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Predictors of parent-reported quality of life of adolescents with cerebral palsy: A longitudinal study

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ABSTRACT

Aim: Parent-reporting is needed to examine Quality of Life (QoL) of children with cerebral palsy (CP) across all severities. This study examines whether QoL changes between childhood and adolescence, and what predicts adolescent QoL.

Method: SPARCLE is a European cohort study of children with CP, randomly sampled from population databases. Of 818 8–12-year-olds joining the study, 594 (73%) were revisited as 13–17-year-olds. The subject of this report is the 551 (316 boys, 235 girls) where the same parent reported QoL on both occasions using KIDSCREEN-52 (transformed Rasch scale, mean 50, SD 10 per domain). Associations were assessed using linear regression.

Results: Between childhood and adolescence, average QoL reduced in six domains (1.3–3.8 points, $p < 0.01$) and was stable in three (*Physical wellbeing, Autonomy, Social acceptance*). Socio-demographic factors had little predictive value. Childhood QoL was a strong predictor of all domains of adolescent QoL. Severe impairments of motor function, IQ or communication predicted higher adolescent QoL on some domains; except that severe motor impairment predicted lower adolescent QoL on the *Autonomy* domain. More psychological problems and higher parenting stress in childhood and their worsening by adolescence predicted lower QoL in five and eight domains respectively; contemporaneous pain in seven domains. The final model explained 30%–40% of variance in QoL, depending on domain.

Interpretation: In general, impairment severity and socio-demographic factors were not predictors of lower adolescent QoL. However, pain, psychological problems and parenting stress were predictors of lower adolescent QoL in most domains. These are modifiable factors and addressing them may improve adolescent QoL.

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Abbreviations: CP, Cerebral Palsy; GMFCS, Gross Motor Function Classification System; PSI, Parenting Stress Index Short Form; QoL, Quality of Life; SDQ, Strength and Difficulties Questionnaire; SPARCLE, Study of PARTICipation of Children with cerebral palsy Living in Europe.

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What this paper adds

- There was a small reduction in average QoL between the childhood and adolescence of individuals with cerebral palsy.
- Child quality of life predicted adolescent quality of life on all domains.
- Severity of impairment and socio-demographic factors had little predictive value.
- Contemporaneous pain predicted lower adolescent quality of life.
- The modifiable factors psychological problems and parenting stress in childhood and their worsening predicted lower adolescent quality of life.

1. Introduction

Quality of life (QoL) is defined as ‘the individual’s perception of their position in life in the context of the culture and value system in which they live, and in relation to their goals, expectations, standards, and concerns’ (WHOQOL, 1995). Thus, QoL is a subjective concept and should be self-reported whenever possible. Capturing such perspectives is a key expectation of Articles 13 and 14 of the UN Convention on the Rights of the Child (United Nations, 1989).

Cerebral palsy (CP) can serve as an exemplar condition of disability (Janssen, Voorman, Becher, Dallmeijer, & Schuengel, 2010; Rosenbaum, Livingston, Palisano, Galuppi, & Russell, 2007; Young et al., 2010) because the severity of CP and the patterns of its motor involvement and associated impairments show much variation and persist across the life course (Nystrand, Beckung, Dickinson, & Colver, 2014).

Children and adolescents with CP who can self-report do not cover all those with CP. Usually those with severe intellectual impairment cannot self-report, and therefore proxy reports are needed to describe QoL (Arnaud, White-Koning, & Michelsen, 2008; Rosenbaum et al., 2007; Young et al., 2010). The proxy should be close to the child or adolescent (Pickard and Knight, 2005). Although a child’s and their parents’ view of their child’s QoL differ, (White-Koning, Arnaud, & Dickinson, 2007) parent reports are closer to those of their children than those of teachers (White-Koning, Grandjean, Colver, & Arnaud, 2008), or therapists (Pickard and Knight, 2005). Interpretation of parent-proxy reports requires information about child factors like impairments (Badia, Riquelme, & Orgaz, 2014), pain or psychological problems and parent factors like stress or life circumstances (Davis, Davies, Waters, & Priest, 2008; Pickard and Knight, 2005; White-Koning et al., 2007). As there are differences between mother and father responses, longitudinal studies should ensure the continuity of one respondent (Davis et al., 2008).

Little is known about the QoL of children with CP across all severity groups as they move from childhood to adolescence. The few studies undertaken have methodological weaknesses: a short time span (van Buuren, 2007; Livingston and Rosenbaum, 2008), a wide age frame from toddlers to adults (Vargus-Adams, 2006), small sample sizes (Janssen et al., 2010) or reporting a mixture of self- and proxy reports in one analysis (Livingston and Rosenbaum, 2008; Vargus-Adams, 2006).

The ongoing Study of Participation of Children with cerebral palsy Living in Europe (SPARCLE) is therefore the first large multi-centre European cohort study of children with cerebral palsy.

The objectives of this report are to examine, whether:

- 1 parent-reported QoL of adolescents with CP changes as young people move from childhood to adolescence
- 2 QoL in adolescence is predicted by:

- a) childhood factors (impairment, pain, psychological problems),
- b) family and personal factors of the reporting parent (socio-economic and parenting stress),
- c) changes in pain, psychological problems and parenting stress between childhood and adolescence.

2. Method

2.1. Study design and participants

The methods of the SPARCLE study, described in detail elsewhere (Colver, 2006; Colver and Dickinson, 2010), are summarised below. Children born between 31/07/1991 and 01/04/1997 were randomly sampled from population-based registers of children with CP in eight European regions that share a standardised definition of CP. 743 (63%) of 1174 target families identified from registers joined the study. One further region (Northwest Germany) ascertained 75 cases from multiple sources. The 818 children who entered the study were interviewed in 2004/5 aged 8–12 years (SPARCLE1), and then again in 2009/10 aged 13–17 years (SPARCLE2) when 594 (73%) remained in the study. Drop out was associated with lower parental educational qualification, higher parenting stress and not being married at time 1 (Dickinson, Rapp, & Arnaud, 2012). These factors were introduced to our regression models.

The 551 (67%) children, where the same parent reported QoL in SPARCLE1 and SPARCLE2, comprised the longitudinal sample of this report (Fig. 1).

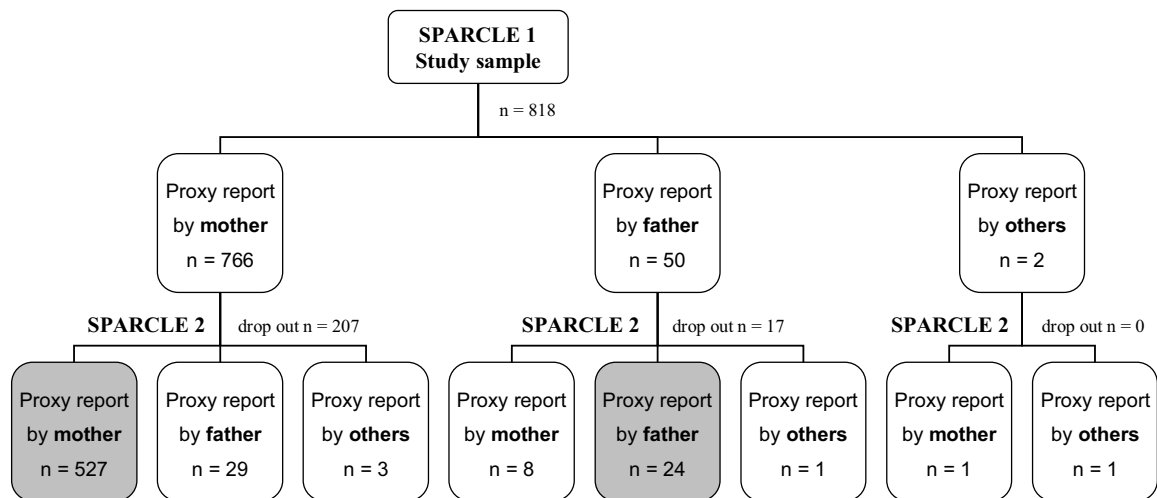


Fig. 1. Flowchart of the SPARCLE 1 (aged 8–12 years) and SPARCLE 2 (aged 13–17 years) study sample by proxy reporting person.

Table 1
Description of each KIDSCREEN domain.

KIDSCREEN domain	No. of items	Measures perceptions of these aspects of life:
Physical wellbeing	5	Physical activity, energy and fitness.
Psychological wellbeing	6	Positive emotions and satisfaction with life.
Moods and emotions	7	Negative moods, boredom and stress.
Self-perception	5	Self, bodily appearance and body-image.
Autonomy	5	Freedom of choice and self-determination in leisure time.
Relationships with parents	6	Interactions and relationships with parents and the socio-emotional atmosphere at home.
Social support and peers	6	Social support available from friends and peers.
School life	6	Learning and feelings about school and teachers.
Financial resources	3	Adequacy of pocket money relative to peers.
Social acceptance	3	Social acceptance or rejection by peers, including bullying.

2.2. Procedures

Trained researchers visited families in their homes to administer questionnaires to parents using the same questionnaires in childhood and adolescence (Colver, 2006; Colver and Dickinson, 2010).

Parents reported their child's QoL using KIDSCREEN-52 long version, a European instrument designed for children and young people (Ravens-Sieberer, Gosch, & Rajmil, 2005). It has 52 items which ask about QoL in the previous week across ten domains (Table 1), and has strong psychometric properties. Items are scored on a 5-point scale. Domain scores are transformed to Rasch person parameters using an algorithm which gives children in the reference population a mean score of 50 with a SD of 10; higher scores indicate better QoL (Ravens-Sieberer et al., 2005). We amended one item on the *Physical wellbeing* domain from "able to run well" to "able to get about easily" to make it suitable for young people with CP (Erhart, Ravens-Sieberer, Dickinson, & Colver, 2009).

Parents provided information about their child's impairments (walking ability as captured by the gross motor function classification system (GMFCS) (Palisano et al., 1997), bilateral fine motor function (BFMF) (Beckung and Hagberg, 2002), seizures, feeding, communication, intellectual ability), family structure and parents' educational qualifications.

Parents reported their child's pain during the previous four weeks in childhood and during the previous week in adolescence. The different time frames were because we did not immediately appreciate that the pain questions (which we took from the Child Health Questionnaire (Landgraf, Abetz, & Ware, 1996)) use a time frame of the past 4 weeks; whereas KIDSCREEN uses the past one week to capture quality of life. Therefore for the visits in adolescence we changed the recall period for the pain questions to one week. The pain severity is reported on a six option scale which we then converted to three levels: No pain (which corresponds to None or Very mild in the CHQ), Mild/moderate pain (which corresponds to Mild or Moderate in the CHQ) and More severe pain (which corresponds to Severe or Very severe in the CHQ).

Parents completed at both time points the Strength and Difficulties Questionnaire (SDQ) (Goodman, 2001). Responses to the SDQ yield a Total Difficulties Score at both ages (8–12 and 13–17) (range 0–40; clinical problems >16, SD in general population 7). We regarded these scores and their difference as a characteristic of the young person with cerebral palsy in our regression models. The SDQ also yields an Impact on Family score (range 0–10; abnormal >= 2) (Goodman, 2001) and we used this as a family factor in the regression models. Parents also completed the Parenting Stress Index Short Form (PSI). We used the 'Total stress score' (range 40–140; clinical stress > 90; SD in general population 15) as a measure of the clinical

stress of the parent concerning parenting and the 'Life stress scale' which captures recent life events experienced by the family causing stress during the last year (range 0–19; high ≥ 2 events) (Abidin, 1995).

2.3. Statistical methods

2.3.1. Psychometric properties

We first examined the psychometric properties (floor and ceiling effects) of the KIDSCREEN scores. We then assessed the change in QoL between childhood and adolescence using paired *t*-tests.

2.3.2. Multiple imputation

Although the overall proportion of missing values was low, approximately half of the 551 children had at least one missing value in one of the investigated characteristics in either SPARCLE 1 or 2. In order to retain all children in the linear regression analyses, we generated ten imputed datasets with no missing values using multiple imputation with chained equations (van Buuren, 2007; van Buuren and Groothuis-Oudshoorn, 2011). We imputed missing values of impairment, QoL, pain, SDQ and PSI from observed values of the same variables and age, gender, region, walking ability, family structure and parental educational qualification. For the imputation we used polytomous regression for categorical variables and predictive mean matching for interval scaled variables. The point estimates and 95% and 99% confidence intervals (CI) of the regression coefficients were obtained using Rubin's rules (Rubin, 1987).

2.3.3. Variables

We undertook analyses separately for each of the ten KIDSCREEN domains. Pain, impairment, family factors and the impact scale were treated as categorical variables. The SDQ, PSI and Life stress scale scores are presented for clarity as categorical data in Table 2; however, these scores (and their changes for SDQ and PSI) were treated as continuous variables in the analyses.

2.3.4. Regression models

All the models are adjusted for region (random effect) and GMFCS (which had determined the sampling strategy), and for gender and age which are known correlates of QoL of adolescents (Bisegger, Cloetta, von Rueden, Abel, & Ravens-Sieberer, 2005).

(a) Baseline model. In this model we used childhood QoL as a potential predictor of adolescent QoL. We treated age as a continuous variable and region, gender and GMFCS as categorical variables. No variable selection was performed for these variables.

Following estimation of this baseline model, the influences on QoL in adolescence of child factors and parent/family factors were investigated in separate models. The additional variables, including an age*gender interaction, were subject to variable selection (see 2.3.5 below).

(b) Child model. In this model we used child factors (impairment, pain in childhood and adolescence, SDQ scores in childhood and their changes between childhood and adolescence) as potential predictors of adolescent QoL.

(c) Parent/family model. In this model we used parent/family factors (impact of child's psychological problems on the family, Life stress scale, PSI scores in childhood and its changes between childhood and adolescence) as potential predictors of adolescent QoL.

(d) Final combined model. In this model we selected variables from (b) and (c). We constructed a combined model in which all the pre-selected child and parent/family factors were included as potential predictors. b-coefficients of regression indicate the change in adolescent QoL score for a change of one point in SDQ, PSI or Life stress scale.

2.3.5. Variable selection

We used stepwise backward variable selection. In order to make the selection more robust, we used bootstrapping—ten bootstrap data sets were drawn from each of the ten imputed data sets (Stine, 1990). The variable selection was based on the model's goodness of fit (Akaike's Information Criterion), not on statistical significance. This criterion selects variables that are not necessarily statistically significant.

2.3.6. Statistical significance

The criterion for statistical significance was that the 95% CIs did not include zero. In our interpretation we also took into account 99% CIs and the findings of sensitivity analyses.

2.3.7. Sensitivity analyses

We undertook two sensitivity analyses. The first included those who had dropped out. The second included those with different parent proxies at childhood and adolescence.

The statistical software R with the package mice for multiple imputation was used for analysis (van Buuren and Groothuis-Oudshoorn, 2011; R Development Core Team, 2014).

Table 2
Characteristics of the study population (n = 551).

Socio-demographic characteristics	Longitudinal sample n = 551 (%)	
	Childhood	Adolescence
Region of residence		
Denmark: East Denmark	72 (13)	
France: Southeast France	45 (8)	
Southwest France	52 (9)	
Ireland: Southwest Ireland	80 (14)	
Italy: Central Italy	37 (7)	
Sweden: West Sweden	59 (11)	
UK: North England	75 (14)	
Northern Ireland	72 (13)	
Germany: Northwest Germany	59 (11)	
Characteristics of the children/adolescents		
Sex		
Boy	316 (57)	
Girl	235 (43)	
Age (years)		
Mean [SD]	10.4 [1.5]	15.1 [1.5]
Range	7.7–13.5	12.0–18.6
Impairment		
Gross Motor Function (GMFCS)		
I Walks and climbs stairs, without limitation	164 (30)	188 (34)
II Walks inside, with limitations	125 (23)	99 (18)
III Walks with assistive devices	95 (17)	71 (13)
IV Unable to walk, limited self-mobility	80 (14)	74 (13)
V Unable to walk, severely limited self-mobility	87 (16)	119 (22)
Missing	0	0
Intellectual impairment (IQ)		
>70	272 (49)	257 (47)
50–70	124 (23)	142 (26)
<50	151 (27)	152 (27)
Missing	4 (<1)	0
Bimanual Fine Motor Function (BFMF)		
I Without limitations	188 (34)	190 (35)
II Both hands limited in fine skills	147 (27)	127 (23)
III Child needs help with tasks	92 (17)	103 (19)
IV Child needs help and adapted equipment	68 (12)	69 (12)
V Child needs total human assistance	56 (10)	60 (11)
Missing	0	2 (<1)
Seizures in the previous year		
No seizures (with or without medication)	453 (82)	447 (81)
Seizures	98 (18)	99 (18)
Missing	0	5 (1)
Communication		
Formal communication (with or without difficulties)	479 (87)	481 (88)
No formal communication	72 (13)	66 (12)
Missing	0	4 (<1)
Feeding		
Feeds by mouth with no problems	402 (73)	417 (76)
Feeds by mouth with problems or by tube	149 (27)	132 (24)
Missing	0	2 (<1)
Pain (parent reported) ^a		
Severity: None	166 (30)	144 (26)
Mild/Moderate	240 (44)	193 (35)
More severe	138 (25)	207 (38)
Missing	7 (1)	7 (1)
Total difficulties score of Strength and Difficulties Questionnaire (parent reported)		
Normal (<14)	330 (60)	329 (60)
Borderline (14–16)	97 (18)	99 (18)
Abnormal (>16)	118 (21)	118 (21)
Missing	6 (1)	5 (2)

Table 2 (Continued)

Socio-demographic characteristics	Longitudinal sample n = 551 (%)	
	Childhood	Adolescence
Family characteristics		
Family status		
Living with a partner (includes being married)	445 (81)	442 (80)
Others	106 (19)	108 (20)
Missing	0	1 (<1)
Other children in the same household		
None	99 (18)	114 (21)
One or more non disabled	386 (70)	369 (67)
One or more disabled	59 (11)	61 (11)
Missing	7 (1)	7 (1)
Educational status of reporting parent		
Low	Not recorded	135 (24)
Middle	"	280 (51)
High	"	134 (25)
Missing	"	2 (<1)
Working status of reporting parent		
Working full-time	158 (29)	157 (29)
Working part-time	181 (33)	200 (36)
Others	208 (38)	189 (34)
Missing	4 (<1)	5 (1)
Impact on family scale		
Normal (0)	Not used in analysis.	261 (47)
Borderline (1)	"	71 (13)
Abnormal (2 and more)	"	216 (39)
Missing	"	3 (<1)
Life stress scale		
Normal (0 or 1 event)	Not used in analysis.	314 (57)
Abnormal (2 or more events)	"	221 (40)
Missing	"	16 (3)
Total stress score of Parenting Stress Index		
Normal (<86)	332 (60)	314 (57)
Borderline (86–90)	40 (7)	36 (6)
Abnormal (>90)	163 (30)	190 (35)
Missing	16 (3)	11 (2)

^a Observational time frame: 4 weeks in childhood, 1 week in adolescence.

2.4. Ethics

We obtained ethical approval as appropriate to each country. We obtained written informed consent from all parents and where possible from the children and adolescents.

3. Results

3.1. Sociodemographic data of the study population

The distributions of the socio-demographic characteristics and impairments for the longitudinal sample (n = 551) are shown in Table 2. The mean ages were 10 years 5 months for SPARCLE1 and 15 years 1 month for SPARCLE2. On average, levels of impairment, psychological problems and parenting stress were similar in childhood and adolescence, but parents reported more severe pain for their children in adolescence than in childhood. Those not included in the analysis (n = 43), due to the change of the reporting proxy, did not differ significantly from those included with respect to impairment and the factors mentioned above.

3.2. KIDSCREEN and QoL on group level

Table 3 shows the psychometric properties of KIDSCREEN-52. The *Financial resources* and *Social acceptance* domains showed ceiling effects, with 28% and 40% of parents reporting the maximum score in childhood and 26% and 40% in adolescence. In the *Financial resources* domain there was 20% missing data at time 1 and the high ceiling effects at both time points; we therefore excluded this domain from further analysis.

Table 3 also presents the mean KIDSCREEN scores by domain. Changes in QoL at group level were small (<4 points or less than ½ a standard deviation). QoL decreased in six domains (−1.3 to −3.8 points; $p < 0.01$), and did not change significantly in three domains ($p > 0.05$).

Table 3

Summary statistics for KIDSCREEN scores of children and adolescents with cerebral palsy, where the same parent reported their child's QoL on each occasion (n = 551).

KIDSCREEN domain		Number (%) of proxy-reporting	Mean (SD) of QoL	Median	[IQR]	Floor (%) ^a	Ceiling (%) ^a	Change in QoL between childhood and adolescence	
								Mean adolescent QoL minus childhood QoL (SE) ^b	p-value, paired t-test
Physical wellbeing	Childhood	517 (94)	42.4 (9.8)	41.1	[34.8–49.5]	1.1	2.4	0.4 (0.5)	0.36
	Adolescence <i>Longitudinal</i>	539 (98) 507 (92)	42.6 (10.1)	41.1	[36.7–49.5]	0.4	2.5		
Psychological wellbeing	Childhood	526 (96)	48.6 (9.8)	48.9	[41.2–55.3]	0.2	3.3	−3.4 (0.5)	<0.001
	Adolescence <i>Longitudinal</i>	538 (98) 517 (94)	45.0 (10.4)	43.5	[36.9–52.1]	0.2	4.6		
Moods and emotions	Childhood	523 (95)	50.4 (9.9)	48.6	[43.9–58.0]	0.2	4.8	−3.6 (0.5)	<0.001
	Adolescence <i>Longitudinal</i>	525 (95) 503 (91)	46.5 (10.6)	46.1	[39.8–51.3]	0.2	6.9		
Self-perception	Childhood	507 (92)	51.0 (10.3)	49.1	[44.3–56.2]	0.2	8.9	−3.8 (0.5)	<0.001
	Adolescence <i>Longitudinal</i>	515 (94) 483 (85)	47.5 (10.3)	46.5	[40.5–52.3]	0.2	12.4		
Autonomy	Childhood	516 (94)	43.6 (9.2)	43.5	[37.6–48.2]	0.2	7.5	0.9 (0.5)	0.07
	Adolescence <i>Longitudinal</i>	530 (96) 502 (91)	44.3 (10.6)	43.5	[37.6–51.0]	0.2	2.9		
Relationships with parents	Childhood	521 (95)	49.6 (10.0)	49.4	[42.3–58.5]	0.2	6.3	−2.5 (0.4)	<0.001
	Adolescence <i>Longitudinal</i>	528 (96) 503 (91)	47.3 (10.5)	46.9	[40.2–55.1]	0.2	8.3		
Social support and peers	Childhood	495 (90)	40.2 (11.9)	40.5	[34.7–48.5]	7.0	1.0	−2.4 (0.6)	<0.001
	Adolescence <i>Longitudinal</i>	516 (94) 468 (85)	37.6 (13.2)	38.6	[30.5–46.4]	2.8	0.8		
School life	Childhood	522 (95)	52.5 (10.0)	52.1	[47.5–59.6]	0.2	4.9	−1.3 (0.5)	0.007
	Adolescence <i>Longitudinal</i>	534 (97) 510 (93)	51.1 (10.1)	49.8	[43.3–57.0]	0.2	5.7		
Financial resources	Childhood	438 (80)	50.3 (12.7)	51.9	[43.3–65.0]	5.1	27.9	1.7 (0.6)	0.003
	Adolescence <i>Longitudinal</i>	512 (93) 420 (76)	52.2 (11.3)	51.9	[45.4–65.0]	9.6	25.6		
Social acceptance	Childhood	527 (96)	48.4 (10.9)	50.6	[39.3–58.8]	0.6	40.4	−0.4 (0.5)	0.43
	Adolescence <i>Longitudinal</i>	539 (98) 518 (94)	48.0 (11.6)	50.6	[39.3–58.8]	0.4	39.8		

All numbers rounded to one decimal place.

^a Floor and ceiling effects indicate the % of scores which had the minimum and maximum values respectively.

^b SE Standard Error.

3.3. Regression models

The results of the baseline, child and parent/family models are presented in the Appendix. The proportion of explained variance (R^2) varied from 16% in *Social support and peers* domain in the baseline models to 47% in *Relationship with parents* domain in the child models. In general, the proportions of explained variance were highest in the child models, except for the domains *Psychological wellbeing* and *Autonomy* in the parent/family models.

The final combined regression models are summarised below and in Table 4. For clarity in Table 4 we present the values of the b-coefficients to one decimal place, but calculations are based on exact values. Relationships are statistically significant if the 95% CI does not cross zero and are shown in bold type. However, as discussed in Methods, we mitigate the risk from multiple testing by also highlighting associations where the 99% CI does not cross zero in bold and red type.

In the final combined model, childhood QoL was a significant predictor of adolescent QoL in all domains. The association was strongest for *Relationship with parents* where an increase of one standard deviation (10 points) in the QoL score in childhood was associated with an increase in adolescent QoL of 5.0 points (95% CI 4.2–5.8). The association was weakest for *Moods and emotions* with an increase in adolescent QoL of 3.1 points (95% CI 2.2–3.9).

In the final combined model, associations of adolescent QoL with age, gender or impairments differed between domains and seldom showed large, significant b-coefficients. Being female had a small negative effect on *Moods and emotions* and *Self-perception*. Whilst severe motor impairment had a negative effect on *Autonomy*, other associations with more severe impairment favoured higher QoL: motor impairment with *School life and social acceptance*; no formal communication with *Relationship with parents*; intellectual impairment with *Psychological wellbeing*, *Self-perception*, *Relationship with parents* and *School life* (for b-coefficients and confidence intervals see Table 4).

In the final combined model, adolescent pain was a significant predictor of QoL in seven domains. The association of severe pain was largest with *Physical wellbeing* where QoL was 6.6 points lower than in those without pain.

In the final combined model, psychological problems in childhood or their worsening by adolescence predicted lower QoL in five domains. For example, an increase in SDQ in childhood of 7 points (1 SD, more psychological problems) was associated with a decrease in QoL of 2.8 points in adolescent QoL in the *Social acceptance* domain (i.e. scaled from an increase of one point in SDQ in childhood being associated with a decrease of 0.4 points in QoL). An increase in SDQ between childhood and adolescence of 7 points was associated with a decrease of 4.2 points in adolescent QoL in the *Social acceptance* domain.

In the final combined model, parenting stress in childhood or its worsening by adolescence predicted lower adolescent QoL in eight domains. For example an increase of 15 points in PSI score (1 SD, more stress) between childhood and adolescence was associated with a decrease in adolescent QoL of –4.5 points in the domain *Psychological wellbeing* (i.e. scaled from an increase of one point in PSI between childhood and adolescence being associated with a decrease of 0.3 points in QoL).

In the final combined model, associations of adolescent QoL with socio-demographic and family characteristics seldom showed large, significant b-coefficients, and none in more than one domain.

The proportion of variance in adolescent QoL explained by these selected variables was 30%–40% in most domains, with lowest R^2 in *Autonomy* (29% (28%–29%)) and highest in *Moods and emotions* (44% (42%–45%)) and *Relationship with parents* (50% (49%–51%)).

Sensitivity analyses for those with a different proxy at childhood and adolescence and for the whole sample of 818 participants gave b-coefficients and R^2 that were generally smaller than those from the primary analysis (data not shown).

4. Discussion

This is the first longitudinal study of the QoL of children, randomly sampled from population registers of children with CP, as they move from childhood to adolescence. It allows examination of the predictors of adolescent QoL. In order to include children with all severity levels, in particular those with severe intellectual impairment, we examined parent-reported QoL.

Between childhood and adolescence, average QoL at group level changed little (between 1.3 and –3.8 points depending on domain). These are less than $\frac{1}{3}$ SD and smaller than differences between the corresponding age groups in cross-sectional and longitudinal data from the general population (Palacio-Vieira, Villalonga-Olives, & Valderas, 2008; Ravens-Sieberer et al., 2005).

Childhood QoL was a consistent predictor of adolescent QoL. Whilst severe motor impairment had a negative effect on *Autonomy*, other associations with more severe impairment favoured higher QoL: motor impairment with *School life and social acceptance*; no formal communication with *Relationship with parents*; intellectual impairment with *Psychological wellbeing*, *Self-perception*, *Relationship with parents* and *School life*. An explanation of our result could be that parents considered their children to be free of the emotional ups and downs of adolescents without intellectual impairment.

Of particular importance are associations which refer to the modifiable factors: pain, psychological problems and parenting stress. All three predicted adolescent QoL.

An adolescent's contemporaneous pain was a negative predictor of adolescent QoL in seven domains. For example, a reduction in pain from more severe pain to no pain was associated with an average increase in adolescent QoL of 6.6 points in *Physical wellbeing*. This is around half a standard deviation and therefore of clinical importance. These findings are consistent with other studies which identify the importance of pain in children and adolescents with CP (Badia et al., 2014; Penner, Xie, Binepal, Switzer, & Fehlings, 2013). Psychological problems and parenting stress in childhood or their worsening between childhood and adolescence predicted lower QoL in five and eight domains respectively. A Dutch longitudinal study

Table 4

Predictors of parent-reported QoL in adolescents with cerebral palsy: regression coefficients of the final combined models (longitudinal sample, n = 551).

KIDSCREEN domain in adolescence:	Physical wellbeing	Psychological wellbeing	Moods and emotions	Self-perception	Autonomy	Relationship with parents	Social support and peers	School life	Social acceptance
b (95% CI) **									
Corresponding KIDSCREEN domain in childhood	0.3 (0.2 to 0.4)	0.3 (0.3 to 0.4)	0.3 (0.2 to 0.4)	0.3 (0.3 to 0.4)	0.3 (0.2 to 0.4)	0.5 (0.4 to 0.6)	0.4 (0.2 to 0.5)	0.4 (0.3 to 0.4)	0.4 (0.3 to 0.5)
Age (years)	0.0 (-0.5 to 0.5)	-0.0 (-0.5 to 0.4)	0.3 (-0.2 to 0.8)	-0.0 (-0.5 to 0.5)	0.2 (-0.3 to 0.7)	0.3 (-0.2 to 0.8)	-0.5 (-1.2 to 0.2)	0.0 (-0.5 to 0.5)	0.6 (0.1 to 1.1)
Sex									
Male	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0
Female	-1.1 (-2.6 to 0.3)	-1.1 (-2.5 to 0.3)	-2.3 (-3.8 to -0.9)	-2.1 (-3.5 to -0.6)	0.1 (-1.5 to 1.8)	-0.0 (-1.3 to 1.3)	-0.8 (-2.8 to 1.2)	0.3 (-1.2 to 1.8)	-1.1 (-2.7 to 0.5)
Gross Motor Function (GMFCS)									
I Walks and climbs stairs, without limitation	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0
II Walks inside, with limitations	-1.0 (-3.0 to 1.1)	1.6 (-0.4 to 3.7)	0.8 (-1.3 to 2.8)	0.0 (-2.0 to 2.1)	0.2 (-2.1 to 2.4)	0.7 (-1.2 to 2.6)	-0.1 (-3.0 to 2.7)	2.8 (0.7 to 4.9)	0.2 (-2.1 to 2.4)
III Walks with assistive devices	-1.5 (-3.8 to 0.8)	2.0 (-0.3 to 4.3)	2.0 (-0.4 to 4.3)	0.8 (-1.5 to 3.0)	-1.2 (-3.7 to 1.2)	2.5 (0.3 to 4.7)	3.1 (-0.2 to 6.5)	1.4 (-0.8 to 3.7)	2.0 (-0.5 to 4.5)
IV Unable to walk, limited self-mobility	0.1 (-2.4 to 2.7)	2.6 (0.1 to 5.0)	1.7 (-1.1 to 4.5)	0.8 (-1.8 to 3.5)	-1.6 (-4.2 to 1.0)	2.8 (0.1 to 5.4)	2.2 (-1.9 to 6.2)	3.9 (1.4 to 6.4)	4.3 (1.5 to 7.1)
V Unable to walk, severely limited self-mobility	-1.5 (-4.3 to 1.2)	1.0 (-1.7 to 3.7)	1.6 (-2.6 to 5.9)	1.0 (-2.1 to 4.1)	-4.6 (-7.4 to -1.7)	1.1 (-2.8 to 4.9)	1.1 (-4.7 to 7.0)	2.9 (0.1 to 5.6)	8.3 (5.1 to 11.5)
Binominal Fine Motor Function (BFMF)									
I Without limitations			0.0			0.0	0.0		
II Both hands limited in fine skills			-2.0 (-3.9 to -0.1)			-1.0 (-2.8 to 0.8)	-2.3 (-5.0 to 0.5)		
III Child needs help with tasks			-0.6 (-2.9 to 1.8)			-2.2 (-4.4 to 0.1)	-0.1 (-3.4 to 3.2)		
IV Child needs help and adapted equipment			1.8 (-1.8 to 5.4)			1.4 (-1.9 to 4.7)	-6.3 (-11.1 to -1.4)		
V Child needs total human assistance			-0.1 (-4.4 to 4.3)			-2.2 (-6.3 to 1.9)	-0.1 (-6.2 to 6.0)		
IQ									
>70	0.0	0.0	0.0	0.0		0.0		0.0	
50-70	0.1 (-1.9 to 2.0)	2.2 (0.3 to 4.1)	1.8 (-0.2 to 3.7)	2.0 (0.0 to 3.9)		1.6 (-0.2 to 3.4)		2.2 (0.3 to 4.2)	
<50	1.5 (-0.6 to 3.5)	4.5 (2.4 to 6.6)	2.8 (0.7 to 5.0)	6.1 (3.8 to 8.4)		2.9 (0.8 to 5.0)		2.6 (0.5 to 4.8)	
Feeding									
Feeds by mouth with no problems				0.0					0.0
Feeds by mouth with problems or by tube				2.0 (-0.2 to 4.2)					-2.2 (-4.6 to 0.1)
Communication									
Formal communication (with or without difficulties)						0.0			
No formal communication						3.7 (1.1 to 6.4)			
Severity of pain in previous week:									
In childhood									
None					0.0				
Mild / moderate					1.1 (-0.8 to 3.0)				
More severe					-0.9 (-3.2 to 1.4)				
In adolescence									
None	0.0	0.0	0.0	0.0	0.0			0.0	0.0
Mild / Moderate	-3.9 (-5.8 to -2.1)	-2.2 (-4.1 to -0.3)	-2.9 (-4.8 to -1.0)	-2.2 (-4.1 to -0.3)	-2.4 (-4.5 to -0.3)			-3.4 (-5.4 to -1.5)	-4.1 (-6.2 to -2.0)
More severe	-6.6 (-8.6 to -4.7)	-3.0 (-4.9 to -1.1)	-4.0 (-6.0 to -2.0)	-1.7 (-3.7 to 0.3)	-1.2 (-3.4 to 1.1)			-3.6 (-5.6 to -1.5)	-4.1 (-6.2 to -1.9)
Total difficulties score of SDQ in childhood		-0.3 (-0.5 to -0.1)	-0.3 (-0.5 to -0.1)				-0.3 (-0.6 to -0.0)	-0.3 (-0.5 to -0.2)	-0.4 (-0.5 to -0.2)
Change in score (SDQ) [†]		-0.3 (-0.5 to -0.1)	-0.4 (-0.6 to -0.2)				-0.4 (-0.7 to -0.1)	-0.5 (-0.7 to -0.3)	-0.6 (-0.7 to -0.4)
Family status									
Living with a partner (included being married)					0.0				
Others					-2.2 (-4.2 to -0.2)				
Other children in the same household									
None		0.0							0.0
One or more non disabled		-2.1 (-4.0 to -0.2)							0.4 (-1.8 to 2.5)
One or more disabled		-0.2 (-3.0 to 2.6)							-0.3 (-6.0 to 0.2)
Educational status of reporting parent									
Low	0.0				0.0				
Middle	-0.6 (-2.4 to 1.2)				-2.5 (-4.5 to -0.5)		-0.8 (-2.4 to 0.9)		
High	1.6 (-0.5 to 3.8)				-3.8 (-6.3 to -1.4)		-1.8 (-3.8 to 0.3)		
Working status of reporting parent									
Working full-time				0.0					0.0
Working part-time				0.3 (-1.6 to 2.1)					0.3 (-1.8 to 2.4)
Others				2.5 (0.6 to 4.4)					2.0 (-0.3 to 4.2)
Impact on family scale (Impact of child's psychological problems on the family)									
Normal (0)			0.0				0.0		
Borderline (1)			-1.2 (-3.4 to 1.0)				2.4 (-0.8 to 5.7)		
Abnormal (2 and more)			-2.5 (-4.4 to -0.6)				-2.0 (-4.7 to 0.6)		
Total stress score of PSI in childhood	-0.1 (-0.1 to -0.0)	-0.1 (-0.2 to -0.1)	-0.0 (-0.1 to 0.0)	-0.1 (-0.1 to -0.1)	-0.1 (-0.1 to -0.0)	-0.1 (-0.1 to -0.1)	-0.1 (-0.2 to -0.0)		
Change in score (PSI) [‡]		-0.2 (-0.2 to -0.1)	-0.1 (-0.2 to -0.1)	-0.1 (-0.2 to -0.1)	-0.1 (-0.1 to -0.0)	-0.2 (-0.2 to -0.1)	-0.1 (-0.2 to -0.0)	-0.1 (-0.1 to -0.0)	
R ² as %									
	34 (33 to 35)	38 (36 to 39)	44 (42 to 45)	36 (34 to 38)	29 (28 to 29)	50 (49 to 51)	31 (28 to 32)	31 (29 to 33)	37 (36 to 38)

All numbers rounded to one decimal place. All regression models adjusted for region, age and gender and Gross Motor Function. Confidence intervals obtained by bootstrapping. The interactions age x sex and the variables seizures and Life stress scale were never chosen. Statistically significant relationships (i.e. with 95%CI excluding zero) are presented in **bold**; those with 99%CI excluding zero are presented in **bold and red**. *b-coefficients for continuous covariates (QoL in childhood, SDQ and PSI scores, changes in SDQ and PSI scores) indicate the change in QoL in adolescence associated with a change of one point in the covariate. †b-coefficients for categorical covariates (impairments, pain, family status) indicate the estimated average difference in QoL between the relevant category and the reference category; b-coefficients below 0 indicate a lower QoL in the corresponding category. ‡Changes were calculated as adolescent scores minus childhood scores.

of children aged 9–16 years with CP also found that mental health problems predicted lower social functioning and mood over a follow-up of three years (Janssen et al., 2010). Although, the effects of psychological problems and parenting stress in our study may not be substantial (with change in QoL of 2–4 points for a change of 1 SD in SDQ score, and of 1.5–3 points for 1 SD in PSI score) the effects might be more important than they first seem. The predictions are across five or more domains and therefore influence many aspects of adolescent life. Further, childhood QoL itself powerfully predicts adolescent QoL in

all domains and models. Therefore, the management of psychological problems and parenting stress in childhood will have a further effect on adolescent QoL via their effect on child QoL.

Comparing this analysis of all adolescents with CP to our earlier report on the 355 adolescents who self-reported their QoL (Colver, Rapp, & Eiseemann, 2015), parent-reported QoL is more influenced by the adolescent's pain and psychological problem and less so by impairments. There is also a stronger negative effect of parenting stress. A comparison of child- and parent-reported QoL³⁰ found that parents with higher parenting stress tended to report a lower QoL for their children than the children themselves reported. Bias may therefore be present but it is not possible to quantify such potential bias directly.

Regarding strengths of our study, the sample was large and had been randomly sampled from population based registers. We took account of the original sampling strategy by controlling for motor impairment. We imputed missing data to retain all children in the analysis. Characteristics of those lost to follow-up were evaluated. We used the well validated KIDSCREEN-52 questionnaire which has psychometric properties that are similar in children with CP and the general population (Erhart et al., 2009).

Regarding weaknesses of our study, child psychological problems and parenting stress may influence each other, and we did not capture depression or anxiety of the reporting parent. The time frame for measuring pain was the past four weeks in childhood and the past one week in adolescence. We did not capture chronic pain, and its influence should be evaluated in further studies. The Parenting Stress index Short Form is only validated for parents of children up to age 12 years (Abidin, 1995).

The main implication for research is that trials should now be undertaken. We need rigorous trials to see if reducing pain, psychological problems and parenting stress improve child QoL. This may be easier to say than do because we first need convincing evidence that interventions can reduce pain, parenting stress and psychological problems. Once this is forthcoming, those interventions should be administered with a view to assessing their impact on QoL.

5. Conclusion

Severity of impairment and socio-demographic factors are difficult to influence but fortunately had little impact on adolescent QoL. However, psychological problems and parenting stress in childhood and their worsening from childhood to adolescence predicted lower QoL. Also contemporaneous pain predicted lower adolescent QoL. These are modifiable and addressing them may improve adolescent QoL.

Conflict of interest

The authors declare that they have no conflicts of interest to disclose.

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Contributors' statements

Marion Rapp: Dr. Rapp took part in the design of the study, the acquisition of data in one region, wrote the manuscript, and approved the final manuscript as submitted.

Nora Eiseemann: Ms. Eiseemann performed the statistical analyses, reviewed and revised the manuscript, and approved the final version as submitted.

Virginie Ehlinger: Ms. Ehlinger participated in performing the statistical analyses, critically reviewed the manuscript, and approved the final manuscript as submitted.

Catherine Arnaud: Dr. Arnaud took part in the conception and design of the study, managed the acquisition of data in one region, drafted the initial manuscript, and approved the final manuscript as submitted.

Jérôme Fauconnier, Marco Marcelli, and Malin Nystrand: Dr. Fauconnier, Dr. Marcelli, and Ms. Nystrand took part in the conception and design of the study, managed the acquisition of data in each region, and approved the final manuscript as submitted.

Allan Colver: Professor Colver directed the overall SPARCLE project, reviewed and revised the paper, and approved the final manuscript as submitted.

Ute Thyen: Professor Thyen gave important intellectual input to all aspects of the study and the paper. She had full access to the data analysed for this paper, and approved the final manuscript as submitted.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.ridd.2016.12.005>.

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