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# The family burden of paediatric heart disease during the chronic phase of illness

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## Original Article

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## Abstract

*Background:* CHD is a lifelong condition with a significant burden of disease to patients and families. With increased survival, attention has shifted to longer-term outcomes, with a focus on social determinants of health. Among children with CHD, socioeconomic status is associated with disparities in outcomes. Household material hardship is a concrete measure of poverty and may serve as an intervenable measure of socioeconomic status. *Methods:* A longitudinal survey study was conducted at multiple time points (at acute hospitalisation, then 12–24 months later in the chronic phase) to determine the prevalence of household material hardship among parents of children with advanced heart disease and quality of life during long-term follow-up. *Results:* The analytic cohort was 160 children with a median patient age of 1 year (IQR 1,4) with 54% of patients <2 years. During acute hospitalisation, over one-third of families reported household material hardship (37%), with significantly lower household material hardship in the chronic phase at 16% (N = 9 of 52). For parents reporting household material hardship during acute hospitalisation, 50% had resolution of household material hardship by the chronic phase. Household material hardship-exposed children were significantly more likely to be publicly insured (56% versus 20%, p = 0.03) with lower quality of life than those without household material hardship (64% versus 82%, p = 0.013). *Conclusion:* The burden of heart disease during the chronic phase of illness is high. Household material hardship may serve as a target to ensure equity in the care and outcomes of CHD patients and their families.

CHD is a lifelong, high-resource utilisation condition with peaks of substantial acuity in the setting of a chronic disease. While only representing a small percentage of overall patients in children's hospitals, CHD is responsible for a significant burden of care, as is common with acute on chronic conditions.[1–4] This burden is experienced by the hospital and health system, in the form of long length of stays, unplanned readmissions, and significant time burden on a large number of sub-specialised providers.[2,3,5–7] In addition, however, are the burden CHD places on families in relation to the value of care their child receives.[8,9]

Over the past decade, mortality from CHD continues to decline, and research has expanded to encompass specific burdens to patients and families. In particular, significant work has been undertaken to assess the effectiveness of transition to adult CHD management, neuropsychiatric and learning abilities, parental stress in the immediate post-operative period, and children's emotional burdens.[10–12] However, most efforts target one point in time during acute illness, rather than the effect of CHD during the stable chronic phase of illness where patients and families spend most of their time. With the exception of assessing neurodevelopmental outcomes in children with CHD, which is now at the forefront of the field, there is a current lack of understanding and research into the stable chronic phase of CHD in terms of overall burden to families.[13–15]

At a time of significant dynamic change in healthcare delivery, there is a need for greater understanding of not only the more longitudinal implications of CHD in terms of symptoms and medical needs but also the burden that families experience over time. It is demonstrated that as mortality declines across paediatric critical disease, morbidity increases yielding more longitudinal medical needs and a higher care burden for families. While McClung, et al. independently assessed financial burden from the National Survey of Children with Special Health Needs database two decades ago, there has not been an assessment of burden of care from individual families within a large Heart Center.[16] Piloting this study to assess the burden of disease to families of children with CHD during the chronic stable phase will offer the opportunity to learn how to better serve patients with CHD with acute and chronic disease and their families.

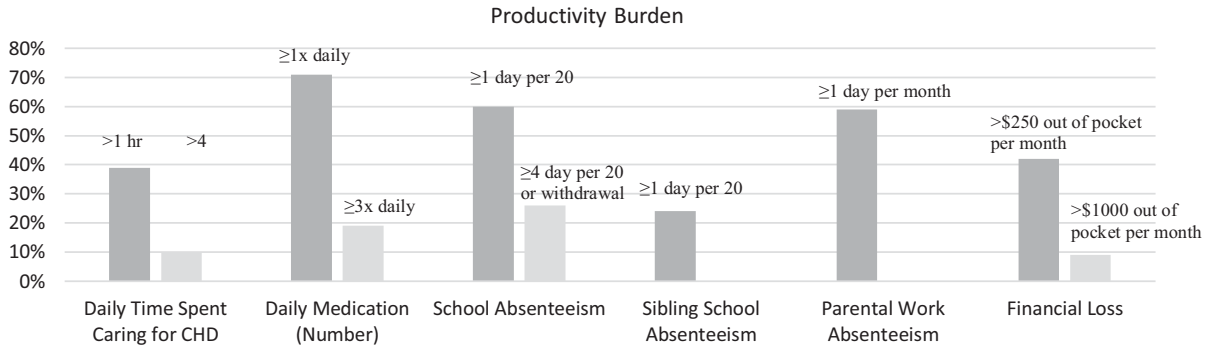


**Figure 1.** Productivity burden percentages of total respondents by type of burden.

## Materials and methods

A prospective longitudinal study was conducted with a cohort of families of children with advanced heart disease. Families were queried for patient-reported outcomes and clinical data at time of acute illness and 12–24 months later to assess (1) overall burden of disease on families and (2) household medical hardship (a concrete measure of poverty including food or housing insecurity) at times of acute illness compared to the time of chronic disease. The study was conducted within a large, quaternary children's hospital heart centre.

The initial cohort of patients and families was assessed in the acute hospital phase of illness as a part of a series of prior projects, which assessed parental and provider prognostic awareness, and household medical hardship.[17] Acute hospital phase of illness was defined as hospitalised patients with advanced heart disease, which included patients < 19 years old with CHD or acquired heart disease and either length of stay > 30 days, mechanical ventilation > 14 days, > 2 admissions for heart disease-related issues in the past year, or actively listed for heart transplant. Stable chronic phase of illness included the same cohort of patients, but during the 12–24 months following initial inclusion in the cohort. It excluded patients who were readmitted at time of follow-up or those who had died following the initial, acute phase of illness. Patients were identified during clinic visits (either in-person or virtual) in the 12–24 month follow-up after initial survey, as all patients with the above criteria for advanced heart disease were followed longitudinally at a minimum of once annually within the quaternary centre, even if followed in parallel at local or regional centres.

Burden of disease was determined via a series of validated questions assessing time loss of school/work, financial losses, overall assessment of self-health, and patient symptom burden (utilising the EQ-5D single question assessment).[18] Predictors of overall burden of disease to families of children with advanced heart disease during stable chronic phase of illness included total number of days hospitalised over the past year, number of readmissions over the past year, number of days since the last surgery, and major complications (as defined by the Society of Thoracic Surgery) during the hospitalisation while enrolled in the study during the acute hospital phase of illness.[19]

To assess household medical hardship, a previously validated tool was utilised, with questions surrounding food insecurity and housing insecurity, through the Hunger and Housing Stability Vital Sign Tools from Children's Health Watch.[20] These tools have previously demonstrated high validity and reliability in psychometric analysis.[20–22] Additionally, a question assessing overcrowding was used (specific questions listed in Fig. 1). Children were classified as household medical hardship-exposed if their family endorsed any single question from the Hunger or Housing Stability Vital Sign Tools or overcrowded housing.[20] The same questions were asked on the follow-up survey

### Data management and chart review

All survey responses were recorded in REDCap (Research Electronic Data Capture database;).[23] Acute data were collected from 2018 to 2019 and chronic data from 2020 to 2021. Research investigators verified the data to ensure accuracy and completeness. Each participant was assigned a unique study number. Some demographic, disease, and healthcare utilisation data were abstracted from the medical record by a trained research assistant and recorded in REDCap. Auto-queries and auto-validations were incorporated into REDCap during the design phase to minimise data entry error.

### Statistical analysis

Descriptive statistics include mean ± standard deviation and median with interquartile range for continuous variables. Categorical data were described as number with frequency. Differences in characteristics between children with vs. without household medical hardship were evaluated with Fisher's exact test, Student's t-test, or Wilcoxon rank sum test as appropriate. A p-value of less than 0.05 was considered statistically significant. Analyses were performed using SAS 9.4.[24]

## Results

In the acute phase of illness, 160 parents or guardians were enrolled. Of the 160 surveys completed, 94% provided data for an analytic cohort of 150 children. Of patients enrolled, 101 (67%) were on the general cardiology wards and 105 (70%) had received the diagnosis of advance heart disease antenatally. Of the acute cohort, 76 (52.5%) of patients were female and in the majority of the families (71.9%) the respondent identified as white (Table 1).

The median age of patients at acute parent enrolment was 1 year (median 1, IQR 1, 5) with the majority of patients (54%) < 2 years of age. Almost all patients (n = 149, 93.3%) had CHD. Half of families (53%) knew about the diagnosis for over one year. Among respondents completing the survey, the median age was 34 years old (range 17 to 56); 130 (87%) were mothers and 114 (76%) were married or living with a partner; the majority were privately insured (N = 109, 68%). Additionally, 97 (64%) had a college education or higher. Patients eligible for follow-up in the chronic

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**Table 1.** Patient demographics.

| Characteristics | Overall cohort (n = 160) | Chronic phase (n = 58) |
|---|---|---|
| Age, years | | |
| Median (IQR) | 1 (1,4) | 2.00 (1,4) |
| Male | 84 (53%) | 30 (56%) |
| Parental race/ethnicity | | |
| Hispanic | 29 (19%) | 3 (6%) |
| White | 115 (72%) | 43 (81%) |
| Black | 12 (8%) | 0 (0%) |
| Asian | 7 (4%) | 3 (6%) |
| Other | 23 (14%) | 4 (8%) |
| Aetiology—cardiomyopathy | | |
| Yes | 9 (6%) | 3 (6%) |
| No | 151 (94%) | 50 (94%) |
| Aetiology—CHD | | |
| Yes | 149 (93%) | 50 (94%) |
| No | 11 (7%) | 3 (6%) |
| Aetiology—heart transplant | | |
| Yes | 9 (6%) | 4 (7%) |
| No | 151 (94%) | 49 (93%) |

cohort included 58 respondents of 160 initially enrolled patients (58/98, capture rate of 59%), with 34 excluded for either not reaching a phase of chronicity (being readmitted to the hospital during the preceding 12 months), loss-to-follow-up, and 28 inter-study mortalities. Compared with the acute cohort, the chronic cohort patient age was older (median 2 years versus 1 year), there were relatively more males (56% vs. 48%), and there was consistency in the predominance of white race (81%), though with lower rates of Black and other race/ethnicity than the acute cohort.[25] The majority of patients in the chronic cohort were privately insured (N = 39, 74%).

Indicators of family burden during the chronic stage of illness included functional status and perceived health status, productivity (time and finances) burdens, and household medical hardship:

### Functional status

When asked about limitations in functional status, including symptoms and activity burden, functional status was a limitation in 60% of patients having some limitation (35/58), with 26% having at least marked limitation (15/58). On the EQ-5D scale of 1–100, with 100 being the highest answer, the average score for perceived health status of the child by the parent was 78%, below the median from other chronic conditions.[21,26]

### Productivity burden (Fig 1)

Over 1/3 (26/56, 39%) of families spend at least one hour per day caring for their child with CHD, with 10% (6/56) spending greater than four hours per day. All patients required daily medication, with 71% (41/58) taking multiple medications daily and 19% (11/58) requiring at least three times daily medications. Child school absenteeism included 60% missing some school on a monthly basis (21/35), with 26% (9/35) either missing at least 4 of 20 days per month or withdrawing from in-person school primarily due to cardiac condition. Siblings of children with CHD missed at least one day of school per month in 24% of families (9/37). The majority of parents of children with CHD missed at least a day of work in a given month (59%, 33/56). Financially, 42% (24/57) of families spend >$250 out-of-pocket (defined as any costs families paid for care from their perspective) in some form to care of their child per month and 9% (N = 1) spend >$1000 per month.

### Household medical hardship

At the time of baseline survey, which was administration to acutely hospitalised families, 59/150 families were household medical hardship-exposed (39%) over the preceding 6 months. Of these, 41 (26%) endorsed food insecurity, 49 (31%) endorsed housing insecurity, with 31 (19%) endorsing both food and housing insecurity in the 6 months prior to the acute illness. In the follow-up assessment, following a period of>12 months out of the hospital, household medical hardship was significantly lower, with 16% of families reporting household medical hardship (9 of 58 reporting families). Most notably, for the parents who reported household medical hardship in the acute hospital phase who completed the follow-up survey (N = 20), 50% of these families had resolution of household medical hardship at the time assessed in chronic illness phase. There were no findings of patients that initially did not express household medical hardship exposure, then developed household medical hardship exposure in the chronic phase of illness.

Households with household medical hardship were more likely to be headed by a single-parent (45% vs. 12%, $p < 0.001$), who was non-college-educated (50% vs. 75%, $p = 0.016$) and of self-reported Hispanic ethnicity (27% versus 10%, $p = 0.001$). These households also had lower household income (median $25,000, IQR (12.5, 50.0) versus $120,000, IQR (70.0, 160.0), $p < 0.001$) (Table 2).

In univariate analyses, during the chronic phase of illness, compared to those without household medical hardship, those exposed to household medical hardship were more likely to be publicly insured than privately insured (56% vs. 20%, $p = 0.03$). QOL of respondents (EQ15) was queried only during the chronic phase of illness. Families who experienced household medical hardship while in the hospital during the acute phase of illness had a lower reported QOL by the parent on the EQ15 during the chronic phase, as compared to those in the acute phase who did not experience household medical hardship (64 ± 32% EQ15 score for respondents with household medical hardship during the acute phase of illness, versus 82 ± 17% for those who did not experience acute household medical hardship; $p = 0.013$; represents 36% of the acute cohort who completed the follow-up survey) (Fig. 2).

Within three potential categories for free-text comments (related to symptom burden and financial burdens), 50% (29/58) provided responses during the chronic phase of illness. Of the symptom responses, 71% (12/17) were related to symptoms adding burden to daily life, while 29% (5/17) commented on a lack of symptoms or ability to achieve activity goals. Of these comments, 35% (6/17) related to burdens or stressors that the symptoms have placed on the family as opposed to the child alone. Of the financial comments, 44% (7/16) addressed either job loss or limited opportunity to work and 56% (9/16) related to costs of travel for care or housing during inpatient hospitalisations.

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**Table 2.** Patient and parent demographics by household material hardship categorisation in the chronic phase of illness.

| Characteristics | Overall (n = 58) | HMH = Yes (n = 9) | HMH = No (n = 49) | p-value |
|---|---|---|---|---|
| Age, years | 3.09 ± 3.67 | 1.67 ± 1.22 | 3.39 ± 3.94 | **0.022** |
| Median (IQR) | 2.00 (1.00, 4.00) | 1.00 (1.00, 2.00) | 2.00 (0.50, 5.00) | 0.485 |
| Male | 30 (57%) | 2 (22%) | 28 (64%) | **0.022** |
| Race/ethnicity | | | | 0.556 |
| Hispanic | 3 (6%) | 1 (11%) | 2 (4%) | |
| White | 43 (81%) | 8 (89%) | 35 (80%) | |
| Black | 0 (0%) | 0 (0%) | 0 (0%) | |
| Asian | 3 (6%) | 0 (0%) | 3 (7%) | |
| Other | 4 (8%) | 0 (0%) | 4 (9%) | |
| Insurance type | | | | **0.030** |
| Private | 39 (74%) | 4 (44%) | 35 (80%) | |
| Public | 14 (26%) | 5 (56%) | 9 (21%) | |
| Aetiology—cardiomyopathy | | | | 0.420 |
| Yes | 3 (6%) | 0 (0%) | 3 (7%) | |
| No | 50 (94%) | 9 (100%) | 41 (93%) | |
| Aetiology—CHD | | | | 0.420 |
| Yes | 50 (94%) | 9 (100%) | 41 (93%) | |
| No | 3 (6%) | 0 (0%) | 3 (7%) | |
| Aetiology—heart transplant | | | | 0.347 |
| Yes | 4 (8%) | 0 (0%) | 4 (9%) | |
| No | 49 (93%) | 9 (100%) | 40 (91%) | |
| Genetic syndrome | | | | 0.907 |
| Yes | 11 (23%) | 2 (25%) | 9 (23%) | |
| No | 36 (77%) | 6 (75%) | 30 (77%) | |
| Number of heart surgeries | 53 | 9 | 44 | |
| Median (IQR) | 2.0 (1.0, 3.0) | 3.0 (2.0, 3.0) | 2.0 (1.0, 3.0) | 0.369 |
| Number of catheterisations | 53 | 9 | 44 | |
| Median (IQR) | 3.0 (1.0, 5.0) | 3.0 (1.0, 3.0) | 2.5 (1.0, 5.0) | 1.000 |
| Number of days hospitalised over past 12 months | 9 | 2 | 7 | |
| Median (IQR) | 22.0 (17.0, 48.0) | 13.0 (9.0, 17.0) | 28.0 (19.0, 59.0) | 0.142 |
| Number of admissions over past 12 months | 9 | 2 | 7 | |
| Median (IQR) | 1.0 (0.0, 3.0) | 2.5 (2.0, 3.0) | 1.0 (0.0, 3.0) | 0.368 |
| Number of ED presentations over past 12 months | 9 | 2 | 7 | |
| Median (IQR) | 0.0 (0.0, 0.0) | 2.0 (0.0, 4.0) | 0.0 (0.0, 0.0) | 0.316 |

## Discussion

Across families with children with advanced heart disease, during times of stability while not in the hospital, there is significant residual symptom burden, productivity burden in terms of work and school absenteeism, and costs. This pilot is the first report on burdens of paediatric heart disease in the chronic phase of illness. While this cohort represents a higher severity of illness than the congenital heart disease population as a whole, the study fills a void in the understanding of burdens families and patients are experiencing at a given point in time while in the chronic phase of illness. Patients with complex CHD quality of life which makes up the majority of the cohort—will require recurrent hospitalisations and procedures in the first several years of life.[27] Less attention is given to their and their families' ability to live productive, low-symptom lives during the majority of their time, which is not spent in the hospital. In addition to what was learned during the acute illness, this study offered multiple opportunities for families to provide free-text comments to serve as feedback and ideas of the struggles families and patients face that care teams may not consider.

### Functional status

The functional limitation and symptom impact on quality of life has had increased attention in the CHD population over the last several years. Neurodevelopmental programmes have begun

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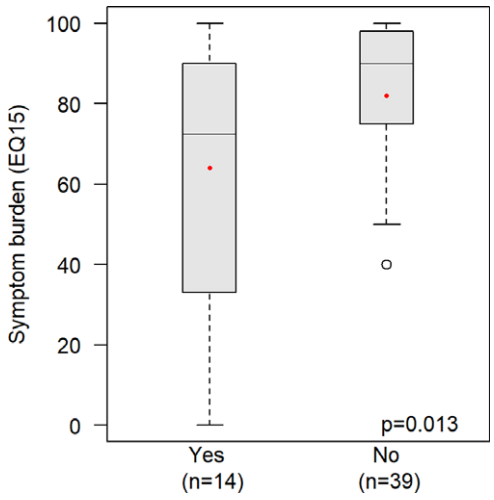


**Figure 2.** Quality of life during the chronic phase of illness. HMH = household medical hardship; IQR = interquartile range; QOL = quality of life.

implementing quality of life assessments and with the exponential growth of adults with CHD, programmes assessing adults with CHD wellness and transition programmes have developed.[12,26–28] However, during the first few years of life, it is not known how symptomatic or functional patients are while not in the hospital. This study found nearly 2/3 of patients had continued limited functional status with>1/4 indicating marked limitation. Recent work by Han, et al utilised a functional status Scale before and after CHD surgery, finding only 5% of patients had new morbidity at time of discharge.[29] Our results suggest that whether the morbidity associated with limited functional status is new or not (which was not assessed without having pre-hospitalisation baseline data), there is a meaningful percentage of patients that could be targeted with services to improve their functional status. More broadly than functional status, patients and families self-reported health status perception was notably lower than reported in other paediatric chronic conditions such as asthma, rheumatologic disease, or diabetes.[16,20] This could be attributable to the selection bias of a more severe cohort of patients with heart disease, but nonetheless identifies a gap in optimal care and can serve as a starting point for improving care for a highly complex, severe population cared for within paediatric heart centres.

### Productivity and costs

School absenteeism is recognised within paediatric public health as a primary indicator of poor health and predictive of future poor health.[28] As outcomes for CHD become increasingly longitudinal, similar to other chronic conditions, indicators such as school absenteeism serve as an opportunity to target to improve overall health. Until this time, school absenteeism was not well-known within CHD patients. A prior single-centre study from Europe found the majority of patients had school or work absenteeism, with 15% of this being attributable to CHD, though the median days missed was only 3 days per year.[29] This cohort was for all CHD, as compared to the current study which focuses on the most severe patients and finds that they miss significantly more school. At>4 days per month of missed school (1/4 of patients in the cohort), patients qualify as chronic absenteeism, which is included in recommendations for repeating school years or having additional educational resources to prevent remediation (https://www2.ed.gov/datastory/chronicabsenteeism.html).

Similarly, parental work absenteeism is anecdotally a known problem within the CHD population, given the often long and recurrent hospitalisations in regionalised facilities. However, there are limited data as to how much time or potential income is lost to families due to their child's CHD. A recent study into CHD parental stressors identified a mean of 37 days in the hospital over the first year of a patient's life with complex CHD.[30] Over half of working parents in this study missed work time in the preceding month, which importantly was during a phase of stability, and does not represent the time of likely highest work time missed. Coupled with the findings that over half of families spend >$250 per month (extrapolated to >$3000 out of pocket costs per year), these findings suggest that to truly optimise end-user outcomes—those of the family—opportunities exist that are not currently being addressed.

### Household material hardship

Living in poverty is associated with poor health outcomes for children, particularly those with chronic illness or medical complexity.[33–38] With 21% of children in America living below the federal poverty limit and 43% living below 200% of the federal poverty limit, this is a particularly prevalent threat to child health and wellbeing.[33,39–41] Among children with CHD, socioeconomic status is associated with disparities in mortality and health resource utilisation.[42,43] Furthermore, children with CHD are living longer, with more co-morbidities, higher medical resource utilisation, and increasing costs.[44,45] These trends pose a dual burden, both for the healthcare system and for patient families. While numerous studies have demonstrated SES-associated outcome disparities among children with CHD, targetable socioeconomic status interventions have not been identified.[45] The household material hardship tool utilised in this project has led to improvements in accessing social support in the oncology population and may serve as an intervenable measure within the CHD population.[46,47]

### Limitations

The external application and broader replicability of this project have notable limitations. The patient population represented those on the severe end of the severity spectrum of CHD. While an important population to focus on given the disproportionate resource utilisation, the results may not be generalisable to the higher number of patients with milder disease. Additionally, while the race/ethnicity of both cohorts were primarily White, the acute cohort does have a higher rate of those indicating Black or Other. This was not statistically significant, but may have been negatively impacted by the follow-up survey being conducted during the COVID-19 era, a time in which access to medical teams disproportionally affected underrepresented minorities. The COVID-19 pandemic could have impacted the data of the chronic phase (2020–2021), particularly in regard to school and work absenteeism, though that was not commented in free-text open-ended responses from families pertaining to absenteeism. While the institution cares for international referrals frequently, the longitudinal nature of the study biased the inclusion to include only patients from the US. As a survey-based study, there is potential for response bias and different characteristics within those who did not respond. Finally, as an observational qualitative study, statistical analyses were not as robust as if there were greater number of patients.

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## Conclusion

While this project represents an initial look into family-reported data of patients with CHD, the ability to systematically and operationally measure outcomes of importance to families is crucial to generating high-value service delivery. Organisations such as the International Consortium of Health Outcomes Measures have begun to develop sets of core outcomes across integrated conditions such as CHD, with the goal of driving value-based healthcare in a patient-centric approach.[9] The CHD set of outcomes, which has significant overlap with the questions asked in this project's survey, offers the opportunity for implementation and generation of similar datasets across centres for benchmarking and improvement.[48] In addition to measuring the outcomes, these data suggest the wide net of stakeholders within an integrated care team required to optimally deliver care to patients and families with CHD.

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**Competing interests.** None.

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