# The Impact on Family of Pediatric Chronic Respiratory Failure in the Home

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**Objective** To assess the family impact of managing severe, chronic respiratory failure (CRF) at home. Better understanding will inform parental counseling and serve as a point of reference for interventions.

**Study design** Families of children with CRF completed the Impact on Family Scale (IFS) and Consumer Assessment of Healthcare Providers and Systems. Using multivariable linear regression, we assessed the relationship between IFS and family, clinical, and utilization characteristics.

**Results** A total of 118 parents (60%) completed the IFS; 114 parents (58%) completed all measures. The 15-item IFS mean total score was 40 (SD = 10) with a possible range of 15-60 (greater scores indicate more impact). Modeling identified a negative association with parent emotional functioning, parent-rated child health, and private insurance only (compared with both private/public), and other family characteristics (eg, parental education, marital status, and income) were not associated with IFS scores.

**Conclusion** Families of children with CRF are greatly impacted by their child's health. In contrast to other children with special health care needs, family characteristics were not associated with IFS scores, excluding insurance type. These results may reflect more uniform demands and stressors related to CRF. Future research should identify interventions to attenuate the impact of CRF. *(J Pediatr 2016;175:40-6)*.

Among the population of children with special health care needs (CSHCN), those with chronic respiratory failure (CRF) represent a particularly vulnerable group.[1-3] CRF in these children may manifest on the severe end of the spectrum with impaired gas exchange (ie, hypercapnea and/or hypoxia), altered minute ventilation, impaired secretion clearance, aberrant work of breathing, failure to thrive, and other clinical signs and symptoms. The complexity of underlying disabilities, including neuromuscular conditions, chronic lung disease, airway abnormalities, and other multisystem disorders, contribute to a degree of prognostic uncertainty and, often, idiosyncratic clinical trajectories.[4-7] Families motivated to care for their children at home have been supported by evolving home-care equipment, adapted practice models, and augmented community services.[8-10] Practical and emotional challenges, however, remain and may increase with the complexity of the child's conditions and the underlying nature of respiratory failure, regardless of the nature and extent of the mechanical supports chosen.[11]

Families of children with CRF assume an altered parental role, serving as parent, care coordinator, respiratory therapist, intensivist, nurse, and any other required function.[12-14] These often-unconscious adaptations represent "natural" investments and responses to challenges.[15] Limited qualitative and quantitative studies have revealed alterations in child and parental health-related quality of life (HRQL) in the presence of CRF.[16-19] Additional exploration of the implications for the family unit is needed.

HRQL is a multidimensional construct, incorporating physical, emotional, and role (family, community, and societal) functioning. In a previous analysis, we identified markedly low levels of HRQL in children with CRF and their families, relative to the general population.[11] Both severity of illness and the etiology of the CRF as an acquired, rather than congenital condition, were associated significantly with compromised HRQL. Parental self-reporting of their own HRQL, notably their emotional functioning, was comparable with previous findings from parents of children embarking on hematopoietic stem cell transplant.[20] Parent emotional functioning scores were markedly lower than those obtained from families living with the chronic challenges of sickle cell anemia,[21] and when compared with age- and sex-matched normative data, these scores correspond to less than the 25th percentile on the Mental Health Scale of the Short Form-36, a conceptually similar construct.[22] In a previous qualitative report, Carnevale et al[18] described an overarching theme of "daily living with distress and enrichment" when parents care for children with CRF.[18]

In the current study we intended to further assess the impact of CRF on the family unit by using the Impact on Family Scale (IFS), a validated, parent-reported

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| CAHPS | Consumer Assessment of Healthcare Providers and Systems |
| CAPE | Critical Care, Anesthesia, Perioperative Extension and Home Ventilation Program |
| CHRIs | Child Health Rating Inventories |
| CRF | Chronic respiratory failure |
| CSHCN | Children with special health care needs |
| HRQL | Health-related quality of life |
| IFS | Impact on Family Scale |

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measure of family impact.[23,24] Previous studies in which authors investigated the impact of children's chronic illness on their families (eg, asthma, hemoglobinopathies,[25] spina bifida,[26] rheumatic diseases,[27] cardiac dysfunction,[28] oncologic diagnoses,[29] prematurity,[30] and hypoxic ischemic encephalopathy[31]) suggest that impact varies with severity of the condition,[32,33] but the average scores across populations suggest some uniformity. Our hypothesis was that IFS scores for parents of children with CRF would be appreciably greater, ie, greater negative impact, compared with other pediatric populations. Despite the heterogeneity of our group, our supposition was that CRF, because of the demands of care and clinical trajectories, which may be characterized by cumulative morbidities and disease progress, had a greater effect than other chronic conditions. We then sought to identify demographic and clinical factors associated with parental reporting of family impact and evaluated the association of health care utilization on IFS. Recognition of the extent and nature of the impact of chronic illness on families may help refine comprehensive care models designed to buttress parental supports, improve patient and family-related outcomes, and improve resource utilization.

## Methods

The Critical Care, Anesthesia, Perioperative Extension and Home Ventilation Program (CAPE) was established in June 2007 at Boston Children's Hospital to care for children with CRF throughout New England. Underlying diagnoses represented known patterns of disease,[3,8] including a small proportion of intrinsic lung disease and a predominance of neuromuscular conditions with secondary respiratory insufficiency (eg, spinal muscular atrophy, muscular dystrophies, spinal cord injury, and complex conditions related to hypoxic ischemic encephalopathy or epilepsy syndromes). Described elsewhere,[34] CAPE program objectives were to provide comprehensive, longitudinal service through individualized care, including home visits, liaison with rehabilitation programs and outpatient clinics, school in-services, inpatient consultation, and 24-hour/day response for remote acute-care management. In February 2012, CAPE Program provision was enhanced with the addition of social work support, a nurse practitioner, and administrative care coordination assistance.

Three hundred twenty-seven CAPE Program patients, who began receiving care through the CAPE program before March 31, 2013, were screened for eligibility for inclusion in a multifaceted family-reported outcomes effort. Ineligible program participants included age ineligibles (ie, <30 days or >22 years) (n = 64), single consults (n = 48), patients living in residential facilities (n = 15), and medical/other reasons (n = 4). All potentially eligible participants (n = 196) received an information letter approved by the Boston Children's Hospital Institutional Review Board describing the study and inviting them to participate with an opt-out card; 16 parents opted out. All remaining parents (n = 180) were contacted by telephone to further assess study eligibility and obtain consent. Parent caregivers had to be at least 18 years of age, have a working knowledge of English or Spanish, and the ability to provide consent for their own participation. If parents had more than one child receiving care from the CAPE program, only one child was selected for the family-reported outcomes study.

Among the 196 eligible parents, 158 (81%) consented to participate and were enrolled in this study; 118 (60%) completed the first (study entry) assessment, although 4 of the 118 did not complete the Consumer Assessment of Healthcare Providers and Systems (CAHPS). Patients were classified as developmental-phase participants (enrolled before February 2012) or enhanced-phase participants, who enrolled in CAPE after staffing expansion. The Boston Children's Hospital Committee on Clinical Investigations reviewed and approved the study protocol. All data from this study were pooled and reported anonymously to assure protection of patient and provider rights.

Parents completed a battery of HRQL questionnaires, including the IFS, Child Health Rating Inventories (CHRIs), a modified CAHPS, and Patient Activation Measure-Parental Adaptation, as well as demographic information on patient and family characteristics. Results of the Patient Activation Measure-Parental Adaptation and complete CHRIs[11] from the developmental phase of the clinical program (before February 2012) are reported elsewhere. On the basis of those findings, the CHRIs global and parent emotional function subscales were used to inform the current IFS analysis. Child age and sex, number of siblings, parent responder characteristics (ie, age, sex, education, job status, race and ethnicity, marital status, and family income), and the child's primary and secondary insurance type (private only, public only, both private/public) were collected. The IFS inventory is a validated instrument designed to evaluate the impact of childhood illness on a family with individual items, to assess/measure global impact on the family, financial impact, familial-social impact, personal strain, and mastery.[23,24,35]

Initially, all 27 items were used in scoring, but with further validation, the authors recommended the use of a psychometrically more robust 15-item total score.[23] Responses are selected from a 4-point Likert-type scale (strongly agree—strongly disagree), yielding a total score (score range, 15-60 on the 15-item version; greater scores connoting greater impact in both cases). Although our analysis primarily focuses on the 15-item score, the 27-item score was calculated for comparison with other studies. The IFS also includes 6 additional items, yielding a separate sibling score (ie, reflecting impact on the brother or sister of the child with CRF) that is not included within the total score. All IFS items use an acute, 1-week recall period. The IFS is available in English and Spanish, and parent participants completed the IFS every 6 months for up to 3 time periods. The current analysis focuses primarily on the first completed IFS (n = 118), although a limited comparison is made with the 6-month IFS score (n = 85).

Parents also completed the parent and child-proxy versions of the CHRIs-General.[20,36] The 7-item parent emotional