Quality of Life of Children Born with a Congenital Heart Defect

Neil Derridj, MD, MPH[1,2], Damien Bonnet, MD, PhD[2], Johanna Calderon, PhD[3,4], Pascal Amedro, MD, PhD[5], Nathalie Bertille, PhD[1], Nathalie Lelong, MSC[1], Francois Goffinet, MD, PhD[1], Babak Khoshnood, MD, PhD[1], and Romain Guedj, MD, PhD[1,6]

Objectives To identify subgroups with a congenital heart defect (CHD) at risk of health-related quality of life (QoL) impairment at 8 years of age according to their medical and surgical management.

Study design From a prospective population-based cohort study, 598 patients with CHD were subdivided according to their medical and surgical management: (1) CHD followed-up in an outpatient clinic, (2) complete repair before age 3 years, (3) complete repair after age 3 years, (4) palliative repair, or (5) CHD with spontaneous resolution (reference subgroup). Self-reported QoL and parent-reported QoL were measured using the Pediatric Quality of Life Inventory version 4.0 (score range, 0-100) at age 8 years. Multivariable regression analysis and Cohen effect size were used to compare outcomes across the CHD groups.

Results Self-reported and parent-reported QoL scores for the palliative repair subgroup were lower (β = −2.1 [95% CI, −3.9 to −0.2] and β = −16.0 [95% CI, −22.4 to −9.5], respectively), with a large effect size (δ = −0.9 [95% CI, −1.4 to −0.4] and δ = −1.3 [95% CI, −1.8 to −0.7], respectively). Parent-reported QoL scores for the complete repair after age 3 years subgroup were lower (β = −9.2; 95% CI, −15.0 to −3.5), with a large effect size (δ = −0.9; 95% CI, −1.4 to −0.5). Self-reported QoL scores for the complete repair before age 3 years subgroup was lower (β = −1.3; 95% CI, −1.9 to −0.6), with a small effect size (δ = −0.4; 95% CI, −0.6 to −0.2).

Conclusions The QoL of children with CHD who experienced a hospital intervention is reduced at age 8 years. Patient age at the last cardiac intervention might influence QoL at 8 years. *(J Pediatr 2022; ■:1-6)*.

Congenital heart defects (CHDs) are the leading group of congenital anomalies and remain the most important cause of infant deaths linked to congenital malformations.[1,2] Despite major improvements in medical and surgical management leading to increased survival,[3,4] long-term morbidities, particularly neurodevelopmental and mental health issues, remain a cause for concern.[5] Thus, the question of health-related quality of life (QoL) in children with CHD is a key issue.

From the time of prenatal diagnosis of CHD, parents' immediate concerns focus not only on survival, but also on their child's future QoL.[6,7] Improved antenatal screening strategies have led to the consideration of QoL issues early in the perinatal path of these families.[3] The decision to terminate a pregnancy for fetal anomaly is no longer based exclusively on medical issues, but also on the child's expected QoL.[6,7] Studies investigating QoL of children with CHD have been numerous in recent years but have yielded conflicting results.[6,8-11] The observed discrepancies may be related in part to a lack of methodologic consistency, as reported in several systematic reviews,[8-10,12] as well as to the absence of population-based studies including all CHDs.

An important challenge for evaluating outcomes, particularly QoL, is related to the great heterogeneity of CHD in terms of prevalence, pathophysiology, and management. To limit this challenge, QoL studies have either focused on specific types of CHD (eg, transposition of the great arteries)[13] or grouped patients according to predefined anatomic and clinical criteria[6] or to severity of the defect.[6,14] Here we sought to define homogeneous groups of patients according to their medical and surgical management: the need for intervention (surgical or percutaneous) and age at and outcome of intervention (complete repair or palliation). We used data from a prospective population-based cohort study of CHD (the EPICARD study) with 2 objectives: to describe QoL at age 8 years in children born with a CHD reported by the children and by their parents, and to evaluate the association between QoL and the medical and surgical management of children with isolated CHD.

Methods

The EPICARD study is a population-based, prospective cohort study of children with a CHD born in the greater Paris area. All cases (ie, live births, terminations

CHD	Congenital heart defect
QoL	Quality of life

From the [1]Center of Research in Epidemiology and Statistics, Inserm, INRA, University of Paris, Paris, France; [2]M3C-Pediatric Cardiology, Necker Enfants Malades, AP-HP, University of Paris, Paris, France; [3]Physiology and Experimental Medicine Heart Muscles, UMR CNRS 9214, Inserm U1046, University of Montpellier, Montpellier, France; [4]Department of Psychiatry, Harvard Medical School, Boston Children's Hospital, Boston, MA; [5]Paediatric and Congenital Cardiology Department, M3C National Reference Centre, Bordeaux University Hospital, IHU Liryc, Electrophysiology and Heart Modelling Institute, Bordeaux University Foundation, Pessac, France; and [6]Pediatric Emergency Department, Faculty of Medicine, AP-HP, Armand Trousseau Hospital, Sorbonne University, Paris, France

Supported by grants from the French Ministry of Health (PHRC 2004, 2008 and 2011), Association pour l'Etude et la Recherche des Maladies Cardiovasculaires, Instituts Thématiques Multi-Organismes, Association pour la Recherche en Cardiologie du Fœtus à l'Adulte, and Fédération Française de Cardiologie. The authors declare no conflicts of interest.

https://doi.org/10.1016/j.jpeds.2022.01.003

of pregnancy for fetal anomalies, and intrauterine fetal deaths) diagnosed in the prenatal period or up to age 1 year between 2005 and 2008 were eligible for inclusion.[16] The planned follow-up of children at age 8 years included QoL questionnaires (self and proxy reports).[17]

Eligibility criteria for this study included children with CHD who were alive at the time of the 8-year follow-up (n = 1493) (**Figure**). Of the 1493 eligible children, 238 families (15.9%) refused to participate and 504 (33.7%) were lost to follow-up. Finally, 751 children (50.3%) were included in the study. For our second aim, the association between medical and surgical management and QoL, only children with isolated CHD were included (n = 598; 79.7%). Informed consent was obtained from all study participants, and the study was approved by the French National Committee of Information and Liberty (2013-A00234-14).

The Pediatric Quality of Life Inventory version 4.0 Generic Core Scales (self-report and proxy-report) was used to assess QoL. It is composed of 23 items in 4 dimensions: physical functioning (8 items), emotional functioning (5 items), social functioning (5 items), and school functioning (5 items). Each item is scored on a 5-point Likert scale from 0 (never) to 4 (almost always). Items are reverse-scored and linearly transformed to a scale of 0-100. The 4 dimensions provide 3 summary scores: psychosocial health summary score (including emotional social and school functioning), physical health summary score (physical functioning), and total score (all items).

We first divided the patients with CHD into those with isolated CHD (without chromosomal abnormalities or malformation in other systems) and those with nonisolated CHD (CHD associated with chromosomal abnormalities or malformations in other systems). Then isolated CHD was subdivided according to the characteristics of the medical and surgical management: need for intervention (surgery or transcatheter interventions), whether the interventions led to complete repair or were palliative, and the age of the last intervention. Finally, we anticipated that the age at which the intervention occurs could influence the perceived QoL for the children and their caregivers. Early childhood memories (ie, long-term episodic memories) reportedly are more consistent from preschool age (3-6 years),[18,19] and the recollective experiences of childhood events (childhood amnesia theory) are scarce before age 3 years.[20]

We defined the following subgroups: (1) CHDs with spontaneous resolution that do not require cardiac follow-up (reference subgroup), (2) CHDs that require follow-up in an outpatient clinic without planned intervention (surgical or catheterization) at age 8 years, (3) complete repair before age 3 years (ie, 1 or more interventions before 3 years and none thereafter), (4) complete repair after 3 years (ie, at least 1 intervention that took place after 3 years), and (5) palliative repair, including univentricular heart palliation with total cavopulmonary connection.

Descriptive data are presented as proportion for categorical variables and as mean and SE for continuous variables. The first part of the analysis consisted of an assessment of QoL scores for all CHD, isolated CHD, and nonisolated CHD. Then QoL scores reported by parents and children in the isolated CHD and nonisolated CHD groups were compared using the Wilcoxon rank-sum test. The correlation between self-reported and parent-reported QoL scores was examined using the Spearman correlation coefficient (rs). Coefficient values >0.1, 0.3, and 0.5 were considered to indicate a weak, medium, or strong linear relationship of correlations, respectively.

Then multiple linear regression was used to compare QoL scores of each of the medical and surgical management subgroups with the reference subgroup. Estimates from the regression models are reported with regression (β) coefficients and 95% CIs. We considered the potentially confounding effects of sex, prematurity, maternal geographic origin, and maternal and paternal occupation. A sensitivity analysis was performed by adding the covariate CHD complexity[21] to the previous model. We also calculated the effect size (δ) to provide clinically meaningful information about the differences between each CHD subgroup and the reference subgroup. Effect sizes of 0.2, 0.5, and 0.8 were considered small, medium, and large effect sizes, respectively.[22,23] Thus, a P value <.05 was defined as significant. All statistical analyses were performed with Stata version 15 (StataCorp). We followed the STROBE guidelines for reporting the study.

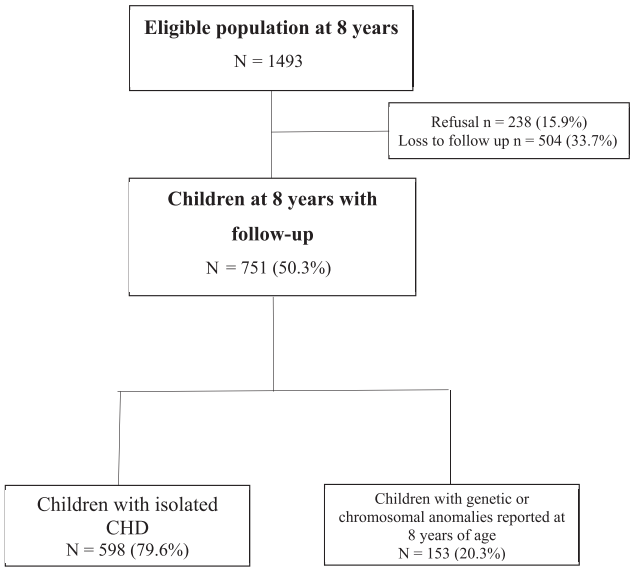

Figure. Flow chart for the study population.

Results

Patient characteristics (n = 751), as well as a comparison between isolated CHD and nonisolated CHD, are presented in **Table I** (available at www.jpeds.com). Specific comparisons of the participants with isolated CHD (n = 598) and

nonparticipants with isolated CHD are shown in Table II (available at www.jpeds.com). Participants were more likely to have a higher socioeconomic status or parental education level, French nationality, and a complex CHD but less likely to have a prenatal diagnosis compared with the nonparticipants. Among the 598 participants with isolated CHD, the number of patients in the medical and surgical management subgroups varied from 16 (2.8%) for the group with CHD who underwent palliative repair to 246 (41.1%) for the group with CHD who underwent complete repair before age 3 years (Table III; available at www.jpeds.com). As shown in Table III, the distribution of children with prenatal diagnosis, the complexity of the CHD at birth, and the sex distribution differed significantly across these subgroups.

As shown in Table IV (available at www.jpeds.com), there were no significant differences in QoL scores according to the mode of follow-up (onsite vs postal mail). Overall, the mean self-reported total QoL score was 75.5 ± 0.1 (61.5 ± 0.2 for the physical score and 89.8 ± 0.3 for the psychosocial score), whereas the mean parent-reported total score was 80.4 ± 0.5 (86.9 ± 0.6 for the physical score and 76.9 ± 0.6 for the psychosocial score) (Table V). When comparing self-reported and parent-reported QoL scores, there was a medium positive correlation for psychosocial and total QoL scores (rs = 0.40 [$P < .001$] and rs = 0.43 [$P < .001$], respectively) but a weak correlation for physical scores (rs = 0.21; $P < .001$).

As shown in Table V, the mean self-reported total and psychosocial scores were significantly lower in the nonisolated CHD group compared with the isolated CHD group (73.9 ± 0.4 vs 75.8 ± 0.2 [$P < .001$] vs 91.9 ± 0.4 vs 92.3 ± 0.2 [$P < .001$]), whereas there was no difference for the physical score. The Cohen d effect size varied between $\delta = -0.3$ (95% CI, −0.5 to −0.1) to $\delta = -0.4$ (95% CI, −0.7 to −0.3), indicating a small effect size between these 2 subgroups (Table VI).

The mean parent-reported total, physical, and psychosocial scores were significantly lower for the nonisolated CHD group compared with the isolated CHD group (70.9 ± 1.5 vs 82.7 ± 0.6 [$P < .001$], 76.5 ± 1.9 vs 89.5 ± 0.6 [$P < .001$], and 68.1 ± 1.4 vs 79.1 ± 0.6 [$P < .001$], respectively) (Table V). The Cohen d effect size varied between $\delta = -0.7$ (95% CI, −0.9 to −0.5) and $\delta = -0.8$ (95% CI, −1.0 to −0.6), indicating a large effect size between these 2 subgroups (Table VI).

Compared with the reference subgroup (76.7 ± 0.2), self-reported total QoL scores were lower in all subgroups (Table V), with a large effect size for the palliative repair subgroup (Table VI). In the multivariable model, the self-reported QoL total scores were significantly lower for the complete repair before 3 years and palliative repair subgroups ($\beta = -1.3$ [95% CI, −1.9 to −0.6] and $\beta = -2.1$ [95% CI, −3.9 to −0.2], respectively) (Table VII).

Compared with the reference subgroup (84.9 ± 0.9), parent-reported total QoL scores were lower in all subgroups (Table V), with a large effect size for the complete repair after 3 years and palliative repair subgroups (Table VI). In the multivariable model, the parent-reported total scores were significantly lower for the complete repair after 3 years and palliative repair subgroups ($\beta = -9.2$ [95% CI, −15.0 to −3.5] and $\beta = -16.0$ [95% CI, - 22.4 to −9.5], respectively) (Table VII).

Compared with the reference subgroup (93.4 ± 0.2), self-reported psychosocial QoL scores were lower in all subgroups (Table V) with a large effect size for the complete repair after 3 years and palliative repair subgroups (Table VI). In the multivariable model, self-reported psychosocial scores were significantly lower for the complete repair before 3 years, complete repair after 3 years, and palliative repair subgroups ($\beta = -1.8$ [95% CI, −2.8 to −0.9], $\beta = -2.5$ [95% CI, −4.8 to −0.4], and $\beta = -3.1$ [95% CI, −5.6 to −0.6], respectively) (Table VII).

Compared with the reference subgroup (81.5 ± 1.0), parent-reported psychosocial QoL scores were lower in all subgroups (Table V) with a large effect size for the complete repair after 3 years and palliative repair subgroups (Table VI). In the multivariable model, parent-reported scores were significantly lower for the complete repair after 3 years and palliative repair subgroups ($\beta = -10.3$ [95% CI, −16.8 to −3.8] and $\beta = -11.6$ [95% CI, −18.8 to −4.3], respectively) (Table VII).

Table V. Self-reported and parent-reported QoL scores

	Self-reported			Parent-reported		
Groups	**Physical score**	**Psychosocial score**	**Total score**	**Physical score**	**Psychosocial score**	**Total score**
All CHD (n = 751)	61.5 ± 0.2	89.8 ± 0.3	75.5 ± 0.1	86.9 ± 0.6	76.9 ± 0.6	80.4 ± 0.5
Nonisolated CHD (n = 153)	60.1 ± 0.4	91.9 ± 0.4*	73.9 ± 0.4*	76.5 ± 1.9*	68.1 ± 1.4*	70.9 ± 1.5*
Isolated CHD (n = 598)	61.8 ± 0.2	92.3 ± 0.2*	75.8 ± 0.2*	89.5 ± 0.6*	79.1 ± 0.6*	82.7 ± 0.6*
CHD with spontaneous resolution not requiring follow-up (reference) (n = 227)	62.4 ± 0.3	93.4 ± 0.2	76.7 ± 0.2	91.2 ± 0.8	81.5 ± 1.0	84.9 ± 0.9
Follow-up outclinic without intervention at 8 y (n = 86)	61.3 ± 0.5	92.9 ± 0.4	75.8 ± 0.4	90.4 ± 1.4	80.0 ± 1.5	83.5 ± 1.3
Complete repair before 3 y (n = 246)	61.4 ± 0.3	91.4 ± 0.4	75.3 ± 0.3	89.8 ± 0.9	78.3 ± 1.0	82.3 ± 0.8
Complete repair after 3 y (n = 23)	61.9 ± 1.1	90.0 ± 1.7	74.8 ± 1.1	81.9 ± 4.7	68.9 ± 3.7	73.3 ± 3.1
Palliative repair (n = 16)	60.9 ± 1.1	90.1 ± 1.6	74.3 ± 1.1	66.7 ± 5.5	70.0 ± 4.3	68.8 ± 4.2

P values are for comparisons between the isolated CHD and nonisolated CHD groups.
*$P < .001$.

Table VI. Cohen d effect size

Variables	Self-reported			Parent-reported		
	Physical score, δ (95% CI)	Psychosocial score, δ (95% CI)	Total score, δ (95% CI)	Physical score, δ (95% CI)	Psychosocial score, δ (95% CI)	Total score, δ (95% CI)
Isolated CHD vs nonisolated CHD	-0.3 (-0.5 to -0.1)*	-0.4* (-0.7 to -0.3)	-0.4* (-0.7 to -0.3)	-0.75* (-0.93 to -0.57)	-0.73* (-0.92 to -0.55)	-0.8* (-1.0 to -0.65)
Follow-up in an outclinic without intervention at 8 y vs reference group	-0.2 (-0.5 to 0.1)	-0.1 (-0.4 to 0.1)	-0.2 (-0.5 to -0.0)	-0.1 (-0.3 to 0.2)	-0.1 (-0.3 to 0.1)	-0.1 (-0.4 to 0.1)
Complete repair before 3 y vs reference group	-0.2 (-0.39 to 0.0)	-0.4* (-0.6 to -0.2)	-0.4* (-0.6 to -0.2)	-0.1 (-0.3 to 0.1)	-0.2* (-0.4 to -0.1)	-0.2* (-0.39 to -0.1)
Complete repair after 3 y vs reference group	-0.1 (-0.5 to 0.3)	-0.9* (-1.3 to -0.4)	-0.5* (-1.0 to -0.10)	-0.7* (-1.1 to -0.2)	-0.9* (-1.3 to -0.4)	- 0.9* (-1.4 to -0.5)
Palliative repair vs reference group	-0.3 (-0.8 to 0.2)	-0.9* (-1.4 to -0.4)	-0.7* (-1.3 to -0.2)	-1.8* (-2.3 to -1.3)	-0.8* (-1.3 to -0.3)	-1.3*(-1.8 to -0.7)

Reference group: CHDs with spontaneous resolution that do not require cardiac follow-up. A δ coefficient of 0.2 is considered a small effect size, δ of 0.5 is considered a medium effect size, δ of 0.8 is considered a large effect size, and δ of 1.3 is considered a very large effect size.
*$P < .05$.

Table VII. Comparison of self-reported and parent-reported QoL according to medical and surgical management, multivariate analyses

Management	Self-reported QoL scores			Parent-reported QoL scores		
	Physical score, adjusted ß (95% CI)	Psychosocial score, adjusted ß (95% CI)	Total score, adjusted ß (95% CI)	Physical score, adjusted ß (95% CI)	Psychosocial score, adjusted ß (95% CI)	Total score, adjusted ß (95% CI)
Follow-up in an outclinic without intervention at 8 y	-0.9 (-2.4 to 0.0)	-0.5 (-1.7 to 0.8)	-0.7 (-1.7 to 0.2)	-0.2 (-3.6 to 3.9)	-1.5 (-5.1 to 2.2)	-0.9 (-4.2 to 2.3)
Complete repair before 3 y	-0.9 (-4.2 to 0.0)	-1.8* (-2.8 to -0.9)	-1.3- (-1.9 to -0.6)	-1.2 (-3.8 to 1.5)	-2.3 (-5.2 to 0.3)	-2.1 (-4.4 to 0.4)
Complete repair after 3 y	-0.1 (-2.1 to 2.2)	-2.5* (-4.8 to -0.4)	-1.1 (-2.8 to 0.6)	-6.6* (-13.1 to -0.1)	-10.3* (-16.8 to -3.8)	-9.2* (-15.0 to -3.5)
Palliative repair	-1.2 (-3.6 to 1.2)	-3.1* (-5.6 to -0.6)	-2.1* (-3.9 to -0.2)	-24.2* (-31.6 to -6.9)	-11.6* (-18.8 to -4.3)	-16.0* (-22.4 to -9.5)

Covariates of adjustment: sex, prematurity, maternal origin in 4 classes, maternal and paternal occupation in 5 classes, complexity of CHD in 3 classes.
*P value $< .05$.

Compared with the reference subgroup (62.4 ± 0.3), self-reported physical QoL scores were lower in all subgroups (Table III). In the multivariable model, self-reported physical scores were not significantly different for any subgroups of CHD (Table VI).

Compared with the reference subgroup (91.2 ± 0.8), parent-reported physical QoL scores were lower in all subgroups (Table V), with a large effect size for the palliative repair subgroup (Table VI). In the multivariable model, parent-reported scores were significantly lower for the complete repair after 3 years and palliative repair subgroups ($\beta = -6.6$ [95% CI, −13.1 to −0.1] and $\beta = -24.2$ [95% CI, −31.6 to −16.9], respectively) (Table VII).

The results of the sensitivity analysis (multivariate model with covariable complexity) showed no significant variation from the model presented (Table VIII; available at www.jpeds.com). The CHD complexity (complex vs simple CHD) was not associated with self- or parent-reported total QoL scores ($\beta = -0.6$ [95% CI, −1.7 to 0.4] and $\beta = -2.1$ [95% CI, −1.6 to 5.8], respectively).

Discussion

Our results show lower self- and parent-reported QoL scores in children with nonisolated CHD compared with children with isolated CHD. However, although self-reported total scores were lower in the nonisolated CHD group, children with nonisolated CHD reported better QoL than their parents overall. These results suggest that comorbidities have an effect on QoL, but that this effect may be milder from the child's perspective.

Furthermore, QoL scores of children with isolated CHD were associated with the characteristics of their medical and surgical management, regardless of the complexity of the CHD. Children and parents who had hospital experiences reported lower QoL than the reference subgroup. For both children and parents, the main dimension impacting QoL was the psychosocial dimension, with similar effect sizes reported by both. Moreover, we observed a moderate correlation between the psychosocial score reported by children and parents. The exception was the palliative repair subgroup, in which parents considered that QoL was related mainly to the physical dimension. Finally, this study shows that children who underwent complete repair after age 3 years reported lower psychosocial QoL scores, and those who underwent palliative repair reported the lowest QoL scores and might be considered at higher risk of QoL reduction at age 8 years.

Our grouping of patients by management characteristics might be questioned, as it has not been used previously for this type of study. Several studies evaluating different clinical characteristics (eg, type of heart defect,[24] duration of circulatory arrest,[25] number of cardiac surgeries[25,26]) found no significant association with QoL. According to Costello et al, clinicians performed poorly when asked to predict QoL for children with CHD[27]; therefore, we grouped patients according to their own life experiences and not according to clinical characteristics. Our hypotheses are based in part on the work of Lawford and Eiser, who considered that the child's ability to adapt to their own experiences and the way in which negative experiences are interpreted will have an impact on QoL.[15] Therefore, it was important to identify common experiences and particularly to define the age at which the memory of these experiences could influence the child. We propose that an experience can influence a child's QoL perception as soon as the child has the ability to remember the intervention (remembered events as a personal experience, ie, episodic memory). Long-term episodic memory develops from preschool age (3-6 years),[18,19] whereas childhood recollection of events occurring before age 3 years (ie, childhood amnesia theory) has not truly emerged.[20] Our results show that children with CHD who underwent surgical interventions after age 3 years report lower psychosocial QoL scores. These findings are in line with the fact that the age of intervention plays a role in the perception of long-term QoL.

Our results also raise questions as to how parents' experience of hospitalization may contribute to the child's future perception of their QoL. The correlations between the psychosocial QoL scores as reported by the parents and child suggest that parental experiences may lead them to show anxiety-related behaviors toward their child (eg, overprotection, anxious attachment). Consequently, this may influence their children's perception of QoL. It is necessary to better understand how the child's long-term overall well-being might depend in part on the lived experience of the parents (ie, the hospitalization of their child) and their own mental health outcomes. Thus, systematic psychological care and support for each family with difficult medical care pathways is important. It is not only a question of one-time support in the acute phase; long-term follow-up may support the development and maintenance of psychological resilience.[15,28] Noteworthy, healthcare systems should adapt to these emerging needs to allow access to care for the most vulnerable groups. The long-term cost/benefit would be largely positive, as parents and children would likely improve their mental health outcomes and reduce societal financial costs.

Our study has several limitations. Given that a large proportion of families completed the questionnaire at home, the question of measurement bias may be raised. However, comparisons with the different populations (Table IV) showed similar scores, supporting the absence or minimization of this potential bias. As in many cohort studies with a long follow-up period, outcome data were not available for a considerable proportion of the eligible study population. Our participants were more likely to have a higher socioeconomic status and a complex CHD. Moreover, there was a large difference in sample size between our subgroups of CHD. This is an inherent issue when forming CHD subgroups but also might indicate a bias of ascertainment. For these reasons, interpretation of the external validity of our study should be done with caution. cautiously.

We did not include children without CHD as the reference group. However, our reference group included children with CHD with spontaneous resolution within the first year of life

considered to have a normal heart. Finally, the group of children who underwent complete repair after age 3 years should have been more thoroughly explored. It is important to assess whether experience of a CHD that requires a single catheterization procedure (eg, atrial septal defect) will have the same impact as multiple surgical procedures performed after age 3 years for another complex CHD. The most at-risk groups are children who underwent an intervention after age 3 years and children who had palliative repair of their CHD. Psychosocial aspects of quality of life appear to be the most impacted in this group, with similar levels of concern reported by both parent and child. ■

We thank all the families for their participation in this study. Many thanks to Ali Ghanchi for proofreading.

Submitted for publication Sep 2, 2021; last revision received Dec 25, 2021; accepted Jan 10, 2022.

Reprint requests: Neil Derridj, MD, MPH, EPOPé, Center of Research in Epidemiology and Statistics, Inserm U 1153. E-mail: neil.derridj@inserm.fr

Data Statement

Data sharing statement available at www.jpeds.com.

References

1. van der Linde D, Konings EEM, Slager MA, Witsenburg M, Helbing WA, Takkenberg JJM, et al. Birth prevalence of congenital heart disease worldwide: a systematic review and meta-analysis. J Am Coll Cardiol 2011;58:2241-7.
2. GBD 2017 Congenital Heart Disease Collaborators. Global, regional, and national burden of congenital heart disease, 1990-2017: a systematic analysis for the Global Burden of Disease Study 2017. Lancet Child Adolesc Health 2020;4:185-200.
3. Khoshnood B, De Vigan C, Vodovar V, Goujard J, Lhomme A, Bonnet D, et al. Trends in prenatal diagnosis, pregnancy termination, and perinatal mortality of newborns with congenital heart disease in France, 1983-2000: a population-based evaluation. Pediatrics 2005;115:95-101.
4. Raissadati A, Nieminen H, Jokinen E, Sairanen H. Progress in late results among pediatric cardiac surgery patients: a population-based 6-decade study with 98% follow-up. Circulation 2015;113:347-53.
5. Mebius MJ, Kooi EMW, Bilardo CM, Bos AF. Brain injury and neurodevelopmental outcome in congenital heart disease: a systematic review. Pediatrics 2017;140:e20164055.
6. Amedro P, Dorka R, Moniotte S, Guillaumont S, Fraisse A, Kreitmann B, et al. Quality of life of children with congenital heart diseases: a multicenter controlled cross-sectional study. Pediatr Cardiol 2015;36:1588-601.
7. Bensemlali M, Stirnemann J, Le Bidois J, Lévy M, Raimondi F, Hery E, et al. Discordances between pre-natal and post-natal diagnoses of congenital heart diseases and impact on care strategies. J Am Coll Cardiol 2016;68:921-30.
8. Latal B, Helfricht S, Fischer JE, Bauersfeld U, Landolt MA. Psychological adjustment and quality of life in children and adolescents following open-heart surgery for congenital heart disease: a systematic review. BMC Pediatr 2009;9:6.
9. Drakouli M, Petsios K, Giannakopoulou M, Patiraki E, Voutoufianaki I, Matziou V. Determinants of quality of life in children and adolescents with CHD: a systematic review. Cardiol Young 2015;25:1027-36.
10. Ladak LA, Hasan BS, Gullick J, Gallagher R. Health-related quality of life in congenital heart disease surgery in children and young adults: a systematic review and meta-analysis. Arch Dis Child 2019;104:340-7.
11. Idorn L, Jensen AS, Juul K, Overgaard D, Nielsen NP, Sørensen K, et al. Quality of life and cognitive function in Fontan patients, a population-based study. Int J Cardiol 2013;168:3230-5.
12. Bertoletti J, Marx GC, Hattge Júnior SP, Pellanda LC. Quality of life and congenital heart disease in childhood and adolescence. Arq Bras Cardiol 2014;102:192-8.
13. Culbert EL, Ashburn DA, Cullen-Dean G, Joseph JA, Williams WG, Blackstone EH, et al. Quality of life of children after repair of transposition of the great arteries. Circulation 2003;108:857-62.
14. Moons P, Van Deyk K, De Geest S, Gewillig M, Budts W. Is the severity of congenital heart disease associated with the quality of life and perceived health of adult patients? Heart 2005;91:1193-8.
15. Lawford J, Eiser C. Exploring links between the concepts of quality of life and resilience. Pediatr Rehabil 2001;4:209-16.
16. Khoshnood B, Lelong N, Houyel L, Thieulin AC, Jouannic JM, Magnier S, et al. Prevalence, timing of diagnosis and mortality of newborns with congenital heart defects: a population-based study. Heart 2012;98:1667-73.
17. Houyel L, Khoshnood B, Anderson RH, Lelong N, Thieulin AC, Goffinet F, et al. Population-based evaluation of a suggested anatomic and clinical classification of congenital heart defects based on the International Paediatric and Congenital Cardiac Code. Orphanet J Rare Dis 2011;6:64.
18. Tulving E. Episodic memory: from mind to brain. Annu Rev Psychol 2002;53:1-25.
19. Scarf D, Gross J, Colombo M, Hayne H. To have and to hold: episodic memory in 3- and 4-year-old children. Dev Psychobiol 2013;55:125-32.
20. Perner J, Ruffman T. Episodic memory and autonoetic consciousness: developmental evidence and a theory of childhood amnesia. J Exp Child Psychol 1995;59:516-48.
21. Warnes CA, Liberthson R, Danielson GK, Dore A, Harris L, Hoffman JI, et al. Task Force 1: the changing profile of congenital heart disease in adult life. J Am Coll Cardiol 2001;37:1170-5.
22. Cohen J. Statistical power analysis for the behavioral sciences. New York: Academic Press; 2013.
23. Rosenthal JA. Qualitative descriptors of strength of association and effect size. J Soc Serv Res 1996;21:37-59.
24. Spijkerboer AW, Utens EMWJ, De Koning WB, Bogers AJJC, Helbing WA, Verhulst FC. Health-related Quality of Life in children and adolescents after invasive treatment for congenital heart disease. Qual Life Res 2006;15:663-73.
25. Dunbar-Masterson C, Wypij D, Bellinger DC, Rappaport LA, Baker AL, Jonas RA, et al. General health status of children with D-transposition of the great arteries after the arterial switch operation. Circulation 2001;104:I138-42.
26. McCrindle BW, Williams RV, Mitchell PD, Hsu DT, Paridon SM, Atz AM, et al. Relationship of patient and medical characteristics to health status in children and adolescents after the Fontan procedure. Circulation 2006;113:1123-9.
27. Costello JM, Mussatto K, Cassedy A, Wray J, Mahony L, Teele SA, et al. Prediction by clinicians of quality of life for children and adolescents with cardiac disease. J Pediatr 2015;166:679-83.e2.
28. Helmreich I, Kunzler A, Chmitorz A, König J, Binder H, Wessa M, et al. Psychological interventions for resilience enhancement in adults. Cochrane Database Syst Rev 2017;2:CD012527.

Table I. Characteristics of the study patients and comparison of children with isolated CHD and nonisolated CHD

Characteristics	Nonisolated CHD (N = 153)	Isolated CHD (N = 598)	All CHD (N = 751)	*P* value
Male sex, %	50.33	50.50	50.47	.96
Prenatal diagnosis, %	24.18	26.09	25.70	.63
Mean term of birth*	37.7	38.3	38.2	.01
Prematurity, %	22.88	16.08	17.47	.04
Weight at birth, kg. mean	2.90	3.08	3.05	.23
Small for gestational age, %	15.03	12.73	13.20	.45
CHD complexity at birth, %				<.01
Simple	43.14	58.19	55.13	
Moderate	45.75	26.76	30.63	
Complex	11.11	15.05	14.25	
Maternal age, y, %				<.01
≥29	30.07	34.95	33.95	
30-34	30.72	38.46	36.88	
35-39	25.49	20.07	21.17	
≥40	13.73	6.52	7.99	
Maternal geographic origin, %				.03
France	43.14	54.61	52.27	
North Africa	22.22	17.92	18.80	
Sub-Saharan Africa	16.34	9.88	11.20	
Others	18.30	17.59	17.73	
Maternal occupation, %				<.01
Unemployed	31.03	17.87	20.50	
Managers	21.38	27.66	26.41	
Intermediate profession	17.93	24.40	23.11	
Employee	11.03	14.78	14.03	
Others	18.62	15.29	15.96	
Level of maternal education, %†				.004
None	8.67	4.03	4.97	
Elementary/junior high	28.00	17.82	19.87	
High school	10.67	11.60	11.41	
University education (up to bachelors)	34.67	42.18	40.67	
University studies (masters or doctoral studies)	18.00	24.37	23.09	
Department of residence at the birth of the child, %				.31
Paris	29.41	34.62	33.56	
Hauts-de-Seine	28.76	26.25	26.76	
Seine-Saint-Denis	26.14	20.57	21.70	
Val-de-Marne	15.69	18.56	17.98	
Level of paternal education, %				.29
None	3.45	3.50	3.49	
Elementary/junior high	30.34	22.42	24.02	
High school	11.72	13.13	12.85	
University education (up to bachelors)	32.41	32.40	32.40	
University studies (masters or doctoral studies)	22.07	28.55	27.23	
Paternal occupation, %				.047
Unemployed	7.43	4.14	4.81	
Managers	36.49	43.10	41.76	
Intermediate profession	10.14	7.59	8.10	
Employee	12.16	18.45	17.17	
Others	33.78	26.72	28.16	

The χ^2 test was used for categorical covariables, and the *t* test or ANOVA was used to compare continuous variables.
*Weeks of gestation.
†Data collected at 3 years.

Table II. Characteristics of participants and nonparticipants with isolated CHD

Characteristics	Nonparticipants (N = 598)	Participants (N = 598)	*P* value*
Male sex, %	44.6	50.5	.04
Prenatal diagnosis, % (N = 1196)	13.4	26.1	<.001
Mean term of birth, mean ± SD†	38.4 ± 2.6	38.3 ± 2.6	.95
Prematurity, % (N = 1193)	13.1	16.1	.14
Weight at birth, kg, mean ± SD (N = 1194)	3.1 ± 0.6	3.1 ± 0.7	.38
Small for gestational age, % (N = 1194)	10.1	12.7	.15
CHD complexity, % (N = 1196)			<.001
Simple	75.8	58.2	
Moderate	17.4	26.8	
Complex	6.9	15.1	
Maternal age, y, % (N = 1185)			.08
≤29	41.7	34.9	
30-34	36.1	38.5	
35-39	17.4	20.1	
≥40	4.8	6.5	
Maternal geographic origin, % (N = 1192)			.027
France	46.9	54.6	
North Africa	18.6	17.9	
Sub-Saharan Africa	14.0	9.9	
Others	20.6	17.6	
Maternal occupation (N = 1134)			<.001
Unemployed	33.9	17.0	
Manager	21.4	27.7	
Intermediate profession	18.3	24.4	
Employee	9.4	14.8	
Others	17.0	15.3	
Level of maternal education, % (N = 750)‡			.008
None	7.7	4.0	
Elementary/junior high	23.9	17.8	
High school	17.4	11.6	
University education (up to bachelors)	32.9	42.2	
University studies (masters or doctoral studies)	18.1	24.4	
Level of paternal education, % (N = 721)‡			.004
None	8.0	3.5	
Elementary/junior high	32.7	22.4	
High school	13.3	13.1	
University education (up to bachelors)	22.7	32.4	
University studies (masters or doctoral studies)	23.3	28.6	
Department of residence at the birth of the child, % (N = 1196)			.5
Paris	32.6	34.6	
Hauts-de-Seine	26.6	26.2	
Seine-Saint-Denis	23.9	20.6	
Val-de-Marne	16.9	18.6	

**P* values comparing participants and nonparticipants.
†Weeks of gestation.
‡Data collected at 3 years.

Table III. Patient characteristics according to medical and surgical management

Groups	No cardiac follow-up (N = 227), %	Follow-up in consultation (N = 86), %	Complete repair before age 3 y (N = 246), %	Complete repair after age 3 y (N = 23), %	Palliative repair (N = 16), %	*P* value
Prenatal diagnosis	6.17	17.44	42.28	43.48	81.25	<.001
Prematurity*	20.80	13.95	13.41	8.70	12.50	.17
Small for gestational age	10.18	16.28	13.41	21.74	6.25	.22
Male sex (male)	40.09	48.84	58.94	60.87	62.50	.001
CHD complexity at birth						<.001
Simple	85.02	82.56	30.08	39.13	0	
Moderate	14.10	17.44	40.24	56.52	0	
Complex	0.88	0.00	29.67	4.35	100	
Maternal age, y						.58
≤29	31.28	38.37	37.40	39.13	25.00	
30-34	41.41	37.21	34.55	39.13	62.50	
35-39	22.03	18.60	19.92	13.04	12.50	
≥40	5.29	5.81	8.13	8.70	0.00	
Maternal geographic origin						.19
France	61.95	56.98	47.56	56.52	43.75	
North Africa	14.60	19.77	18.70	26.09	31.25	
Sub-Saharan Africa	8.85	9.30	11.38	8.70	6.25	
Others	14.60	13.95	22.36	8.70	18.75	
Maternal occupation						.49
Unemployed	15.14	16.87	19.67	19.05	31.25	
Manager	30.28	22.89	26.64	23.81	37.50	
Intermediate profession	21.56	32.53	24.59	28.57	12.50	
Employee	18.81	12.05	13.52	9.52	0.00	
Others	14.22	15.66	15.57	19.05	18.75	
Maternal education level						.51
None	6.19	1.16	3.27	4.55	0.00	
Elementary/junior high	17.70	16.28	16.73	31.82	25.00	
High school	8.41	11.63	14.29	13.64	12.50	
University education (up to bachelors)	40.71	48.84	42.04	31.82	43.75	
University studies (masters or doctoral studies)	26.99	22.09	23.67	18.18	18.75	
Paternal education level						.33
None	5.05	3.85	1.69	9.09	0.00	
Elementary/junior high	20.18	23.08	23.63	27.27	25.00	
High school	11.47	11.54	15.61	4.55	18.75	
University education (up to bachelors)	29.36	38.46	33.76	22.73	37.50	
University studies (masters or doctoral studies)	33.94	23.08	25.32	36.36	18.75	
Paternal occupation						.15
Unemployed	4.91	2.53	3.35	4.55	12.50	
Manager	47.77	36.71	41.42	45.45	31.25	
Intermediate profession	6.25	10.13	7.95	0.00	18.75	
Employee	17.41	27.85	17.99	9.09	6.25	
Others	23.66	22.78	29.29	40.91	31.25	
Maternal place of residence (department)						.29
Paris	37.89	41.86	29.27	26.09	43.75	
Hauts-de-Seine	27.31	20.93	26.83	34.78	18.75	
Seine-Saint-Denis	15.86	22.09	23.98	17.39	31.25	
Val-de-Marne	18.94	15.12	19.92	21.74	6.25	

The χ^2 test was used for categorical covariables, and the *t* test or ANOVA was used to compare continuous variables.
*Less than 37 weeks of gestation.

Table IV. Self-reported and parent-reported QoL scores according to the type of participation: onsite vs postal mail

Variables	Total (N = 598), mean ± SE	Onsite (N = 481), mean ± SE	Postal mail (N = 117), mean ± SE	*P* value
Parents				
Physical score	89.5 ± 0.6	89.7 ± 0.7	89.0 ± 1.5	.70
Psychosocial score	77.1 ± 0.6	77.1 ± 0.7	77.1 ± 1.6	.95
Total score	81.4 ± 0.6	81.5 ± 0.6	81.2 ± 1.4	.85
Children				
Physical score	61.8 ± 0.2	61.8 ± 0.2	61.6 ± 0.4	.70
Psychosocial score	92.3 ± 0.2	92.1 ± 0.2	93.0 ± 0.4	.1
Total score	75.8 ± 0.2	75.8 ± 0.2	76.2 ± 0.3	.28

The *t* test was used to compare variables.

Table VIII. Sensitivity analysis: Multivariable linear regression model

	Self-reported			Parent-reported		
Management	**Physical score, ß (95% CI)**	**Psychosocial score, ß (95% CI)**	**Total score, ß (95% CI)**	**Physical score, ß (95% CI)**	**Psychosocial score, ß (95% CI)**	**Total score, ß (95% CI)**
Follow-up in an outclinic without intervention at 8 y	-0.9 (-2.1 to 0.3)	-0.5 (-1.8 to 0.8)	-0.8 (-1.7 to 0.2)	0.3 (-3.4 to 4.0)	-1.5 (-5.1 to 2.2)	-0.9 (-4.2 to 2.4)
Complete repair before 3 y	-0.4 (-1.4 to 0.6)	-2.0* (-3.1 to -0.9)	-1.1* (-1.8 to -0.3)	-1.6 (-4.7 to 1.5)	-3.1* (-5.2 to 0.0)	-2.6 (-5.4 to 0.1)
Complete repair after 3 y	0.3 (-1.9 to -2.4)	-2.5* (-4.8 to -0.3)	-1.0 (-2.7 to 0.7)	-6.4 (-13.1 to 0.3)	-10.7* (-17.3 to -4.1)	-9.4* (-15.3 to -3.5)
Palliative repair	0.1 (-2.6 to 2.7)	-3.7* (-6.5 to -1.0)	-1.6 (-3.6 to 0.5)	-26.1* (-34.2 to -17.9)	-13.3* (-21.3 to -5.2)	-17.7* (-24.9 to -10.6)

Covariates of adjustment: sex, prematurity, maternal origin in 4 classes, maternal and paternal occupation in 5 classes.
**P* value <.05.