

# The Pediatric Quality Appraisal Questionnaire: An Instrument for Evaluation of the Pediatric Health Economics Literature

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## ABSTRACT

**Objectives:** Currently there is no tool available to adequately appraise the quality of the pediatric health economics literature. A comprehensive pediatric-specific instrument would be valuable in informing allocation decisions related to pediatric interventions and services. The goal of this study was to develop the Pediatric Quality Appraisal Questionnaire (PQAQ).

**Methods:** A draft instrument was constructed from published checklists and questionnaires. New questions pertaining to the pediatric population were incorporated. An expert panel reviewed the draft instrument and the proposed scoring scheme for face and content validity. A revised version was pilot tested by three independent appraisers. After addressing discrepancies in scores, a final version was created and subjected to interrater and test-retest reliability assessment.

**Results:** The 57 items in the final PQAQ were mapped onto 14 domains: economic evaluation, comparators, tar-

get population, time horizon, perspective, costs and resource use, outcomes, quality of life, analysis, discounting, incremental analysis, sensitivity analysis, conflict of interest, and conclusions. Among the 57 items, 46 have response options that are scored from 0 to 1. Interrater reliability was 0.75 (95% confidence interval [CI] 0.66–0.81) and test-retest reliability was 0.92 (95% CI 0.71–0.98).

**Conclusions:** The PQAQ is a comprehensive instrument demonstrating face and content validity and strong interrater and test-retest reliability in the appraisal of pediatric economic evaluations. This tool will be valuable to health economists, methods researchers, and policy decision makers involved in allocation decisions for pediatric health care.

**Keywords:** economic evaluation, pediatrics, publication, quality appraisal.

## Introduction

As health-care delivery becomes increasingly constrained by economic considerations, decision makers demand high-quality economic evaluations of interventions, services, and technology to facilitate allocation decisions. In recent years, standard methods for the conduct of economic evaluations have evolved, catalyzed by growth in the pharmaceutical sector [1,2]. Nevertheless, the validity of applying standard health economic methods to a pediatric population has yet to be examined. Health economic evaluations of pediatric interventions must recognize that children rely on parents, teachers, and others to provide access to health care and to

report on the use of health resources; that recommended outcomes such as quality-adjusted life-years or willingness to pay may be difficult to assess in children; and that different outcome measures corresponding to successive stages of physical development may be required [3–7].

As a first step in a research program in pediatric health economic methods, a comprehensive database of all pediatric economic evaluations published from 1980 to 1999 was created [8]. With this database available, an important research objective was to evaluate the quality of published pediatric economic evaluations. Nevertheless, existing economic appraisal instruments appeared inadequate for the formal appraisal of the pediatric literature. These instruments do not include items related to parent/caregiver proxy reporting for outcomes and for health resource use, do not capture time losses of parents and caregivers, do not assess the need for an adequate time horizon to capture future costs and

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consequences, do not consider pediatric quality of life, and do not address the family as a unit of analysis. Thus, there remained a need to develop a pediatric-specific quality appraisal instrument. This article describes the systematic development and testing of a new instrument, the Pediatric Quality Appraisal Questionnaire (PQAQ).

## Methods

The PQAQ was developed through a formal process of item selection and reduction, domain construction, assignment of response options, development of a scoring scheme, external review, pretesting, and reliability assessment.

### *Initial Questionnaire Construction*

A comprehensive initial version of the PQAQ was constructed based on published checklists and questionnaires that evaluated the quality-of-health economic evaluations [9–24]. These published instruments appraised research published in pharmacy, medical, or health economics journals [12,14–18] or economic studies employing a particular analytic technique, such as cost–benefit analysis (CBA), cost-effectiveness analysis (CEA), or cost-utility analysis (CUA) [9,11,13,22,24]. In addition to instruments found in peer-reviewed journals, those available from public agencies and textbooks were reviewed [19,20]. Because available instruments lacked items that address the measurement of pediatric and parental costs and consequences, questions pertaining to the pediatric population were newly formulated. Several existing instruments ask the appraiser to rate the appropriateness of analytic steps, such as costing and modeling. However, many issues in economic evaluation method remain controversial, and rating the appropriateness of the evaluation components could be subjective. In the development of the PQAQ, a deliberately comprehensive approach was taken. “Appropriate” costing could be defined as including all relevant cost items, including productivity and time costs when necessary, including costs borne outside the health-care system, and citing the sources for resource use volume and prices. The preferred approach was to break down the definition of “appropriate” by including a series of yes or no questions regarding each step. Whereas this increases the number of items in a questionnaire, it reduces subjectivity. Related items were grouped together in domains based on established instruments and based on themes emphasized in pub-

lished guidelines for economic evaluation [9–12,14–20,22–24].

### *External Review*

A seven-member expert panel was assembled to assess the face and content validity of the draft PQAQ. This consisted of ensuring that all relevant items were included and evaluating the proposed scoring approach. The panel was chosen to represent expertise in health economic methods, quality appraisal of health economic publications, and pediatric research. Several members were non-Canadian or had extensive experience in international health economic appraisal. As part of the external review, the experts were asked to: 1) comment on the wording of the questions and recommend the deletion or addition of items; 2) rate each question as having high, medium, or low importance for inclusion in the PQAQ; and 3) comment on the various options for scoring the PQAQ and/or provide suggestions for alternative scoring schemes.

Based on the experts’ ratings of each item in the PQAQ as having high, medium, or low importance for inclusion, a distribution pattern of ratings for each item was determined. The frequency for each possible pattern of response distribution was calculated. “Strong” agreement among experts was specified if five or more experts agreed on the particular rating of high, medium, or low. If four experts agreed on the rating, a “medium” level of agreement was assigned. If agreement was found between three experts or fewer, the level of agreement was designated as “weak.”

The agreement between the experts’ rating of highly important items and published economic evaluation quality appraisal instruments was evaluated by calculating the percent observed agreement and a kappa coefficient. A rating was classified as high if five or more experts rated the item as having high importance and if the item appeared in four or more published instruments.

### *PQAQ Scoring*

The PQAQ responses were all categorical. Most of the items had response options that followed the following format: yes (explicitly stated); yes (inferred from text, tables, or figures); no; unknown/not stated/cannot tell; or not applicable. Distinguishing between explicitly and implicitly stated responses was based on instruments from the literature that used similar approaches [14,15,17,18,22]. For these items, full weight could be given to explicit required elements, and partial weight to elements that were implied. Several

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