Quality of life of extremely preterm school-age children without major handicap: a cross-sectional observational study

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ABSTRACT

Objective To determine the quality of life (QoL) of school-aged children who were born <28^{+0} weeks of gestation and who have no resultant major disabilities.

Design, setting and patients A cross-sectional multicentre study of extremely preterm (EPT) infants born <28^{+0} weeks, discharged alive and free from severe impairments (cerebral palsy, autism, major cognitive disabilities). Two generic, self-evaluation and hetero-evaluation (by parent) QoL measurement questionnaires (Kidscreen 10/VSP-A) were used and then compared with French population reference.

Main outcome measures Clinical examination, an assessment of cognitive functions and QoL between 7 and 10 years of age.

Results 40 (7.5%) severely disabled children were excluded. Among those 471 eligible, the lost to follow-up group (169 (36%)) paralleled those 302 (64%) included in the study. The mean gestational age was 26.2 (±0.8), birth weight was 879 (±181) g and the mean age was 8.4 (±0.87) years. 48% of participants had minor or moderate cognitive disabilities based on their Full-Scale Index Quotient. Working memory, attention and mental flexibility scored as low-average. Except for family relationships, the EPT QoL VSP-A and Kidscreen 10 assessment were significantly lower based on the children's and parent's perspectives. Children reported the most significant QoL decline as (1) friends' relationships, (2) self-esteem and (3) leisure, while parents indicated (1) psychological well-being, (2) schoolwork and (3) vitality.

Conclusion The QoL of a school-age EPT child without severe impairment was lower relative to a reference population from both the parents' and child's points of view. This evaluation should help to better understand the long-term outcomes and to provide better support for them and their families.

Trial registration number NCT01675726, pre-results.

What is already known?

- Quality of life (QoL) is inadequately evaluated for those school-aged children born <28 weeks' gestational age.
- There are no studies of children free from serious sequelae, which constitute the majority of extremely premature school-aged children.
- Methodologically weak studies show the parents perceived their children's QoL as low. A single, unicentric health questionnaire assessment of child's QoL reported by both the child and the parent shows an identical QoL as compared with the control population.

What this study adds?

- Except for the area of family relationships, there is a decreased school-aged QoL for former extremely preterm children who had no serious sequelae.
- There is a decreased QoL in combined assessments by the child and that reported by the parent.
- A lowered QoL of these children should be considered when making therapeutic choices.

INTRODUCTION

Infants born at the limit of viability are increasingly being admitted to neonatal intensive care units. As survival rates increase, extremely preterm (EPT) birth (<28 weeks of gestational age (GA)) has become an important public health issue. More than half of EPT children have moderate or mild disabilities and 10%–15% have severe disabilities when they reach school age.[1–4]

During the pivotal transition of their elementary school years, the EPT children's overall disabilities disrupt their learning and schooling. This disruption compromises social integration and self-fulfilment[3–6] and highlights the importance of this population's quality of life (QoL) evaluation during this period.[7 8] There are only three studies that have a highly heterogeneous design in method, population and outcomes.[9–11] Two of these used a health questionnaire rather than a QoL generic questionnaire as their measurement instrument,[9 10] and two used only a parental approach.[9 11] Hack *et al*'s study used a combined parent–child QoL evaluation with CHIP (Child Health and Illness Profile).[10] The parental and children's self-questionnaires are multidimensional and use good psychometric properties.[12] However, their weakness is that the sample on which the questionnaires are standardised is neither representative nor large. On the other

hand, the CHIP measured the health status and what would be expected for a good QoL.[12] Finally, Hack *et al*'s study is an American unicentric study, and thus the conclusions are difficult to transpose to a country's scale.[10]

Consequently, for EPT school-aged children, there is no study exploring their QoL by means of a validated, self-administered, generic questionnaire from the child's point of view.[5 8 13]

Moreover, major disabilities are known to impact QoL, but there are no studies focusing on EPT children that have no major disabilities.[14 15]

Our main objective was to evaluate the QoL of EPT children between the ages of 7 and 10 years, who had no major disabilities during follow-up. This evaluation was then compared with a general reference population using combined parent–child standardised, validated, generic questionnaires, which measured how the individual's health status impacted his well-being.

DATA AND METHODS

Experimental design

A cross-sectional, multicentre observational study was carried out between 2012 and 2015 within five French Level III facilities authorised to care for extreme preterm infants less than 28 week's gestational age (GA) (*Marseille Conception*, *Marseille Nord*, *Nantes*, *Nîmes* and *Rouen*).

Population

Inclusion criteria were an EPT child (less than 28 week's GA), born between 1 January 2004 and 31 December 2007, and hospitalised after birth in one of the participating facilities, school-aged (7 to 10 years) at the time of inclusion, capable of answering a French-language questionnaire parents or legal representatives accepting the study's participation principles and signing an informed consent.

Non-inclusion criteria were children having died after discharge, blindness or amblyopia (visual acuity <3), deafness (prescribed hearing aid) and severe cerebral palsy (CP) (according to Bax *et al*[16] determined through the Gross Motor Function Classification System (>2)).[17] We chose to define severe cognitive disability (Full-Scale Intelligence Quotient (FSIQ) score less than 65).[18]

General framework

After the investigating physician verified full compliance with the inclusion criteria, the families were contacted by phone, and a day-long evaluation visit to the birth centre was proposed. If the participating family could not arrange a site visit, a phone interview was arranged.

Measurements collected

Clinical examination

Physician-conducted clinical examinations were done for current diseases, treatments in progress, any specialist educational assessments, height and weight measurements, neurological gross and fine motor skills evaluation by Touwen Infant neurological examination,[19] and any recorded visual and/or auditory disorders.

Psychometric assessment

Psychometric assessment was performed by a neuropsychologist, using the Wechsler Intelligence Scale for Children—Fourth Edition (WISC-IV)[20] to calculate four sub-indices and an overall intelligence index (FSIQ) (mean 100 and SD 15); the Rey figure,[21] a short perceptual organisation and memory test; and the NEuroPSYchological assessment (NEPSY) subtests evaluating attention and executive functions (mean 10 and SD 3).[22] Behaviour evaluation was obtained by the Goodman Strengths and Difficulties Questionnaire with 25 items for the parents.[23] Anxiety evaluation was obtained by the Spielberger questionnaire (State-Trait Anxiety Inventory for Children), containing 20 items addressed to children.[24]

FSIQ categories were defined according to the mean SD of the FSIQ: no disability, FSIQ greater or equal to 89; mild disability, FSIQ less than 89 and greater than or equal 79 SDs; and moderate disability, FSIQ less than 79 and greater than or equal to 65.[18]

Data collection

Perinatal and pregnancy data included maternal age, type of prematurity, antenatal corticosteroid therapy, multiple pregnancies, GA (weeks), birth weight, gender and neonatal morbidities. The sociodemographic and family data were collected at the time of the assessment included age, gender, parental education, parents' employment, the family's material wealth as reported by the child and evaluated by the Family Affluence Scale (FAS)[25] and, finally, the child's school life.

Quality of life assessment

The children's life assessment quality was measured by using two standardised, validated questionnaires in French: VSP-A (*Vécu et Santé Perçue des Adolescents*) and Kidscreen 10, with scores ranging from 0 to 100. The higher score reflects a higher QoL level.[12 26 27]

The VSP-A is a self-administered questionnaire with an index total score along with nine scores measuring a total of nine dimensions (vitality, psychological well-being, relationships with friends, hobbies, relationships with family, physical well-being, relationships with teacher, school work and self-esteem).[26] The questionnaires used were the VSP-A child version (VSP-Ae: 35 items) and the parent version (VSP-Ap: 34 items). Psychological and physical well-being dimensions of the VSP-Ap are gathered together under the general well-being dimension for the VSP-Ae. The full version of the Kidscreen questionnaire explores the following dimensions: physical well-being, psychological well-being (positive and negative), emotions, relations with parents and autonomy, relations with friends, and social and school support.[12 27] Our study used the 10-item child and parent versions (short Kidscreen-10) to obtain an index total score.[27]

Reference group

The reference population was derived from a 2003 established European database, which included a French sample (n=989) obtained by randomly dialling telephone numbers (CATI method: Computer Assisted Telephone Interview—RDD: Random Digital Dialling), and compared with Eurostat data.[28] This sample included children aged 8 to 10 who responded to the VSP-A and Kidscreen QoL questionnaires. This national data sampling of approximately 1000 children in the general population provided a baseline sample comparable to the preterm group studied for the confounding factors of age and gender.[29 30]

Statistical analysis

A descriptive analysis was conducted with qualitative variables presented as numbers and percentages. Quantitative variables were presented as averages and SD. All variables had an approximate normal distribution, which was assessed graphically for each quantitative variable using histograms and QQ plots. Data

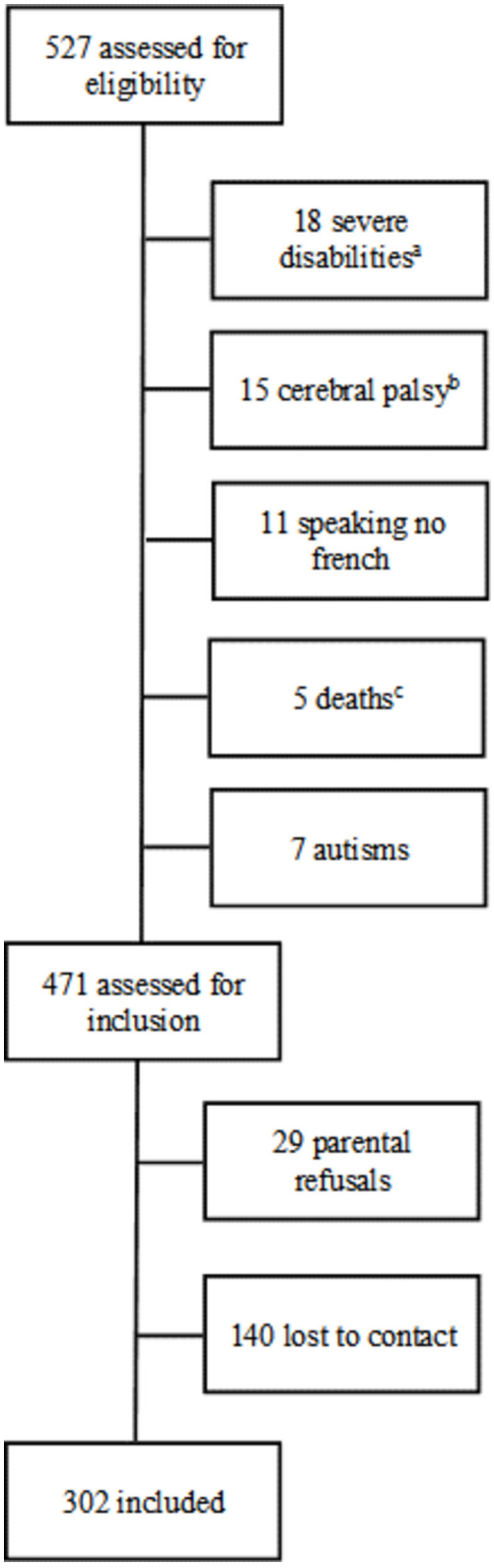

Figure 1 [a]FSIQ <65 (FSIQ, Full-Scale Intelligence Quotient measured by the Wechsler Intelligence Scale for Children—Fourth Edition). [b]Gross Motor Function Classification System >2 (gross motor function). [c]Deaths between hospital discharge and evaluation at school age.

on GAs, birth weights and parental occupations were compared with those that were lost to contact or for whom there was no consent. We used χ^2 test or Fisher's test for the qualitative variables, and Student's t-test and Mann-Whitney U test for continuous variables.

For QoL, the different dimensions of the questionnaires' scores and the score were established using the scoring algorithms made available by the designers of the questionnaires. The distribution normality of the QoL scores were systematically verified using the Shapiro-Wilks and Kolmogorov-Smirnov tests. If one or more scores were not considered normally distributed, logarithmic transformation techniques or normalisation algorithms (Blom or Tukey algorithms) were implemented. The results were presented using different descriptive statistical tools: position and dispersion indicators (mean, median, mode, SD, extent), and different intervals were determined (fluctuation intervals, CIs, interquartile intervals). This analysis was identically conducted on the reference population.

The statistical analyses were carried out using the software R. The significance threshold was set at 5%. Missing data were too low (<3%) to be put into the included population.

RESULTS

Population

There were 40 (7.5 %) excluded for a major disability. A total of 302 (64.1%) were included in the study with only 29 (6.2%) refusals (figure 1).

Perinatal characteristics

The mean GA at birth was 26.25 weeks (±0.87) with a mean birth weight of 879.03 g (±181.73). These data are not significantly different from the non-participating eligible population. The neonatal morbidity was rated high (table 1).

School-aged children's characteristics

The children's mean age was 8.45 years (±0.75). There were 55.9% of parents with an educational level (father or mother) beyond a high school diploma. Material wealth was rated high by 57.4% of families (score FAS) (table 2).

With regards to schooling, 64 (21.4%) repeated grades, 107 (41.3%) received academic support and 228 (79.2 %) were followed by a healthcare professional. There were 48.0% of participants who had a minor or moderate cognitive disability based on their FSIQ index. The working memory, attention and mental flexibility were in the range of low average.

Quality of life data as compared with the same gender and age in the reference population

Participants had significantly lower QoL scores than the reference population for most dimensions of the VSP-A. The most significant QoL evaluation lowering from the children's point of view was (1) relationships with friends, (2) self-esteem and (3) leisure. For the parents, it was (1) psychological well-being, (2) schoolwork and (3) vitality (table 3).

The EPT children's QoL assessment by Kidscreen was significantly lower from the children's and parent's report.

DISCUSSION

As compared with the reference population, we showed that school-aged EPT children having no major disability had a significantly lower QoL.

The characteristics of our study population are valid since they are comparable to a Swedish cohort EXPRESS[18] of EPT children aged 6 ½ years born during the same time period. Additionally, our participating centres annually care for 20% of EPTs born in France each year.

Table 1 Baseline perinatal characteristics of study and non-included population

	Study population (n=302)	Non-included population* (n=169)	P values
Antenatal data			
Mother's age in years (mean±SD)	29.33 (±5.51)		
<20	6 (2.03)		
20–35	250 (84.75)		
>35	39 (13.22)		
Multiple pregnancies	103 (34.22)		
Antenatal steroids	263 (88.55)		
Cause of prematurity			
Spontaneous	209 (69.44)		
Schedule	92 (30.56)		
Perinatal data			
Male gender	143 (51.53)	92 (55.76)	0.38
GA at birth in WA (mean±SD)	26.25 (±0.87)	26.23 (±0.89)	0.62
23	1 (0.33)	0 (0)	
24	10 (3.31)	9 (5.77)	
25	50 (16.56)	21 (13.46)	
26	94 (31.13)	51 (32.69)	
27	147 (48.68)	75 (48.08)	
Mean BW in grams (±SD)	879.03 (±181.73)	866.40 (±170.91)	0.46
SGA†	167 (59.86)		
Neonatal morbidities			
CLD	161 (54.21)		
NEC (all stages)	72 (23.84)		
Late-onset sepsis‡	162 (54.18)		
PDA treated§	165 (54.64)		
IVH ≥grade 3¶	10 (3.32)		
Retinopathy of prematurity ≥stage 3	1 (0.34)		

Data expressed as n (%) or mean (±SD); P value: value for difference between groups with available data was obtained with χ^2 test. P>0.05.
*140 lost to contact and 29 refusal.
†Birth weight less than −2 SD of the French intrauterine growth standard.
‡Sepsis after 72 hours of life.
§Haemodynamically significant PDA, ibuprofen treatment and/or surgical ligation.
¶Grade 3: subependymal haemorrhage (>50%) with extension into lateral ventricles with ventricular enlargement, grade 4: intraparenchymal haemorrhage, periventricular leucomalacia.
BW, birth weight; CLD, chronic lung disease; GA, gestational age; IVH, intraventricular haemorrhage; NEC, necrotising enterocolitis; PDA, patent ductus arteriosus; SGA, small for gestational age; WA, weeks of amenorrhoea.

Table 2 School-aged children's characteristics (n=302)

	Number (n)	% or mean (± SD)
Sociodemographic data		
Mean age in years	302/302	8.47 (±0.75)
Education (highest scholastic level of mother or father)	295/302	
Primary, secondary and technical training	54/295	18.31%
High school	76/295	25.76%
University	165/295	55.93%
Professional activity of the parents	296/302	
Neither: both parents	22/296	7.43%
One of two parents	100/296	33.78%
Both parents	174/296	58.78%
Family Affluence Score	293/302	
Low	7/293	2.39%
Average	118/293	40.27%
High	168/293	57.34%
Weight child data	282/302	
Normal	210/282	74.47%
Overweight	28/282	9.93%
Obesity	7/282	2.48%
Insufficient weight	37/282	13.12%
School data		
Repetition of a class	64/299	21.40%
Tutoring	107/259	41.31%
Paramedical follow-up (at least one)	228/288	79.17%
Neurocognitive evaluation*		
WISC-IV	250/302	
Mean FSIQ (±SD)		91.54 (±15.35)
Mean Verbal Comprehension Index (±SD)		98.24 (±16.19)
Mean Perceptual Reasoning Index (±SD)		91.30 (±15.31)
Mean Working Memory index (±SD)		90.95 (±14.40)
Mean Processing Speed Index (±SD)		92.30 (±14.48)
FSIQ disability categories†		
None (FSIQ >89)	130/250	52.00%
Mild (FSIQ 77–89)	71/250	28.40%
Moderate (FSIQ 65–77)	49/250	19.60%
NEPSY	250/302	
Mean score executive function/planning (Tower) (±SD)		10.61 (±2.72)
Mean score design fluency (±SD)		8.22 ± (2.96)
Mean score auditory attention (±SD)		8.93 (±1.68)
Mean score visual attention (±SD)		10.31 (±3.35)

*Cognition was tested in 250 EPT (extremely preterm) children, quality of life was measured in 302 EPT children.
†FSIQ categories were defined according to the mean SD of the WISC-IV: no disability as FSIQ greater or equal to −1 SD, mild disabilities as FSIQ less than −1 SD and greater than or equal to −2 SDs, moderate disability as FSIQ less than −2 SDs and greater than or equal to −3 SDs.
FSIQ, Full-Scale Intelligence Quotient; WISC-IV, Wechsler Intelligence Scale for Children—Fourth Edition.

QoL assessment varies with the measured concept, the age, the instruments used and whether or not a self-evaluation or hetero-evaluation is used.[26 29] In our work, we therefore used generic measuring instruments: Kidscreen and VSP-A, designed to measure the QoL in both healthy and chronically ill children.[29] The strength of these measuring instruments is (1) their simultaneous development in several European countries in consideration of age-appropriate and interculturally relevant content, and (2) their relevance for school-age children in allowing measurement of the most meaningful WHO-recommended dimensions for a child-suitable measurement of QOL. Jardine *et al* showed that a combined QoL appraisal, even in school-aged children, is necessary in order to recognise the divergent points of view between the parent and child.[30] These divergent QoL perspectives are usually observed in various pathologies seen in the general population.[12 27] Typically, parents report a lower QoL than do their children, especially their adolescent children.[29] Our work does not confirm this at school age when the QoL levels are lower and concordant between the parents and their infant. Generally, even if the parents' perceptions agree with their children's regarding physical health, they diverge in the more subjective areas such as psychological and psychosocial domains.[8] In children 8 to 12 years old, it is the psychosocial domain (self-esteem,

Table 3 Quality of life (QoL) data

	Study population (n = 302)		Reference data*	Difference				Effect size (ranking)†
	Mean	SD	Expected mean	Mean	SD	95% CI	P values	
VSP-Ae (children assessment)								
Vitality	76.45	19.91	82.46	−6.01	20.00	−8.31 to −3.72	<0.0001	5
General well-being	73.06	17.68	78.43	−5.37	17.77	−7.41 to −3.33	<0.0001	4
Relationship	45.12	27.04	58.87	−13.74	27.19	−16.88 to −10.61	<0.0001	1
Leisures	61.83	20.13	69.64	−7.80	20.10	−10.12 to −5.49	<0.0001	3
Relationship with family	72.33	19.70	73.23	−0.90	19.81	−3.17 to 1.38	0.4377	
School work	76.20	23.12	82.05	−5.85	23.11	−8.52 to −3.19	<0.0001	6
Self esteem	75.24	20.66	84.61	−9.38	20.66	−11.76 to −7.00	<0.0001	2
Index	68.68	13.46	75.58	−6.90	13.62	−8.49 to −5.32	<0.0001	
VSP-Ap (parent assessment)								
Vitality	70.46	15.56	77.36	−6.90	15.71	−8.74 to −5.06	<0.0001	3
Psychological well-being	70.27	20.70	81.34	−11.07	20.73	−13.48 to −8.65	<0.0001	1
Relationship	58.65	20.34	64.52	−5.87	20.41	−8.34 to −3.40	<0.0001	6
Leisures	50.87	19.48	56.99	−6.12	19.42	−8.38 to −3.86	<0.0001	5
Relationship with family	76.87	14.00	78.60	−1.73	14.06	−3.35 to −0.10	0.0376	
Physical well-being	76.01	16.81	78.58	−2.57	16.88	−4.54 to −0.61	0.0105	
Relationship with teachers	73.73	18.72	75.06	−1.33	19.06	−3.62 to 0.96	0.2536	
School work	69.90	19.50	79.72	−9.82	19.38	−12.09 to −7.54	<0.0001	2
Self-esteem	79.06	26.59	88.33	−9.26	26.97	−12.42 to −6.11	<0.0001	4
Index	70.76	11.19	75.92	−5.90	11.20	−7.35 to −4.45	<0.0001	
Kidscreen—children								
Index	70.76	17.13	76.88	−6.12	17.24	−8.09 to −4.14	<0.0001	
Kidscreen—parents								
Index	69.66	14.32	71.84	−2.18	14.32	−3.83 to −0.52	0.0101	

VSP-Ae and VSP-Ap: Life and Perceived Health of Child and Adolescent, QoL questionnaires (children and parent assessments, respectively) whose scores vary between 0 and 100, higher scores indicating better QoL; Kidscreen—children and Kidscreen—parents: QoL questionnaires (children and parent assessments) whose scores vary between 0 and 100, higher scores indicating better QoL.

*Reference data. This sample included children aged 8 to 10 who responded to the VSP-Ae and Kidscreen QoL questionnaires, as well as their parents who responded to the VSP-Ap and Kidscreen QoL questionnaires. This national data sampling of approximately 1000 children in the general population provided a baseline sample comparable with the preterm group studied for the confounding factors of age and gender.

†Effect size: rank of decrease in QoL in each domain for the VPA-Ae and VSP-Ap QoL questionnaires (standardised effect size obtained by dividing the mean difference by the SD), 1, 2 and 3 numbers indicating the top three of the most decreased QoL domains.

relationship with friends, then school).[30] These data from the literature are consistent.

This project's design is based on a retrospective population with EPT subjects being clearly identified in places of exclusive enrolment. Final results of the study might be limited if 'non-participation' is related to their socioeconomic or clinical characteristics, as these characteristics impact on QoL. To ensure that our population is representative of the target population and to discuss the extrapolation power of the highlighted results, we compared perinatal characteristic of our inclusive population (lost to follow-up, non-consenting) to the included populations without noting a difference. The non-participant's socioeconomic status is unknown but probably low. However, a lower socioeconomic level has more severe sequelae such as severe disability and/or cerebral palsy[15] and is also a QoL lowering determinant.[8 14] As a result, the impact of non-participation of our QoL results was minimised but might affect the analysis.

The psychometric properties of our two QoL questionnaires were validated on a large sample of children. These populations provided a reference database due to their numbers and multicultural diversity. This multicountry diversity is more representative since a voluntary control group in each centre is biased by both the city and centre effects.

We could have strengthened our study by choosing to investigate the quality of life in a population of full-term infants of the same age in each centre, but the study's feasibility process would have been burdened with minimal benefits from this option.

Usually, cohort EPT studies in the USA, Canada or Europe document long-term health and cognitive status.[4 18 31 32]

A study describing the perspective of French experts on QoL assessment of EPT children showed that QoL appeared as a subjective notion which was difficult to implement but could influence treatment choices.[33]

A preterm QoL meta-analysis in 2008[7] and 2016[8] selected 37 studies, 5 of which were school-aged children who were born very preterm (<32 GA). Only three EPT children's studies have been undertaken (395 total children). The three studies report a low QoL when assessed by parents.[9–11] Parents may underestimate the QoL of their EPT child by correlating it to their own memories of the child's traumatic birth and neonatal hospitalisation.[8] Only one study had comparable methodology with combined QoL evaluation. That study's 220 EPT children showed an identical QoL compared with the control group's children born at term.[10] However, these results contradict our findings although explanations can be found for these differences. As an example, CHIP measured health status and the health

status of school-age children born EPT are generally good.[8] Furthermore, there is an assumption that, depending on the country, cultural differences impact QoL.[34] The lessening QoL in our population is comparable to a recent adult population study where only autonomous adults (without impairment) were respondent.[35]

These QoL data could shed light on making public decisions through a better appreciation of EPT's social and economic impact. Moreover, the idea of 'burden' appears in the literature,[36] in reference to the heavy demands placed on medical and rehabilitation follow-up. This raises questions regarding the intensive medical and economic measures implemented in the perinatal period as compared with those reduced after the infant's follow-up discharge. That is, there exists a limited number of places in medical and social facilities, as well as a recent and limited development of follow-up networks. There is also difficulty in providing schooling, and finally, there exists non-reimbursement for some rehabilitation treatments.

These QoL data may be used as a patient management tool to improve both care and prematurity outcome information as described by both Stahlmann *et al*[11] and Greenhalgh *et al*[37] from a more general standpoint. Parents need more concrete information than the conventional data on morbidity or mortality. While information on the QoL of ETP children appears to be useful to parents, it is also useful to physicians.[38]

These QoL data can clarify the meaning of the initial therapeutic act with regards to these children's future social integration, interpersonal capacity and physical and moral suffering.[34 36] Given the specific nature of the problems raised by ETP, in particular the dilemma of the limits of viability,[36] the experts would benefit from a tool to rationalise individual decision-making, notably to reflect on the 'burden–benefit' balance of neonatal surveys.

Finally, there is a need to go further and explore the QoL determinants of EPT children that are now of school age. These data are available from our cohort and are currently being analysed.

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Contributors CG and PA conceptualised and designed the study, drafted the initial manuscript and approved the final manuscript as submitted. VB-M and BT organised and conducted the project, and undertook data management and data analysis. NR and JB carried out the initial analyses, reviewed and revised the manuscript, and approved the final manuscript as submitted. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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