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# Parent Health-Related Quality of Life for Infants with Congenital Anomalies Receiving Neonatal Intensive Care

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# Abstract

## Objective

To examine factors associated with parent quality of life during and after neonatal intensive care unit (NICU) discharge among parents of infants with congenital anomalies admitted to the NICU.

## Study design

This secondary analysis of 2 prospective cohort studies between 2016 and 2020 at a level IV NICU included parents of infants with major congenital anomalies receiving NICU care. The primary outcomes were parent health-related quality of life (HRQL) during the NICU stay and at 3 months post-NICU discharge.

## Results

A total of 166 parent–infant dyads were enrolled in the study, 124 of which completed the 3-month follow-up interview. During the NICU stay, parent history of a mental health disorder (−13 points), earlier gestational age (−17 points), consultation by multiple specialists (−11 points), and longer hospital stay (−5 points) were associated with lower HRQL. Parents of infants with a neonatal surgical anomaly had higher HRQL (+4 points). At 3 months after NICU discharge, parent receipt of a psychology consult in the NICU, the total number of consultants involved in the child's care, and an infant with a nonsurgical anomaly were associated with lower parent HRQL. Parents of infants with a gastrostomy tube (−6 points) and those with hospital readmission (−5 points) had lower HRQL. Comparing same-parent differences in HRQL over time, parents of infants with anomalies did not show significant improvement in HRQL on discharge home.

## Conclusion

Parents of infants with congenital anomalies reported low HRQL at baseline and at discharge. Parents of infants with nonsurgical, medically complex anomalies requiring multispecialty care represent a vulnerable group who could be better supported during and after their NICU stay.



# Keywords

quality of life, NICU, anomalies, surgical anomalies


# Abbreviations

CDH - Congenital diaphragmatic hernia
HRQL - Health-related quality of life
NICU - Neonatal intensive care unit
PedsQL - Pediatric Quality of Life
TEF - Tracheoesophageal fistula

Infants with congenital anomalies represent up to a one-third of children's hospital neonatal intensive care unit (NICU) admissions.[1] Parents of infants with congenital anomalies face significant stressors that impact their health-related quality of life (HRQL), which begin at diagnosis and continue through a prolonged NICU hospitalization and subsequent health needs.[2, 3, 4] We previously found that unlike parents of preterm infants, whose HRQL improves on discharge home, parents of term infants do not follow the same pattern and continue to have low HRQL after NICU discharge.[5] Because many term infants admitted to the NICU are born with congenital anomalies, understanding the factors associated with lower parent HRQL before and after NICU discharge would help target appropriate family counseling and discharge planning.

HRQL is a multidimensional concept of the impact of a person's health status on their overall quality of life.[6] For parents of infants with congenital anomalies, HRQL could be impacted by multiple factors. Infant illness severity has been associated with lower parent HRQL, but characteristics associated with congenital anomalies have not been evaluated previously.[4,5] Multiple factors may affect parent HRQL, including surgery in the neonatal period, multisystem involvement, long-term prognosis, complicating conditions such as growth restriction or prematurity, demographic factors, and postdischarge healthcare utilization.[5,7] The interaction of NICU illness, demographic factors, anomaly characteristics, and postdischarge health utilization as it impacts parent HRQL for infants born with congenital anomalies has not yet been reported.

The objective of this study was to describe the associations between characteristics of neonatal anomalies and parent HRQL while in the NICU and after discharge. We hypothesized that lower parent HRQL would be associated with NICU illness and specific anomaly characteristics (eg, surgically correctable vs not), and that parents of infants with more post-NICU healthcare utilization needs would report lower HRQL after discharge.

## Methods

This was a secondary analysis of 2 prospective cohort studies of parent–infant dyads hospitalized for at least 14 days in a single level IV NICU, the first from November 2016 to July 2017 and the second from September 2018 to March 2020.[5,8] Parent HRQL was measured in both cohorts, but the cohorts differed with respect to other measured outcomes.

Our level IV NICU admits infants of all gestational ages requiring intensive care born at a co-located birth hospital and receives patients transferred for medical and surgical evaluation. The NICU's parent support resources include single-patient rooms with video cameras, psychologists, case managers, social workers, and a family support coordinator. Our NICU admits all infants with congenital heart disease except those requiring cardiac surgery or extracorporeal membrane oxygenation immediately after delivery. All infants with cardiac anomalies who require medical management alone are cared for in our NICU until discharge. Infants who require cardiac surgery are transferred to the cardiac ICU, typically 1-2 days before surgery, and recover in the cardiac intensive care unit unless they are very preterm (<34 weeks), in which case they are transferred back to the NICU after recovering from cardiac surgery. For this study, we included all infants with major congenital anomalies who had been identified by manual chart review in each of the original cohort studies. We included all infants admitted for NICU care for management of either the anomaly alone or another complicating factor in addition to the anomaly (eg, need for respiratory support, prematurity). In accordance with the original study criteria, we excluded non–English-speaking families, nonbiological parents who could not provide consent, infants previously discharged home, infants transferred to cardiac intensive care, and infants for whom death was imminent. Parents of multiples chose 1 child to enroll if both children were eligible.

The primary outcomes were parent HRQL in the NICU and at 3 months after NICU discharge, measured using the validated Pediatric Quality of Life (PedsQL) Family Impact Module.[9] This 36-item self-report tool assesses parents' HRQL related to their child's illness among 8 domains: physical, emotional, social, and cognitive functioning; communication; worry; family relationships; and daily activities. A 5-point Likert scale is used; mean scores are transformed to a 0-100 scale, with higher scores indicating better HRQL.

Because this cohort included infants with a diverse group of anomalies, we categorized them a priori in different ways to assess how specific anomaly characteristics were associated with parent HRQL. Anomalies are traditionally classified by organ system and specific type in birth defect registries and medical textbooks[10, 11, 12]; however, this classification does not necessarily correlate with what a baby might need in the NICU, or what the long-term outlook would be. Thus, we used a classification system that identified babies based on what they might need in the NICU (eg, surgery),[13] how that would impact NICU stay, and long-term prognosis, as described

previously.[13, 14] Therefore, we categorized anomalies by the need for neonatal surgery (neonatal surgical, non-neonatal surgical, nonsurgical); by overall prognosis (moderate, severe, life-limiting), based on previously published research from our group; and by organ system (eg, thoracic, cardiac), with individual types of anomalies included separately within the organ system (eg, congenital diaphragmatic hernia [CDH], gastroschisis). The definitions and examples of included anomalies are listed in Table I (available at www.jpeds.com).

Study procedures for identification of eligible subjects, enrollment, and data collection were similar in the 2 cohorts.[5] Eligible parents were approached for consent by a research assistant; after enrollment, the parents completed the questionnaires using a tablet. Responses were entered directly into a secure database.[15] On enrollment, parents answered demographic questions, including self-reported history of a mental health disorder and the PedsQL Family Impact Module. At discharge, we reviewed the chart for variables that would reflect NICU illness, comorbidities, and discharge medical needs across all anomalies. At 3 months after discharge, healthcare utilization was evaluated using chart review. Parents were contacted 3 months after discharge to repeat the PedsQL Family Impact Module and confirm healthcare utilization; this post-NICU assessment was performed using a secure electronic questionnaire,[15] by phone, or in person depending on parent preference.

### Statistical Analyses

We calculated that a sample size of 111 patients would be sufficient to test for a change in HRQL of 5 points between pre–NICU discharge and post–NICU discharge with 80% power at a probability level of 0.05. Anomaly characteristics were comparable in the baseline and 3-month samples. We compared demographic characteristics, infant illness, and post-NICU healthcare utilization among infants with anomalies. Then we compared NICU and 3-month post-NICU parent HRQL scores by demographic characteristics, anomaly type, infant illness, and post-NICU healthcare use. Between-group comparisons were performed with Kruskal–Wallis or Wilcoxon rank-sum test, chi-square test, or Fisher exact test, as appropriate; within-group changes in HRQL were compared using the paired-samples Wilcoxon signed-rank test. Multivariable regression was used to assess the impact of multiple predictors on baseline HRQL and 3-month HRQL separately. The model included variables with a *P* value <.2 in bivariate analysis. For 3-month HRQL, the baseline HRQL was included as a predictor. This study was approved by the Institutional Review Board at the Children's Hospital of Wisconsin.

## Results

Between July 2016 and February 2020, a total of 514 parent–infant dyads were enrolled in 2 separate cohort studies. The first study enrolled 214 parents, and the second study enrolled 300 parents. In those 2 cohorts, 166 infants (32%) had a major congenital anomaly and were included in the present study; 8 of these infants died in NICU or before the 3-month follow-up postdischarge, and 4 were still in NICU at the end of the study period, leaving 154 parents eligible for follow-up. Of the 154 eligible parents, 124 (81%) completed the 3-month interview. Eighty-eight percent of the enrolled parents were mothers.

The most common infant anomalies were genetic (n = 53; 32%), followed by thoracic, cardiac, craniofacial, and abdominal wall defects (Table II). Based on our prognostic classification, 106 (64%) of the anomalies were classified as moderate severity, 53 (32%) were severe, and 7 (4%) were life-limiting. The majority of the anomalies (102; 61%) were classified as neonatal surgical.

Table II. Bivariate analysis of parent HRQL and parent, infant, illness, and anomaly characteristics

| **Variables** | **NICU (N = 166)** | | | | **3 mo postdischarge (N = 124)** | | | |
|---|---|---|---|---|---|---|---|---|
| | **n** | **%** | **Median (IQR)** | ***P* value** | **n** | **%** | **Median (IQR)** | ***P* value** |
| Parent characteristics | | | | | | | | |
| Age, y | | | | | | | | |
| ≤18 | 3 | 2 | 82 (80-87) | .16 | 3 | 2 | 88 (68-89) | .07 |
| 19-25 | 37 | 22 | 64 (50-81) | | 28 | 23 | 70 (61-82) | |
| 26-35 | 95 | 57 | 67 (55-76) | | 70 | 56 | 63 (52-80) | |
| ≥36 | 31 | 19 | 68 (60-77) | | 23 | 19 | 70 (62-89) | |
| Race/ethnicity | | | | | | | | |
| Black | 30 | 18 | 66 (58-74) | .08 | 19 | 15 | 68 (57-75) | .14 |
| White | 115 | 69 | 66 (52-77) | | 92 | 74 | 67 (52-82) | |
| Hispanic | 8 | 5 | 71 (48-77) | | 5 | 4 | 68 (54-80) | |
| Asian | 4 | 2 | 80 (77-84) | | 3 | 2 | 84 (80-89) | |
| Other | 9 | 5 | 76 (63-90) | | 5 | 4 | 83 (71-94) | |
| Has a car | | | | | | | | |
| Yes | 137 | 83 | 66 (54-76) | .07 | 104 | 84 | 68 (56-82) | .71 |
| No | 29 | 17 | 74 (61-82) | | 20 | 16% | 71 (53-84) | |
| History of mental health disorder* | | | | | | | | |
| Yes | 46 | 28 | 56 (46-68) | **<.001** | 33 | 27% | 64 (50-77) | .20 |
| No | 80 | 48 | 69 (59-78) | | 59 | 48% | 75 (57-88) | |
| Parent psychology consult in NICU | | | | | | | | |
| Yes | 22 | 13 | 50 (42-62) | .06 | 14 | 11 | 56 (48-59) | **<.01** |
| No | 144 | 87 | 68 (65-70) | | 110 | 89 | 69 (57-84) | |
| Number of siblings (for child) | | | | | | | | |
| 0 | 53 | 32 | 69 (55-81) | .28 | 41 | 33 | 71 (58-84) | .15 |
| 1 | 55 | 33 | 64 (52-74) | | 41 | 33 | 67 (49-75) | |
| 2 or more | 58 | 35 | 68 (57-77) | | 42 | 34 | 69 (56-83) | |
| NICU illness characteristics | | | | | | | | |
| Gestational age, wk | | | | | | | | |
| ≤28 | 7 | 4 | 51 (49-76) | .09 | 6 | 5 | 68 (44-72) | .71 |
| 29-36 | 82 | 49 | 65 (53-76) | | 58 | 47 | 71 (56-84) | |
| ≥37 | 77 | 46 | 68 (60-78) | | 60 | 48 | 68 (56-82) | |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| Sex | | | | | | | | |
| Male | 91 | 55 | 67 (52-80) | .98 | 67 | 54 | 64 (52-83) | .17 |
| Female | 75 | 45 | 67 (58-76) | | 57 | 46 | 71 (62-82) | |
| Number of consultants∗ | | | | | | | | |
| 0-1 | 25 | 15 | 77 (64-89) | **.01** | 17 | 14 | 80 (63-87) | **.05** |
| ≥2 | 141 | 85 | 66 (53-76) | | 107 | 86 | 67 (54-81) | |
| Palliative care consulted | | | | | | | | |
| Yes | 16 | 10 | 57 (51-65) | | 10 | 8 | 62 (46-70) | |
| No | 150 | 90 | 68 (56-78) | .07 | 114 | 92 | 68 (57-83) | .08 |
| Received mechanical ventilation | | | | | | | | |
| Yes | 101 | 61 | 66 (53-77) | .14 | 75 | 60 | 67 (54-81) | .06 |
| No | 65 | 39 | 68 (60-80) | | 49 | 40 | 75 (59-87) | |
| Received vasopressors, % | | | | | | | | |
| Yes | 36 | 22 | 68 (55-80) | .48 | 27 | 22 | 74 (62-84) | .18 |
| No | 130 | 78 | 66 (55-76) | | 97 | 78 | 67 (56-82) | |
| Total hospital LOS, d | | | | | | | | |
| ≤28 | 46 | 28 | 69 (62-80) | **.04** | 33 | 27 | 71 (61-85) | .15 |
| 29-60 | 70 | 42 | 68 (55-81) | | 56 | 45 | 67 (53-83) | |
| >61 | 50 | 30 | 64 (51-72) | | 35 | 28 | 68 (48-77) | |
| Anomaly-specific characteristics | | | | | | | | |
| Classification by need for surgery | | | | | | | | |
| Neonatal surgical | 102 | 61 | 69 (57-80) | .13 | 82 | 66 | 71 (57-84) | **.05** |
| Non-neonatal surgical | 26 | 16 | 63 (52-69) | | 19 | 15 | 58 (48-69) | |
| Nonsurgical | 38 | 23 | 65 (53-75) | | 23 | 19 | 68 (58-80) | |
| Classification by prognosis | | | | | | | | |
| Moderate | 106 | 64 | 68 (59-77) | .46 | 81 | 65 | 68 (56-84) | .88 |
| Severe | 53 | 32 | 66 (52-80) | | 41 | 33 | 68 (53-82) | |
| Life-limiting | 7 | 4 | 60 (44-75) | | 2 | 2 | 63 (58-68) | |
| Classification by organ system | | | | | | | | |
| Genetic | 53 | 32 | 64 (52-75) | .14 | 35 | 28 | 66 (49-81) | .16 |
| Associated with a syndrome | 25 | 15 | 62 (53-77) | .85 | 18 | 15 | 62 (39-77) | .76 |
| Thoracic | 30 | 18 | 72 (59-85) | **.04** | 24 | 19 | 69 (56-90) | .52 |
| TEF/EA | 13 | 8 | 67 (55-86) | .69 | 11 | 9 | 73 (58-82) | .71 |
| CDH | 12 | 7 | 76 (68-80) | **.05** | 11 | 9 | 64 (53-91) | .76 |
| CPAM | 3 | 2 | 70 (52-88) | .75 | 1 | 1 | 92 | .16 |
| Pulmonary lymphatic malformation | 2 | 1 | 75 (65-88) | .25 | 1 | 1 | 58 | .50 |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| Cardiac | 29 | 17 | 60 (46-70) | **.02** | 20 | 16 | 59 (48-70) | **.02** |
| CNS | 27 | 16 | 62 (51-75) | .29 | 21 | 17 | 67 (51-74) | .36 |
| Craniofacial | 27 | 16 | 68 (58-76) | .61 | 19 | 15 | 67 (48-82) | .55 |
| Airway | 15 | 9 | 67 (58-74) | .95 | 11 | 9 | 67 (49-89) | .83 |
| Cleft lip/palate | 12 | 7 | 61 (54-74) | .64 | 9 | 7 | 56 (48-81) | .23 |
| Eye | 6 | 4 | 74 (65-86) | .12 | 4 | 3 | 74 (69-90) | .24 |
| Ear | 3 | 2 | 73 (28-77) | .85 | 2 | 2 | 56 (24-89) | .79 |
| Abdominal wall defect | 25 | 15 | 69 (60-81) | .43 | 21 | 17 | 71 (62-84) | .15 |
| Gastroschisis | 20 | 12 | 68 (50-81) | .88 | 16 | 13 | 72 (62-84) | .24 |
| Omphalocele | 4 | 2 | 81 (70-89) | .06 | 4 | 3 | 75 (68-94) | .22 |
| Cloacal exstrophy | 1 | 1 | 73 | .60 | 1 | 1 | 56 | .38 |
| Gastrointestinal | 18 | 11 | 70 (58-81) | .39 | 13 | 10 | 69 (57-87) | .52 |
| Intestinal atresias | 12 | 7 | 65 (60-76) | .75 | 7 | 6 | 73 (57-85) | .53 |
| Hirschsprung disease | 4 | 2 | 83 (60-94) | .12 | 4 | 3 | 88 (71-93) | .05 |
| Imperforate anus | 4 | 2 | 76 (52-83) | .47 | 4 | 3 | 62 (33-77) | .37 |
| Renal/genitourinary | 15 | 9 | 69 (58-75) | .98 | 8 | 6 | 66 (33-86) | .40 |
| Musculoskeletal | 13 | 8 | 59 (52-66) | .10 | 9 | 7 | 62 (47-73) | .24 |
| Multiple anomaly complex | 40 | 24 | 64 (56-77) | .62 | 9 | 7 | 64 (49-80) | .24 |
| Healthcare utilization at NICU discharge | | | | | | | | |
| Home oxygen | | | | | | | | |
| Yes | | | | | 31 | 25 | 67 (49-77) | .20 |
| No | | | | | 93 | 75 | 69 (57-84) | |
| Gastrostomy tube | | | | | | | | |
| Yes | | | | | 48 | 39 | 63 (48-81) | |
| No | | | | | 76 | 61 | 69 (58-84) | .06 |
| Emergency department visits | | | | | | | | |
| None | | | | | 69 | 56 | 69 (57-85) | **.05** |
| ≥1 | | | | | 55 | 44 | 67 (52-77) | |
| Hospital readmissions | | | | | | | | |
| None | | | | | 68 | 55 | 71 (58-84) | .06 |
| ≥1 | | | | | 56 | 45 | 66 (49-80) | |

*CPAM*, congenital pulmonary airway malformation; *CNS*, central nervous system; *EA*, esophageal atresia; *LOS*, length of stay. For parent and infant characteristics, only variables with a *P* value <.2 in either the initial or 3-month evaluation, or the pre-post difference are shown. *P* values ≤ .05 are in bold type. Other variables that were tested and found to be nonsignificant were parent education, distance from hospital, single-parent household, insurance type, presence of a prenatal diagnosis, adults nearby to care for children, birth weight, multiple gestation, number of surgeries, and mode of delivery. ∗ Contains missing values.

Table II shows bivariable associations between parent demographics, NICU illness, and anomaly-specific characteristics with parent HRQL at baseline and at the 3-month follow-up. Parent history of a mental health disorder, consultation by multiple specialists, and longer hospital stay were associated with lower parent HRQL in the NICU. Presence of a neonatal surgical anomaly was associated with higher parent HRQL at 3 months. Anomaly prognostic classification did not impact parent HRQL. Post-NICU healthcare utilization was also associated with 3-month HRQL differences; parents of infants with a gastrostomy tube and those with hospital readmission had lower HRQL.

Table III shows HRQL subdomain scores in the NICU and at 3 months postdischarge. In the NICU, parents of infants with anomalies scored the lowest on the subdomains of worry and daily activities. From the NICU to 3 months postdischarge, daily activity scores declined significantly (−8 points), whereas emotional functioning scores improved significantly (+10 points).

Table III. HRQL subdomain scores for parents of infants with congenital anomalies at baseline and at 3-month follow-up

| **Subdomain** | **NICU (N = 166), median (IQR)** | **3 mo follow-up (N = 124), median (IQR)** | **Change, median (IQR)** | ***P* Value for change** |
|---|---|---|---|---|
| Physical functioning | 62 (50-75) | 67 (50-75) | 0 (−8 to +12) | .58 |
| Emotional functioning | 65 (45-75) | 72 (55-90) | +10 (−5 to +20) | **<.001** |
| Social functioning | 75 (56-87) | 69 (50-94) | 0 (−14 to +15) | .12 |
| Cognitive functioning | 70 (55-90) | 72 (55-90) | 0 (−14 to +15) | .92 |
| Communication | 67 (50-83) | 67 (50-83) | 0 (−8 to +8) | .92 |
| Worry | 60 (45-75) | 65 (50-80) | 0 (−10 to +15) | .16 |
| Daily activities | 58 (42-75) | 50 (33-75) | −8 (−17 to +8) | **<.01** |
| Family relationships | 80 (65-95) | 80 (60-100) | 0 (−10 to +5) | .18 |

*IQR*, interquartile range. *P* values calculated using the paired Wilcoxon rank-sum test. *P* values <.05 are in bold type.

Figure 1 shows parent HRQL associated with specific types of anomalies or organ systems with at least 10 patients for assessment. During the NICU stay, parents of infants with CDH had higher scores than parents of infants with other anomalies; parents of infants with musculoskeletal and cardiac anomalies had lower than median NICU scores. At 3 months postdischarge, infants with CDH had lower scores, and infants with tracheoesophageal fistula (TEF), intestinal atresia, and gastroschisis had higher scores. When comparing parents in the NICU vs at 3 months (Figure 2), parents of infants with intestinal atresias, TEF/esophageal atresia, neurologic anomalies showed improvement over time, whereas parents of infants with CDH, genitourinary anomalies, and cleft lip/palate had decreased HRQL after discharge. Parents of infants with multiple anomaly complex showed no change.

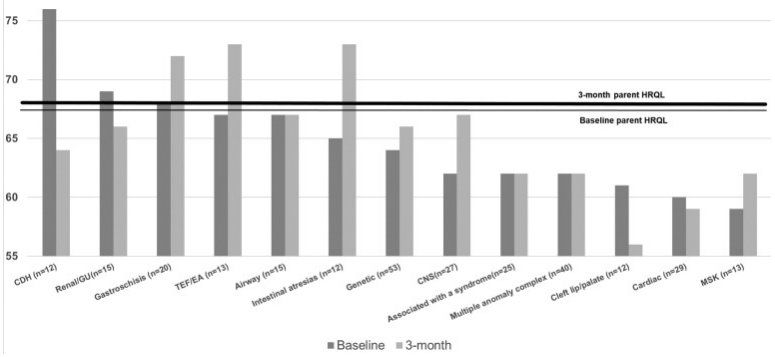

Figure 1. Baseline and 3-month HRQL by individual anomalies or organ systems. The median HRQL score is on the *y*-axis. Only anomalies with >10 patients are shown. EA, esophageal atresia; CNS, central nervous system. ∗*P* < .05 for difference between baseline and 3 month post–NICU discharge HRQL scores.

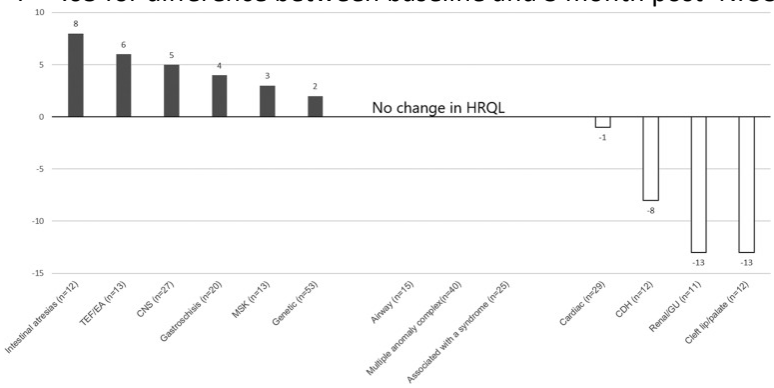


Figure 2. Change in parent HRQL from baseline to 3 months postdischarge by individual anomalies or organ systems. Only anomalies with >10 patients are shown. ∗*P* < .05 for change in HRQL score from baseline to 3 months post–NICU discharge.

Table IV (available at www.jpeds.com) shows the results of a multivariable regression model assessing the effect of multiple predictors on baseline and 3-month parent HRQL. Factors associated with a lower parent HRQL in the NICU were parent history of mental health disorder (9 points lower) and multiple consultants involved in a patient's care (10 points lower). At 3 months, hospital readmissions were associated with an 8-point decrease in parent HRQL compared with their own HRQL in the NICU, adjusted for other covariates.

## Discussion

In this prospective single-center cohort study of infants with anomalies requiring NICU care, we found that parents of infants admitted with major anomalies to the NICU report low HRQL, both at baseline, and after NICU discharge. Parents of infants with neonatal surgical anomalies had higher HRQL, and anomaly prognosis did not affect parent HRQL in the NICU or at follow-up. The main factors associated with lower parent HRQL were poor parent mental health, need for multiple consultants, presence of a syndrome, and need for continued medical care.

Parents of infants admitted with major anomalies to a level IV NICU reported low overall HRQL both in the NICU (median, 67) and at 3 months postdischarge (median, 68). As a reference, the average reported HRQL is approximately 84 in a healthy pediatric population and 74 in those with chronic disease.[16, 17, 18] In our earlier study of infants without anomalies admitted to the NICU, the baseline HRQL was 70 and the 3-month HRQL was 75.[5] In our current cohort of infants with anomalies, at 3 months after NICU discharge, the overall HRQL of their parents did not improve significantly. When we examined subdomain scores, we found that parents of infants with anomalies reported more worry and significantly more difficulty in performing daily activities (eg, daily activities taking more time and effort, difficulty finding the time and energy to finish household tasks), and that this subdomain is negatively affected after discharge to home. The higher proportion of parents reporting a mental health history in our parent cohort also may have affected these subdomain scores. In contrast to our findings in parents of extremely preterm infants, who reported improvement in most subdomain scores on discharge to home, parents of infants with anomalies do not report such an improvement.[5] Research on parents with older children with certain anomalies also have shown effects on their physical and emotional functioning, although this finding is not consistent across studies.[19] Our findings suggest that parents of infants admitted with anomalies are a more vulnerable population than parents of infants requiring NICU admission for prematurity.

We explored different anomaly classification methods (by, eg, prognosis, organ system) as predictors of parent HRQL. Parents of infants with neonatal surgical anomalies reported higher NICU HRQL, and those with nonsurgical anomalies reported lower HRQL. Parents of infants with surgical anomalies such as intestinal atresias, TEF, and gastroschisis had significantly better HRQL, but this was not true for parents of infants with CDH, omphalocele, or imperforate anus. We speculate that the postdischarge health care needs of these infants could at least partly explain the lower parent HRQL after discharge. Amin et al showed that in infants with CDH, the ongoing need for a feeding tube is associated with lower quality of life,[16] as was also seen in our study population. Ongoing care requirements (eg, in omphalocele, colostomy care in infants with an imperforate anus) may impose additional burdens on families.[16,17] In addition, for some anomalies, such as CDH, the lower HRQL may reflect parents recognizing that their baby has a serious chronic medical condition.[20, 21] Some of these families could be better supported by incorporating parent education and better preparation for the care needs after the transition to home. Parents of infants with non-neonatal surgical anomalies, such as cleft lip or palate, also reported lower HRQL after discharge to home. This may be related to ongoing feeding difficulties at home, the need for feeding assistance, association with a syndrome of multiple anomalies, or a physically more obvious defect that has not yet been repaired. The reasons for lower parent HRQL with some of these anomalies needs further exploration.

We acknowledge that HRQL is a dynamic construct that changes over time; a parent's attitude about their child's quality of life likely changes with time, and the 3-month time frame of our study does not reflect more long-term changes. For many anomalies, HRQL has been shown to be worst for the youngest children (<4 years) and improves over time.[22,23] Nonetheless, our short-term time frame does provide some perspective on the well-being of these families as they transition to home and should inform communication with the primary care providers that take over their care following NICU discharge.

The type of anomaly and complexity of care (number of consultants, length of stay, palliative care involvement) were influential in determining parent HRQL, and they likely are interrelated factors. For example, infants with genetic anomalies had longer length of stay, had more consultants, and were more likely to carry a worse prognosis. Whether the anomaly itself or its associated medical complexity contributes to lower parent HRQL remains to be explored. We speculate that parents of infants with certain surgical anomalies may derive hope in viewing their child's problems as temporary and able to be fixed, whereas those with nonsurgical anomalies may perceive that problems are more permanent and not fixable. Moreover, infants with surgical anomalies tended to have fewer consultants involved, as opposed to a patient with a more complex genetic diagnosis with whom multiple specialists may be involved. Prior work on parent experience in the NICU for infants born with anomalies has shown that parents often feel overwhelmed and desire more empathic listening and better communication.[24] Parents with a history of mental health concerns and parents of premature infants with anomalies may be especially vulnerable to poor quality of life. Addressing the needs of the baby and the family as a whole rather than relegating individual organ systems to multiple specialists may improve parent quality of life.

Overall, our study cohort had significant home health care needs and post-NICU healthcare utilization. One-quarter of the infants were discharged with home oxygen, and 40% were discharged home with a gastrostomy tube. More than 40% of infants were readmitted to the hospital within the study period. Parents of children with a gastrostomy tubes or with hospital readmission had lower 3-month HRQL.

Parent quality of life is multifactorial and likely driven by factors beyond what we report in our study. Some families may adapt readily to the need for home medical equipment, while others may struggle. For some children, these needs may be temporary, and for others, they may be lifelong.[20] Regardless, fostering early independence with care, better education for families being discharged with home medical equipment, and setting realistic expectations in terms of need for future healthcare utilization might better prepare families to

cope with their child's health care needs. In addition, a better understanding of what outcomes are important to families of infants with anomalies is vital for effective communication.

We acknowledge certain limitations to our study. As a single-center study, certain anomalies were underrepresented. Parents of infants who were discharged to home before 14 days of their NICU stay were excluded. Although the majority of anomalies represented in our cohort would necessitate a longer NICU stay, it is possible that infants with more favorable prognoses were excluded. We did not have a large enough sample to fully evaluate the effect of disease severity on HRQL within each anomaly category. Although we represent quantitative differences in quality of life, we did not explore the reasons why some parents reported worse quality of life. We measured post-NICU HRQL at 3 months but do not know the long-term quality of life of these families. Despite these limitations, this is one of the first studies to explore parent HRQL for infants with anomalies during the NICU and post-NICU period and to examine factors associated with quality of life.

In conclusion, parents of infants with anomalies admitted to NICUs represent a population at risk for lower HRQL in the NICU and after discharge to home. The complexity of NICU care and post-NICU healthcare needs likely impose burdens on families that affect their HRQL. These parents could be targeted earlier in their NICU course by the NICU team with education, hands-on training, and anticipatory guidance as to what to expect after discharge home. Future research should focus on qualitative evaluation of parental HRQL in this population, as well as large, multi-institution analyses of data for specific congenital anomalies.

## Data Statement

Data sharing statement available at www.jpeds.com.

## Appendix

Table I. Anomaly classification schemes and examples of included anomalies

| **Anomaly classification schemes** | |
|---|---|
| **Classification by prognosis∗ (≠)** | **Examples of anomalies included** |
| Moderate: significant NICU course with either need for surgery in NICU or mechanical ventilation or LOS >14 days, but with overall good prognosis and good expected long-term HRQL based on previously reported data | Gastroschisis, CDH, CPAM, TEF, bowel atresias, bladder outlet obstruction, ventricular septal defect<br>Genetic conditions such as Pierre-Robin sequence, Turner syndrome |
| Severe: expected long-term healthcare burdens that are typically life-long; possibility of need for repeat surgeries or procedures | MMC, cloacal exstrophy, hydrocephalus, single ventricle physiology, skeletal dysplasia<br>Genetic conditions such as Trisomy 21, CHARGE syndrome |
| Life-limiting: anticipated neonatal or infant death; few long-term survivors | Trisomy 13 or 18, neonatal Marfan syndrome |
| Classification by need for surgery | |
| Neonatal surgical: will require surgery during the neonatal admission | Bowel atresias, congenital diaphragmatic hernia, tracheoesophageal fistula, gastroschisis, hypoplastic left ventricle |
| Non-neonatal surgical: will require surgery at some point, but usually not during the neonatal admission | VSD, AV canal, cleft lip or cleft palate, club feet, CPAM, limb anomalies |
| Nonsurgical: do not/will not typically require surgery at any point or cannot be surgically corrected; infants in this group may have required surgery for reasons other than the | Agenesis of corpus callosum, congenital myotonic dystrophy, and multiple other genetic conditions |

| | |
|---|---|
| anomaly, such as a gastrostomy tube for infants with severe developmental delays | |
| Classification by organ system/type of anomaly | |
| Genetic | Diagnosis associated with a genetic mutation or change, eg, trisomy 13, single gene disorders |
| Associated with a syndrome | Subgroup of Genetic; constellation of anomalies associated with a known syndrome, such as CHARGE |
| Thoracic | TEF, CDH, CPAM, pulmonary lymphatic malformations |
| Cardiac | AV canal, large VSD, double-outlet right ventricle, Ebstein anomaly, hypoplastic left ventricle; minor anomalies, such as ASD, or PDA not included |
| Central nervous system | Myelomeningocele, aqueductal stenosis, absent septum pellucidum, agenesis of corpus callosum, ventriculomegaly, Chiari malformation, colpocephaly, Dandy–Walker malformation/spectrum, encephalocele, holoprosencephaly, hydrocephalus, polymicrogyria |
| Craniofacial | Cleft lip/palate, eye, ear, or airway anomalies |
| Airway | Choanal atresia or stenosis, choanal stenosis, Pierre-Robin sequence, mandibular hypoplasia, microretrognathia |
| Eye | Coloboma, cataract, retinitis pigmentosa, septo-optic dysplasia |
| Ear | Any ear, nose, throat malformations |
| Abdominal wall defect | Gastroschisis, omphalocele, cloacal exstrophy |
| Gastrointestinal | Bowel atresia (including duodenal, jejunal, ileal or colonic), Hirschsprung disease, imperforate anus |
| Renal/genitourinary | Any kidney malformations or those affecting genitourinary system, such as cystic kidney disease, horseshoe kidney, polycystic kidney disease; or absent or micropenis, ambiguous genitalia, bladder outlet obstruction with or without bilateral hydronephrosis, hydronephrosis, posterior urethral valves |
| Musculoskeletal | Congenital myotonic dystrophy, limb anomalies, osteogenesis imperfecta, skeletal dysplasia, vertebral anomaly, clubfoot |
| Multiple anomaly complex | Multiple anomalies present, either with or without an associated genetic or syndromic diagnosis; this term encompasses genetic anomalies associated with a syndrome as well as multiple anomalies not associated with any known syndrome or genetic diagnosis. |

*ASD*, atrial septal defect; *AV* canal, atrioventricular canal defect; *CHARGE,* coloboma, heart defects, atresia choanae (also known as choanal atresia), growth retardation, genital abnormalities, and ear abnormalities; *CPAM*, congenital pulmonary airway malformation; *LOS*, length of stay; *MMC,* myelomeningocele; *PDA*, patent ductus arteriosus; *VSD*, ventricular septal defect. When 2 or more anomalies were present, the more severe of the 2 anomalies was recorded (eg, trisomy 21 with intestinal atresia would be classified as severe). When 2 or more anomalies were present, at least 1 of which was neonatal surgical, then the patient was classified as having a neonatal surgical anomaly. Classification adapted from Baker A, Lagatta J, Leuthner S, Acharya K. Does prenatal counseling for pregnancies complicated by multiple fetal abnormalities concord with postnatal outcomes? Prenat Diagn 2020;40:538-48.[25] ∗ Anomalies with a wide-ranging prognosis are included in the most anticipated prognostic category.

Table IV. Multivariable regression model showing predictors of baseline and 3-month parent HRQL

| **Variables** | **Coefficient** | **95% CI** | ***P* value** |
|---|---|---|---|
| Baseline HRQL | | | |
| Parent age | −1.25 | −0.65 to 0.4 | .64 |
| Race | | | |
| Black | Reference | | |
| White | −3.33 | −11.16 to 4.5 | .401 |
| Hispanic | 3.2 | −12.4 to 18.8 | .686 |
| Asian | 10.6 | −7.06 to 28.3 | .236 |
| Other | 6.66 | −5.76 to 19.09 | .29 |
| Has a car | 1.24 | −7.59 to 10.08 | .781 |
| History of mental health disorder | −9.2 | −15.1 to −3.2 | **.003** |
| Anomaly classification by surgery | | | |
| Neonatal surgical | Reference | | |
| Non-neonatal surgical | −2.15 | −10.1 to 5.8 | .593 |
| Nonsurgical | −0.9 | −7.5 to 5.6 | .779 |
| Gestational age, wk | | | |
| ≤28 | Reference | | |
| 29-36 | 2.7 | −12.3 to 17.7 | .723 |
| ≥37 | 7.4 | −8.6 to 23.5 | .362 |
| Multiple consultants | −10.3 | −18.4 to −2.2 | **.013** |
| Palliative care involved | −8.3 | −18.7 to −2.2 | .115 |
| Psychology consult in the NICU | | | |
| Mechanical ventilation in the NICU | −2.9 | −9.2 to 3.5 | .377 |
| LOS | 0.01 | −0.08 to 0.11 | .825 |
| 3-month HRQL | | | |
| Enrollment HRQL (each 1 point) | 0.83 | 0.58 to 1.09 | **<.001** |
| Gestational age, wk | | | |
| ≤28 | Reference | | |
| 29-36 | 10.8 | −7.9 to 9.8 | .25 |
| ≥37 | 8.1 | −10.7 to 26.9 | .39 |
| Anomaly classification by surgery∗ (1) | | | |
| Neonatal surgical | Reference | | |
| Non-neonatal surgical | −2 | −13.8 to 9.7 | .73 |
| Nonsurgical | −0.1 | −10.9 to 10.5 | .97 |
| Discharged with gastrostomy tube | 1.2 | −7 to 9.6 | .76 |
| Hospital readmission | −7.9 | −15.6 to −0.2 | **.04** |
| Discharged with home oxygen | −2.1 | −11.8 to 7.6 | .67 |

*P* values <0.05 are in bold font.

# References


1. J. Weiner, J. Sharma, J. Lantos, H. Kilbride. **How infants die in the neonatal intensive care unit: trends from 1999 through 2008.** Arch Pediatr Adolesc Med, 165 (2011), pp. 630-634
2. C.E. Ratliffe, R.C. Harrigan, J. Haley, A. Tse, T. Olson. **Stress in families with medically fragile children.** Issues Compr Pediatr Nurs, 25 (2002), pp. 167-188

3. K. Kuhlthau, K.S. Hill, R. Yucel, J.M. Perrin. **Financial burden for families of children with special health care needs.** Matern Child Health J, 9 (2005), pp. 207-218
4. N. Golfenshtein, A.L. Hanlon, J.A. Deatrick, B. Medoff-Cooper. **Parenting stress in parents of infants with congenital heart disease and parents of healthy infants: the first year of life.** Compr Child Adolesc Nurs, 40 (2017), pp. 294-314
5. S. McAndrew, K. Acharya, J. Westerdahl, D.C. Brousseau, J.A. Panepinto, P. Simpson, *et al.* **A prospective study of parent health-related quality of life before and after discharge from the Neonatal Intensive Care Unit.** J Pediatr, 213 (2019), pp. 38-45.e3
6. G.H. Guyatt, D.H. Feeny, D.L. Patrick. **Measuring health-related quality of life.** Ann Intern Med, 118 (1993), pp. 622-629
7. R.J. Graham, A.M. Rodday, R.A. Weidner, S.K. Parsons. **The impact on family of pediatric chronic respiratory failure in the home.** J Pediatr, 175 (2016), pp. 40-46
8. J.M. Lagatta, M. Uhing, K. Acharya, J. Lavoie, E. Rholl, K. Malin, *et al.* **Actual and potential impact of a home nasogastric tube feeding program for infants whose neonatal intensive care unit discharge is affected by delayed oral feedings.** J Pediatr, 234 (2021), pp. 38-45.e2
9. J.W. Varni, S.A. Sherman, T.M. Burwinkle, P.E. Dickinson, P. Dixon. **The PedsQL™ Family Impact Module: preliminary reliability and validity.** Health Qual Life Outcomes, 2 (2004), p. 55
10. Centers for Disease Control and Prevention. **Birth Defects Surveillance Toolkit. 5.1 International Classification of Diseases. 2018.** https://www.cdc.gov/ncbddd/birthdefects/surveillancemanual/facilitators-guide/module-5/mod5-1.html, Accessed 27th Oct 2021
11. D.W. Bianchi. **Fetology: diagnosis and management of the fetal patient.** (Second edition), McGraw-Hill, New York (NY) (2010).
12. Centers for Disease Control and Prevention. **What are birth defects? 2020.** https://www.cdc.gov/ncbddd/birthdefects/facts.html, Accessed 27th Oct 2021
13. K. Acharya, S. Leuthner, R. Clark, T.H. Nghiem-Rao, A. Spitzer, J. Lagatta. **Major anomalies and birth-weight influence NICU interventions and mortality in infants with trisomy 13 or 18.** J Perinatol, 37 (2017), pp. 420-426
14. S.R. Leuthner. **Palliative care of the infant with lethal anomalies.** Pediatr Clin North Am, 51 (2004), pp. 747-759
15. **Research Electronic Data Capture (REDCap).** https://projectredcap.org/about/, Accessed 9th Jun 2021
16. R. Amin, M. Knezevich, M. Lingongo, A. Szabo, Z. Yin, K.T. Oldham, *et al.*. **Long-term quality of life in neonatal surgical disease.** Ann Surg, 268 (2018), pp. 497-505
17. J.W. Varni, T.M. Burwinkle, M. Seid, D. Skarr. **The PedsQL™ 4.0 as a pediatric population health measure: feasibility, reliability, and validity.** Ambul Pediatr, 3 (2003), pp. 329-341
18. J.W. Varni, C.A. Limbers, K. Neighbors, K. Schulz, J.E.C. Lieu, R.W. Heffer, *et al.* **The PedsQL™ Infant Scales: feasibility, internal consistency reliability, and validity in healthy and ill infants.** Qual Life Res, 20 (2011), pp. 45-55
19. S.V. Glinianaia, N.D. Embleton, J. Rankin. **A systematic review of studies of quality of life in children and adults with selected congenital anomalies.** Birth Defects Res A Clin Mol Teratol, 94 (2012), pp. 511-520
20. D. McCann, R. Bull, T. Winzenberg. **The daily patterns of time use for parents of children with complex needs: a systematic review.** J Child Health Care, 16 (2012), pp. 26-52
21. K. Javalkar, E. Rak, A. Phillips, C. Haberman, M. Ferris, M. Van Tilburg. **Predictors of caregiver burden among mothers of children with chronic conditions.** Children (Basel), 4 (2017), p. 39
22. J.A. Deurloo, S. Ekkelkamp, E.E. Hartman, M.A.G. Sprangers, D.C. Aronson. **Quality of life in adult survivors of correction of esophageal atresia.** Arch Surg, 140 (2005), pp. 976-980

23. M.J. Poley, E.A. Stolk, D. Tibboel, J.C. Molenaar, J.J.V. Busschbach. **Short term and long term health related quality of life after congenital anorectal malformations and congenital diaphragmatic hernia.** Arch Dis Child, 89 (2004), pp. 836-841
24. B. Áskelsdóttir, S. Conroy, G. Rempel. **From diagnosis to birth: parents' experience when expecting a child with congenital anomaly.** Adv Neonatal Care, 8 (2008), pp. 348-354
25. A. Baker, J. Lagatta, S. Leuthner, K. Acharya. **Does prenatal counseling for pregnancies complicated by multiple fetal abnormalities concord with postnatal outcomes?** Prenat Diagn, 40 (2020), pp. 538-548