancy over time, we plotted the three categories at baseline and follow-up (Figure 2). In those with category ‘Agreement (*child~parent*)’ at baseline, 29% (*Social support and peers*) to 49% (*Social acceptance*) still agreed at follow-up (percentages not shown). In the multivariate mixed model (eTable 2), the mean of child/parent discrepancy was significantly higher when the child was older in four QoL domains: *Moods and emotions*, *Self-perception*, *Relations with parents and home life* and *Social acceptance*.

### ***Factors associated with child/parent discrepancy in childhood and adolescence***

The factors associated with child/parent discrepancy are shown in eTable 2.

No interaction with the age was found to be significant, suggesting that the association between co-variables and child/parent discrepancy was not modified by the child age. Larger

discrepancy in child/parent reports of domain *Physical well-being* was significantly associated with more severe motor impairment ( $p=0.001$ ); and the domain *School environment* and *Social acceptance* with child behaviour problems (respectively,  $p=0.005$  and  $p<0.001$ ). Having pain significantly reduced the differences between child/parent ratings in the domains of *Psychological well-being* ( $p<0.05$ ), *Autonomy* ( $p<0.01$ ) and *School environment* ( $p<0.05$ ). Higher parenting stress significantly related to a greater discrepancy in the domains *Physical well-being* ( $p<0.05$ ), *Psychological well-being* ( $p<0.01$ ) and *Moods and emotions* ( $p<0.05$ ). Lastly, the child/parent mean score of QoL (added into the models on *Physical well-being* and *School environment* because of evidence of patterns in the Bland-Altman plots) was significantly related to child/parent discrepancy: the higher was the average score, the higher was the discrepancy for *Physical well-being* ( $p<0.001$ ) and *School environment* ( $p=0.001$ ).

### ***Sensitivity analyses***

Results of the multivariate quantitative analysis did not differ according to whether the reporting proxy was the mother or the father. However, the mean child/parent discrepancy was significantly lower in *Social support and peers* if the reporting person was neither the child's mother nor father, but another person well-known to the child. Outliers did not change the conclusion. Overall, analyses on the ten imputed datasets for complete child/parent dyads at childhood or at adolescence ( $n=354$ ) resulted in similar findings. Motor impairment was associated with discrepancies in *Autonomy* ( $p=0.002$ ). Age was significant at the 5% level ( $p=0.017$ ) in *Self-perception*, but pain was significant at the 1% level ( $p=0.005$ ) in *Psychological well-being* and parenting stress significant in *Physical well-being* ( $p=0.005$ ) and *Moods and emotions* ( $p=0.008$ ).

## **DISCUSSION**

Discrepancy between self- and parent-proxy-reported QoL persisted from childhood to adolescence for children with CP, with parents giving lower ratings, on average, than children across all domains. ICC values ranged from 0.16 to 0.48 in both age groups; only two domains, *School environment* and *Social acceptance*, attained an ICC of 0.35 or over. The extent of the discrepancy (*child > parent*) increased in adolescence in four domains (*Moods and emotion*, *Self-perception*, *Relationships with parents and home life* and *Social acceptance*). The association of child and family characteristics with child/parent discrepancy were similar in both age groups.

Our results are consistent with previous literature that reported that parents of children with chronic conditions often underestimate their child's QoL,<sup>23,4,24-26,7</sup> but few previous studies have investigated the questions in both childhood and adolescence.<sup>27,11</sup> Adolescence is a period of rapid biological, social and emotional change that may affect child/parent relationships. Parental judgment on domains related to their child's feelings and social experience might be distorted during this time, leading to further child/parent discrepancy. The large increase in discrepancy between childhood and adolescence for *Moods and emotion* may be explained by the increased independence and greater difficulties in expressing emotions and feelings in adolescence.<sup>4</sup> In contrast, for the domains of *Physical well-being*, *Autonomy* and *School environment*, which may reflect more factual and objective functioning, discrepancy was unchanged.

A large child/parent discrepancy, being stable over time, was observed for the *Physical well-being* and *Autonomy* domains which have been defined as measuring, respectively, "the level of the child's physical activity, energy and fitness", and "the respondents' opportunities to shape their social and leisure time".<sup>28</sup> Large differences are also reported in such domains between 8-18-year-old children with autism spectrum disorder and their parents.<sup>29</sup> The

interpretation of these results should be in light of the QoL instrument used (e.g. generic or condition-specific instrument) and the potential differences in the conceptualisation of physical well-being.<sup>30</sup> Both in childhood and adolescence, the severity of the impairment (in terms of motor function and behaviour problems) increased discrepancy in at least one QoL domain, with parents underestimating their child's QoL. Our analysis also suggests that child/parent discrepancies were reduced when pain was reported, while higher level of parenting stress related to greater discrepancies in some domains. Severe pain in particular is likely to be appreciated by both child and parent; higher levels of parenting stress may have adverse effect on the parent/child relationship and therefore lead to greater child/parent discrepancy.

Quality of life is an important health outcome. Proxy-report instruments have been developed to assess a child's QoL in circumstances where self-report is not possible (e.g. younger age, or in presence of cognitive impairment). Our findings confirm a lack of agreement in report of a child's QoL between the children themselves and their parents. Whilst a parent's perception of their child's QoL is important, we have also shown that parents overestimated their child's QoL when the child had much pain, and that parents' own difficulties led them to underestimate their child's QoL. Given that we can never know how a child who cannot self-report views their QoL, we argue that the same factors that appear to influence how a parent reports the QoL of a child who can self-report may also influence how a parent reports the QoL of a child who cannot self-report. Innovative approaches are needed to develop parent proxy-report instruments that incorporate recognition that parents can never be totally objective observers and that their own stress levels may influence how they report their child's QoL.

The major strength of our study is the large sample size and the data collection at two time points. Moreover, statistical analysis was based on a clear definition of discrepancy. Reasons

for loss to follow-up between childhood and adolescence were evaluated and did not appear to introduce bias. In accordance to the general purpose of the SPARCLE study to examine the QoL in children with CP compared to the general population, we used a generic instrument to assess QoL,<sup>2</sup> with both child and parent versions available, which has demonstrated ability to detect change over time.<sup>31</sup> Finally, a sensitivity analysis, with full dyads analysis after multiple imputation of missing data, confirmed our results.

In conclusion, our study showed an increase in child/parent discrepancy in reporting of the child's in some domains of QoL. Child pain was associated with lower discrepancy, while the severity of the disability and parenting stress was associated with higher discrepancy in childhood as well as in adolescence.

## REFERENCES

- 1 Wallander JL, Koot HM. Quality of life in children: A critical examination of concepts, approaches, issues, and future directions. *Clinical Psychology Review* 2016; **45**: 131-43.
- 2 Fayed N, de Camargo OK, Kerr E, et al. Generic patient-reported outcomes in child health research: a review of conceptual content using World Health Organization definitions. *Dev Med Child Neurol* 2012; **54**: 1085-95.
- 3 Pickard AS, Knight SJ. Proxy evaluation of health-related quality of life: a conceptual framework for understanding multiple proxy perspectives. *Med Care* 2005; **43**: 493-9.
- 4 Eiser C, Varni JW. Health-related quality of life and symptom reporting: similarities and differences between children and their parents. *Eur J Pediatr* 2013; **172**: 1299-304.
- 5 Upton P, Lawford J, Eiser C. Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Qual Life Res* 2008; **17**: 895-913.
- 6 Sattoe JN, van Staa A, Moll HA. The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. *Health Qual Life Outcomes* 2012; **10**: 10.
- 7 White-Koning M, Arnaud C, Dickinson HO, et al. Determinants of child-parent agreement in quality-of-life reports: a European study of children with cerebral palsy. *Pediatrics* 2007; **120**: e804-14.
- 8 Kobayashi K, Kamibeppu K. Quality of life reporting by parent-child dyads in Japan, as grouped by depressive status. *Nurs Health Sci* 2011; **13**: 170-7.
- 9 Morrow AM, Hayen A, Quine S, Scheinberg A, Craig JC. A comparison of doctors', parents' and children's reports of health states and health-related quality of life in children with chronic conditions. *Child Care Health Dev* 2012; **38**: 186-95.
- 10 Fayed N, Avery L, Davis AM, et al. Parent Proxy Discrepancy Groups of Quality of Life in Childhood Epilepsy. *Value in Health* 2019; **22**: 822-8.
- 11 Rajmil L, Lopez AR, Lopez-Aguila S, Alonso J. Parent-child agreement on health-related quality of life (HRQOL): a longitudinal study. *Health Qual Life Outcomes* 2013; **11**: 101.
- 12 Johnson A. Prevalence and characteristics of children with cerebral palsy in Europe. *Dev Med Child Neurol* 2002; **44**: 633-40.
- 13 Colver A. Study protocol: SPARCLE--a multi-centre European study of the relationship of environment to participation and quality of life in children with cerebral palsy. *BMC Public Health* 2006; **6**: 105.
- 14 Colver AF, Dickinson HO. Study protocol: determinants of participation and quality of life of adolescents with cerebral palsy: a longitudinal study (SPARCLE2). *BMC Public Health* 2010; **10**: 280.
- 15 KIDSCREEN Group. The KIDSCREEN questionnaires—quality of life questionnaires for children and adolescents. Handbook. *Lengerich: Pabst Science Publishers* 2006.
- 16 Erhart M, Ravens-Sieberer U, Dickinson HO, Colver A. Rasch measurement properties of the KIDSCREEN quality of life instrument in children with cerebral palsy and differential item functioning between children with and without cerebral palsy. *Value Health* 2009; **12**: 782-92.
- 17 Arnaud C, White-Koning M, Michelsen SI, et al. Parent-reported quality of life of children with cerebral palsy in Europe. *Pediatrics* 2008; **121**: 54-64.
- 18 Koch GG, Landis JR, Freeman JL, Freeman DH, Jr., Lehnen RC. A general methodology for the analysis of experiments with repeated measurement of categorical data. *Biometrics* 1977; **33**: 133-58.

- 19 Norman GR, Sloan JA, Wyrwich KW. Interpretation of changes in health-related quality of life: the remarkable universality of half a standard deviation. *Med Care* 2003; **41**: 582-92.
- 20 Royston P. Multiple imputation of missing values: further update of ice, with an emphasis on categorical variables. *The Stata Journal* 2009; **9**: 466-77.
- 21 Stine R. An introduction to bootstrap methods: Examples and ideas. *Sociological Methods & Research* 1989; **18**: 243-91.
- 22 Dickinson HO, Parkinson KN, Ravens-Sieberer U, et al. Self-reported quality of life of 8-12-year-old children with cerebral palsy: a cross-sectional European study. *Lancet* 2007; **369**: 2171-8.
- 23 Danckaerts M, Sonuga-Barke EJ, Banaschewski T, et al. The quality of life of children with attention deficit/hyperactivity disorder: a systematic review. *Eur Child Adolesc Psychiatry* 2010; **19**: 83-105.
- 24 Galloway H, Newman E. Is there a difference between child self-ratings and parent proxy-ratings of the quality of life of children with a diagnosis of attention-deficit hyperactivity disorder (ADHD)? A systematic review of the literature. *Atten Defic Hyperact Disord* 2017; **9**: 11-29.
- 25 Houwen-van Opstal SL, Jansen M, van Alfen N, de Groot IJ. Health-related quality of life and its relation to disease severity in boys with Duchenne muscular dystrophy: satisfied boys, worrying parents--a case-control study. *J Child Neurol* 2014; **29**: 1486-95.
- 26 Vetter TR, Bridgewater CL, McGwin G, Jr. An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? *Health Qual Life Outcomes* 2012; **10**: 85.
- 27 Murray CB, Holmbeck GN, Ros AM, Flores DM, Mir SA, Varni JW. A longitudinal examination of health-related quality of life in children and adolescents with spina bifida. *J Pediatr Psychol* 2015; **40**: 419-30.
- 28 Ravens-Sieberer U, Gosch A, Rajmil L, et al. KIDSCREEN-52 quality-of-life measure for children and adolescents. *Expert Rev Pharmacoecon Outcomes Res* 2005; **5**: 353-64.
- 29 Egilson ST, Olafsdottir LB, Leosdottir T, Saemundsen E. Quality of life of high-functioning children and youth with autism spectrum disorder and typically developing peers: Self- and proxy-reports. *Autism* 2017; **21**: 133-41.
- 30 Makris T, Dorstyn D, Crettenden A. Quality of life in children and adolescents with cerebral palsy: a systematic review with meta-analysis. *Disability and rehabilitation* 2019: 1-10.
- 31 Palacio-Vieira JA, Villalonga-Olives E, Valderas JM, et al. Changes in health-related quality of life (HRQoL) in a population-based sample of children and adolescents after 3 years of follow-up. *Qual Life Res* 2008; **17**: 1207-15.

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## TABLES

**Table 1** Impairments, socio-demographic and family characteristics in those available for analysis and those not available

	Available for analysis (n=354)		Not available for analysis (n=145) Baseline n(%)	P-value Chi-square <sup>a</sup>
	Baseline n(%)	Follow-up n(%)		
<b>SOCIO-DEMOGRAPHIC CHARACTERISTIC</b>				
<b>Region of residence</b>				
UK				< 0.001
North England	52 (14.7)		26 (17.9)	
Northern Ireland	53 (15.0)		5 (3.5)	
Southwest Ireland	46 (13.0)		21 (14.5)	
West Sweden	36 (10.2)		10 (6.9)	
East Denmark	48 (13.6)		33 (22.8)	
Northwest Germany	39 (11.0)		6 (4.1)	
France				
Southeast France	31 (8.7)		7 (4.8)	
Southwest France	33 (9.3)		13 (9.0)	
Central Italy	16 (4.5)		24 (16.5)	
Missing	0 (0.0)		0 (0.0)	
<b>Child's sex</b>				
Male	201 (56.8)		85 (58.6)	0.71
Female	153 (43.2)		60 (41.4)	
Missing	0 (0.0)		0 (0.0)	
<b>Child's school setting</b>				
Mainstream school	212 (59.9)	129 (36.9)	78 (54.5)	0.05
Mainstream school and visits special unit	38 (10.7)	100 (28.6)	27 (18.9)	
Special unit/Special school	104 (29.4)	119 (34.0)	38 (26.6)	
Not at school	0 (0.00)	2 (0.5)	0 (0.0)	
Missing	0 (0.0)	4 (1.1)	2 (1.4)	
<b>IMPAIRMENT</b>				
<b>GMFCS</b>				
I - Child walks and climbs stairs	145 (41.0)	171 (48.3)	73 (50.3)	0.06
II - Child walks inside	92 (26.0)	67 (18.9)	26 (17.9)	
III- Child walks with limitations	61 (17.2)	46 (13.0)	32 (22.1)	
IV - Moving about is limited	39 (11.0)	37 (10.5)	10 (6.9)	
V - Moving about is severely limited	17 (4.8)	33 (9.3)	4 (2.8)	
Missing	0 (0.0)	0 (0.0)	0 (0.0)	
<b>Intellectual impairment</b>				
IQ >70	257 (73.2)	248 (70.1)	105 (72.4)	0.85
IQ ≤ 70	94 (26.8)	106 (29.9)	40 (27.6)	
Missing	3 (0.8)	0 (0.0)	0 (0.0)	

<b>Self-reported frequency and severity of pain in previous week</b>				
No pain	160 (46.2)	105 (29.9)	57 (40.1)	0.03
Slight/moderate	127 (36.7)	154 (43.9)	46 (32.4)	
Severe/very severe	59 (17.1)	92 (26.2)	39 (27.5)	
Missing	8 (2.3)	3 (0.8)	3 (2.1)	
<b>Communication difficulties</b>				
Normal communication	288 (81.4)	297 (84.4)	120 (82.8)	0.71
Mild to profound impairment	66 (18.6)	55 (15.6)	25 (17.2)	
Missing	0 (0.0)	2 (0.6)	0 (0.0)	
<b>SDQ, Total difficulties score, parent report</b>				
Normal	231 (65.3)	229 (64.9)	86 (59.3)	0.45
Borderline	50 (14.1)	57 (16.1)	23 (15.9)	
Abnormal	73 (20.6)	67 (19.0)	36 (24.8)	
Missing	0 (0.0)	1 (0.3)	0 (0.0)	
<b>FAMILY CHARACTERISTIC</b>				
<b>Family structure/ marital status</b>				
Married or living with partner	290 (81.9)	282 (79.9)	116 (80.0)	0.62
Single	64 (18.1)	71 (20.1)	29 (20.0)	
Missing	0 (0.0)	1 (0.3)	0 (0.0)	
<b>Siblings in the same household</b>				
None	59 (16.9)	68 (19.5)	27 (19.0)	
Yes, none disabled	246 (70.7)	235 (67.3)	100 (70.4)	
Yes, some disabled	43 (12.4)	46 (13.2)	15 (10.6)	
Missing	6 (1.7)	5 (1.4)	3 (2.1)	
<b>Parental qualification</b>				
Undergraduate or postgraduate degree	-	62 (17.7)	8 (36.4)	0.09 F
Upper secondary or vocational education	-	227 (64.6)	10 (45.4)	
Primary or lower secondary education	-	62 (17.7)	4 (18.2)	
Missing	-	3 (0.8)	123 (84.8)	
<b>Parenting stress, Total score</b>				
Normal (<72)	134 (38.1)	137 (39.4)	50 (36.8)	0.31
Borderline (72-90)	137 (38.9)	112 (32.2)	46 (33.8)	
Abnormal (>90)	81 (23.0)	99 (28.4)	40 (29.4)	
Missing	2 (0.6)	6 (1.7)	9 (6.2)	

<sup>a</sup> comparing available versus not available on baseline data

Abbreviations: F: Fisher exact test; GMFCS: Gross Motor Function Classification System; SDQ: Strength and difficulties questionnaire

**Table 2** - Child and parent reports of QoL domains and mean discrepancy, in childhood and adolescence

Domain	N	Childhood (baseline)			Adolescence (follow-up)			Discrepancy mean (SD) <sup>a</sup>	
		Mean (SD)		ICC <sup>b</sup>	Mean (SD)		ICC <sup>b</sup>	Childhood (baseline)	Adolescence (follow-up)
		Child report	Parent report		Child report	Parent report			
Physical well-being	332	51.0 (11.8)	44.0 (9.5)	0.24	49.3 (9.8)	43.2 (9.3)	0.26	7.0 (11.8)	6.1 (10.7)
Psychological well-being	344	51.5 (9.3)	48.0 (9.4)	0.22	48.5 (9.1)	44.2 (9.7)	0.28	3.5 (11.4)	4.3 (10.8)
Moods and emotions	332	52.1 (10.2)	49.6 (9.7)	0.27	51.7 (10.0)	45.6 (10.1)	0.20	2.5 (11.8)	6.1 (11.8)
Self-perception	332	53.6 (10.3)	49.7 (9.7)	0.26	51.5 (10.1)	45.2 (9.2)	0.20	3.9 (12.0)	6.3 (10.9)
Autonomy	339	50.1 (10.0)	44.5 (8.9)	0.16	51.1 (9.6)	45.8 (10.7)	0.23	5.6 (11.6)	5.3 (11.9)
Relationships with parents and home	332	52.1 (9.3)	49.5 (10.0)	0.23	51.3 (9.4)	45.4 (9.9)	0.23	2.6 (11.9)	5.9 (11.2)
Social support and peers	320	48.6 (12.6)	42.8 (11.4)	0.34	46.8 (12.4)	39.4 (12.7)	0.30	5.8 (13.0)	7.5 (13.8)
School environment	336	56.0 (11.5)	52.1 (9.9)	0.38	53.3 (10.1)	50.0 (10.2)	0.48	3.9 (11.6)	3.4 (10.0)
Social acceptance	327	49.2 (11.1)	46.4 (11.5)	0.41	50.7 (10.2)	46.1 (11.9)	0.35	2.8 (12.1)	4.7 (12.0)

<sup>a</sup>All paired comparisons were significant,  $p < 0.001$ .

<sup>b</sup> estimated from a one-way ANOVA model

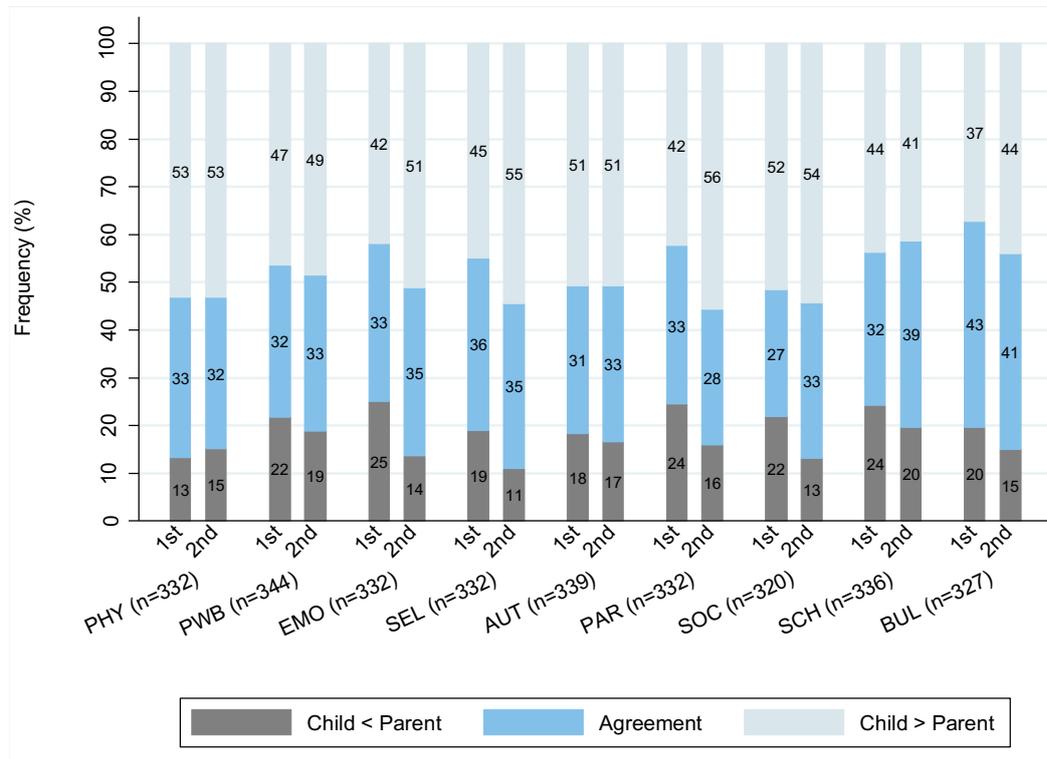
Abbreviations: ICC: intra-class correlation coefficients; SD: Standard Deviation

## **FIGURE LEGENDS**

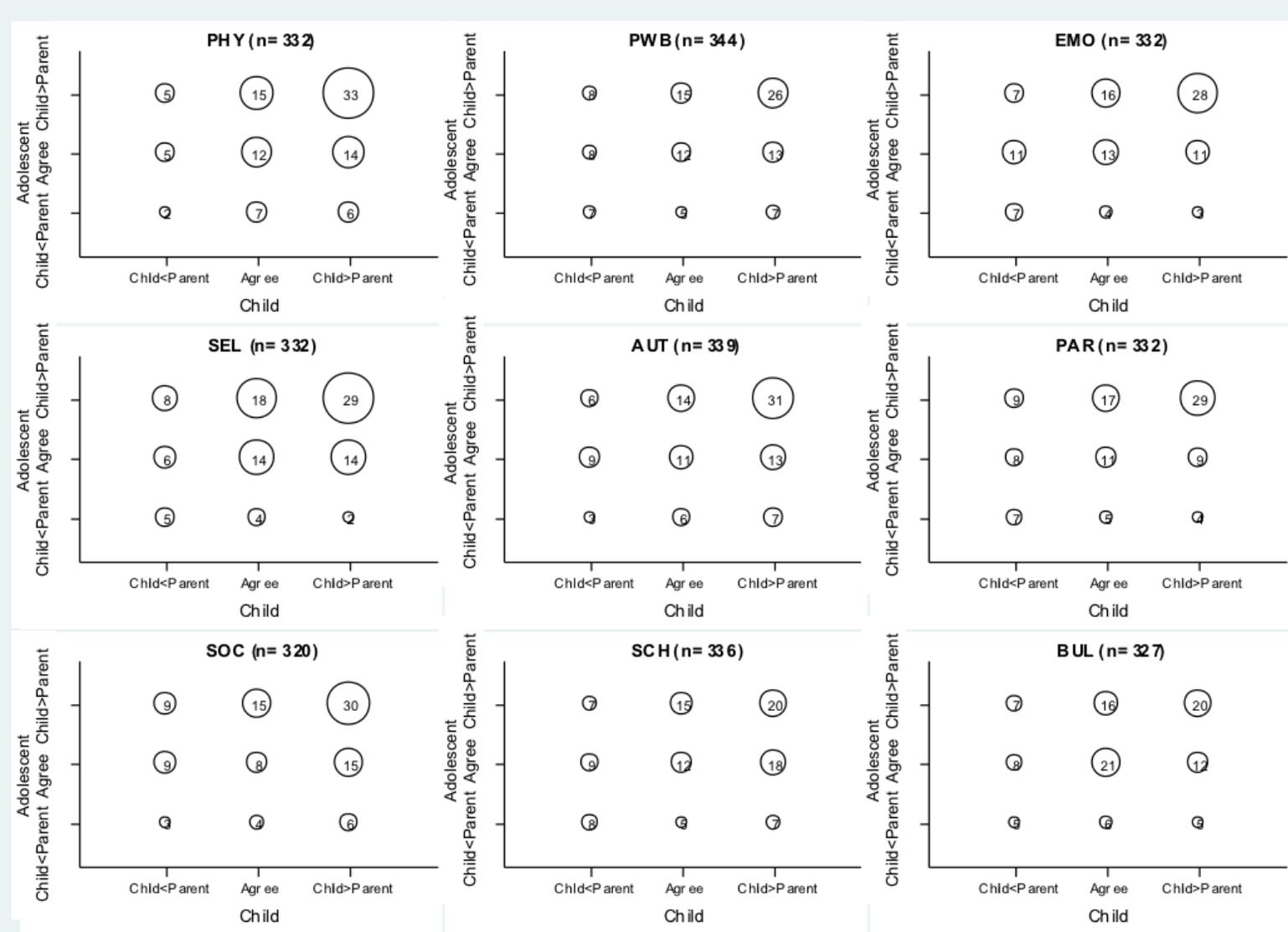
**Figure 1** - Distribution of discrepancy between child/parent reports (% of complete pairs) in childhood and adolescence

**Figure 2** - Individual change in child/parent discrepancy between childhood (at baseline) and adolescence (at follow-up)

**Figure 1** - Distribution of discrepancy between child/parent reports (% of complete pairs) in childhood and adolescence



Abbreviation: Physical well-being (PHY), Psychological well-being (PWB), Moods and emotions (EMO), Self-perception (SEL), Autonomy (AUT), Relationship with parents and home life (PAR), Social support and peers (SOC), School environment (SCH), and Social acceptance (BUL).



Note: the circle size is proportionate to the number of dyads

Abbreviations: Physical well-being (PHY), Psychological well-being (PWB), Moods and emotions (EMO), Self-perception (SEL), Autonomy (AUT), Relationship with parents and home life (PAR), Social support and peers (SOC), School environment (SCH), and Social acceptance (BUL).

Example of interpretation: 33% of the 332 dyads with complete data on physical well-being domain of quality of life (PHY), belong to the discrepancy category ‘Positive agreement (child>parent)’ in childhood (at baseline) and in adolescence (at follow-up)”