Establishing Disability Weights for Congenital Pediatric Surgical Disease: A cross-sectional, multi-modal study

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Background

Congenital anomalies are one of the top 10 global causes of mortality for children under 5. These conditions typically require life-saving surgical intervention, and even with treatment can lead to life-long disabilities.

The Global Burden of Disease study and the Developing Countries Project (DCP) utilize the Disability-Adjusted Life Year (DALYs) framework:
- Quantifies the impact of both mortality and morbidity in a single health utility measure
- Requires a disability weight (DW) for each condition, a measure between 0 (perfect health) and 1 (death)

Previous evidence includes a variety of single-method study designs in the development of DWs, but suffers from multiple limitations. There is currently no data using a multi-method approach to establish DWs in pediatric surgery, neither in Canada nor in a Low- and Middle-Income Country (LMIC).

The objective of this study was to develop a set of DWs for 15 congenital anomalies prevalent in both Kenyan and Canadian pediatric surgical patients using a multi-method Burden of Disease approach.

Methods

A wide variety of Kenyan and Canadian healthcare professionals and community members were recruited from Kijabe, Kenya and Hamilton, Canada.
- Participants were provided with 15 expert-panel derived health states, using 6 disability domain descriptions adapted from the EuroQol-5D.
- Focus groups were conducted
- Participants rated each health state using 4 health valuation exercises (2 psychometric and 2 economic), from which the DW data was subsequently derived:
  - Preference Ranking
  - Visual Analog Scale
  - Paired Comparison
  - Time Trade-Off

Findings

In total 154 participants were recruited, 78 from Kenya and 76 from Canada. Mean DWs from each exercise were calculated and combined to yield overall DWs for each health state on a scale from 0 (perfect health) to 1 (death).

We examined country data and statistically analyzed any differences in DWs between the two countries.

Interpretation

Overall DWs were fairly concordant between countries, with a few disparities:

- Internal validity between the four methods was confirmed through statistical analysis
- External validity was demonstrated through comparisons of the DWs with those from the Disease Control Priorities in DCP2

Disability weights for most health states were consistent between DCP2 and DAPS, however:
- Two sets of conditions (cleft lip and cleft palate) received significantly lower DWs in the DCP2
- The actual disability associated with these health states may be presently underestimated by DCP2

We established novel DWs for pediatric congenital anomalies using a low cost multi-method approach. These DWs:
- do not appear to differ significantly across cultural contexts
- can be used to calculate burden of global pediatric surgical disease using the Disability-Adjusted Life Year metric
- will ultimately serve to inform global public health priorities

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