

Kawasaki Disease Substantially Impacts Health-Related Quality of Life

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Objective To prospectively evaluate the acute impact of Kawasaki disease (KD) on health-related quality of life (HRQoL) and to assess deterioration in the HRQoL experienced by children with KD compared with other childhood diseases.

Study design We merged the Outcomes Assessment Program database obtained prospectively with the existing KD database and queried for KD admissions between 1 month and 13 years of age. HRQoL was evaluated with the parent-proxy Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core and Infant Scales. We compared the KD HRQoL results with those obtained from newly diagnosed patients with cancer and pneumonia, matched for age, sex and race. PedsQL total scores over time were assessed with ANCOVA models, adjusted for matching variables and PedsQL score prior to admission.

Results We identified 89 patients with KD and compared 65 subjects with an equal number with pneumonia and with 67 subjects with newly diagnosed cancer. Patients with demonstrated lower PedsQL total score on admission and suffered a significantly greater HRQoL decline from baseline to admission than the other groups. KD diagnostic subtype (complete or incomplete) and coronary artery dilatation were not associated with HRQoL outcomes. However, non-intravenous immunoglobulin responders showed greater HRQoL decline than responders ($P = .03$).

Conclusions Children with KD suffer acute and significant HRQoL impairment exceeding that of children newly diagnosed with cancer. Lack of immediate treatment response may exert an additional HRQoL burden, whereas KD subtype and coronary artery dilatation do not. (*J Pediatr* 2017;■■■:■■■-■■■).

Kawasaki disease (KD) is an acute vasculitis occurring primarily in young children, and showing a predilection for the coronary arteries.^{1,2} Coronary artery inflammation can result in aneurysm formation with subsequent thrombosis or stenosis or both.³⁻⁵ A portion of the population with KD exhibits lifelong cardiac disability, and some patients even suffer myocardial infarction.⁶ The etiology of KD remains elusive. Moreover, the diagnosis is based on symptoms and inflammatory signs that mimic other childhood diseases, often creating difficulties and delays in treatment.^{1,2,7} Clinical trials performed over 30 years ago show some efficacy of intravenous immunoglobulin (IVIG) infusion treatment in reducing incidence of aneurysm.^{8,9} However, these trials were performed in children with fever occurring within an initial 10-day period. Adding potentially serious cardiovascular sequelae with vague long-term prognosis contributes to prolonged uncertainty and psychological distress to families.^{10,11}

Despite witnessed anxiety, few studies have evaluated the effect of acute and subchronic KD from the caregivers' perspective. The multidimensional health-related quality of life (HRQoL) assessment encompasses physical, mental, and social domains that affect health.^{12,13} Patient-reported outcomes, including HRQoL measures, provide important information regarding clinical and behavioral interactions in a wide variety of medical conditions.^{14,15} The Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core Scales and the PedsQL Infant Scales are standardized population HRQoL instruments with recognized reliability and validity in assessing physical and psychological functioning, both in the outpatient and the hospitalized pediatric population.¹⁶⁻²²

Based upon clinical experience, the extant literature on KD-induced stress,^{10,11} and prior studies of HRQoL in children with severe acute and chronic illnesses,¹⁷⁻¹⁹ we hypothesized that parents perceive that their children with KD exhibit severe physical and psychosocial deterioration, comparable with that experienced by other newly established acute or chronic diagnoses. Accordingly, we compared HRQoL in KD with community-acquired pneumonia and cancer. In addition, we sought

HRQoL	Health-related quality of life
IVIG	Intravenous immunoglobulin
KD	Kawasaki disease
OAP	Outcomes assessment program
PedsQL	Pediatric Quality of Life Inventory
SCH	Seattle Children's Hospital

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to determine whether specific KD characteristics, including KD subtype, responsiveness to IVIG, and coronary artery dilatation, influence HRQoL decline.

Methods

We performed a single-center, prospective cohort study that used the framework of 2 existing institutional databases at Seattle Children's Hospital (SCH): the KD Research Program and the Outcomes Assessment Program (OAP). Patient enrollment occurred between October 1, 2011 and October 31, 2015. The SCH KD Research Dataset encompasses multidisciplinary prospectively collected information including clinical, treatment, and surveillance echocardiographic data for patients diagnosed with complete or incomplete KD as defined by the 2004 American Heart Association guidelines.¹ The OAP is designed to routinely assess child health-related quality of life and family experiences of patients admitted to SCH by the use of self-reported outcome measures. All study procedures regarding the KD Research Program, the Outcomes Assessment Program, and the creation of our subsequent dataset have been reviewed and approved by the Seattle Children's Research Institute Institutional Review Board.

The KD cohort included patients older than 1 month and younger than 13 years of age, who were admitted from October 1, 2011 through October 31, 2015, and whose caregivers participated in the OAP surveys. Patients with KD were identified by *International Classification of Diseases, Ninth Revision* code 446.1 appearance in any position in the patient's medical record diagnosis list. In case of multiple admissions, we included the chronologically oldest encounter. Direct inpatient admissions and transfers from outside institutions were excluded. Two comparison cohorts with alternate diagnoses were formed. The first group consisted of previously healthy patients with a principal discharge diagnosis of uncomplicated community-acquired pneumonia. Patients with respiratory illness associated with complex medical conditions, such as cystic fibrosis, malignancy, lung disease of prematurity, neurologic and neuromuscular diseases, known congenital or acquired immunodeficiency, hereditary anemias, and ventilator-dependency were excluded from this cohort. The second comparison group included previously healthy children, admitted from the Emergency Department with various medical complaints and ultimately discharged from the hospital with a de novo malignancy. Patients in protective isolation, with developmental delays, and those who had been approached to participate within the past 2 months were excluded per OAP protocol eligibility criteria.

The OAP measured patient- and family-centered outcomes related to the care of children admitted to SCH. The PedsQL Infant Scales instrument was addressed to caregivers of admitted patients 1-24 months old, while caregivers of children 2-13 years old completed the PedsQL 4.0 Generic Core Scales instrument. The use of Infant Scales instrument along with the Generic Core Scales instrument has been previously validated.²² The HRQoL scores are derived by assessing for (1) physical functioning, (2) emotional functioning, and (3) social

functioning. In addition to the above elements, the (PedsQL) 4.0 Generic Core Scales considers (4) school functioning, whereas the PedsQL Infant Scales consists of (5) physical symptoms and (6) cognitive functioning when compiling the total score.²⁰⁻²²

Each parent-proxy response is reverse-scored in a scale format and linearly converted to a scale of 0-100. Those individual items are combined and divided by the number of items answered to derive the total physical and psychosocial scores, respectively. Higher total scores indicate better HRQoL.

Data Collection

The primary outcome measure was the PedsQL total score (0-100 scale) composed by the physical and psychosocial subscale scores. Within 72 hours of hospital admission, caregivers were instructed to fill out a "baseline" PedsQL form reflecting HRQoL at 1 month prior to admission, and a form reflecting HRQoL at the time of admission. A follow-up PedsQL form was completed 2-12 weeks after hospital discharge. Improvement scores were derived by calculating the difference between the total, physical, and psychosocial scores on admission from the scores at follow-up. Return to baseline was defined as follow-up score within 4.5 points below the baseline value based on previously published data defining the minimal clinically important decrease in HRQoL.^{18,21}

Sociodemographic characteristics of participating patients and their caregivers were derived from the OAP dataset. Patient's age, sex, race, parental age, level of education, primary language, and insurance type were recorded at the initial phase of recruitment.

Clinical data of all eligible subjects with KD were extracted from electronic medical records and the SCH Kawasaki Research Database. Patients were divided into subgroups according to the type of KD, complete or incomplete, responsiveness to IVIG treatment, and coronary artery status. We defined the time to establishing the diagnosis as the interval between the patient's presentation to the emergency department until the initiation of IVIG infusion. Categorization, as complete and incomplete KD, was based on the diagnostic criteria by the American Academy of Pediatrics and the American Heart Association joint guidelines.¹ Successful response to IVIG treatment was defined as resolution of fever within 36 hours from the end of the initial infusion without requirement of a second IVIG dose or additional KD treatment interventions (except aspirin). Coronary artery classification was based on echocardiographic features either at the time of diagnosis or at 2- and 6-week follow-up time points per KD protocol. In accordance with the 2004 the American Heart Association scientific statement, echocardiography was considered positive if any of 3 conditions were met: z score of left anterior descending or right coronary artery ≥ 2.5 ; coronary arteries met the Japanese Ministry of Health criteria for aneurysms, or ≥ 3 other suggestive features, including perivascular brightness, lack of tapering, decreased LV function, mitral regurgitation, pericardial effusion, or z score in left anterior descending or right coronary artery of 2-2.5. Patients with KD were divided in 3 groups: (1) no or minimal coronary artery

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