The burden of waiting: DALYs accrued from delayed access to pediatric surgery in Kenya and Canada

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A B S T R A C T

Background: Disability-adjusted life years (DALYs) have become the standard metric for estimating burden of disease (BoD), but have not yet been applied to delayed access to surgical procedures. This study estimates the DALYs accrued from delayed access to surgical care in two pediatric surgical units in Kenya and Canada.

Methods: Records of operations for 13 congenital health states in a Kenyan and a Canadian hospital were prospectively collected for 2012. DALYs caused by delayed presentation were estimated using disability weights and ideal and actual age at surgery.

Results: 1208 first-time procedures in general surgery, neurosurgery, plastic surgery, and urology were included. Delays were longest in general surgery and longer in Kenya than in Canada in all specialties. The longest delays in Kenya were for orchidopexy (72 months) and anorectoplasty (PSARP) (74 months), and in Canada for orchidopexy (40 months). Corresponding total delayed BoD was highest in general surgery and neurosurgery and higher again in Kenya than in Canada (484 cf. 84 DALYs).

Conclusions: Estimating BoD resulting from delayed surgery is feasible and reflects both late presentation and limited access to care. Further exploration of these factors can make delayed DALYS a useful measure of health care coverage and waitlist prioritization.

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While a few recent studies are starting to explore the impact of surgery through averted DALYs in various specialties and to compare this impact between high-income countries (HICs) and LMICs [15–19], there is to date no information on the magnitude of the BoD caused exclusively by delayed access to surgical care. This “delayed burden of disease” component represents the burden suffered by patients who live with a given surgical condition for months or years, while awaiting treatment.

The objective of this study was to use a recently developed surgical DALY methodology to compare the delayed burden of surgical disease accrued through 13 congenital health states addressed by pediatric surgery in a high-income country (Canada) with a low-income country (Kenya).

1. Methods

1.1. Settings

BethanyKids at Kijabe Hospital (BKKH) is an international faith-based pediatric surgical unit located in a secondary and tertiary general hospital 65 kilometers outside of Nairobi, Kenya. The unit relies on external funding through various agencies and donors to provide affordable surgical care and rehabilitation.

McMaster Children’s Hospital (MCH) is a regional pediatric academic tertiary care center located in Hamilton, a major metropolitan city in southern Ontario, Canada. Comparative data regarding the two sites have been presented in a recent publication [20].

1.2. Data collection

We chose to compare 13 health states addressable through pediatric surgical procedures in both centers. The reasons and procedures used in choosing these health states are detailed in our recent publication [20]. Twelve distinct surgical procedures were identified as proxies for treating these health states, as two of the health states (hypospadias and abdominal defect) each included a mild and a severe form, while one state (severe imperforate anus) required two procedures.

After research ethics approval at both sites (Hamilton Health Sciences Integrated Research Board (#11-328) and the Kijabe Hospital Research Ethics Board), medical records were reviewed for all patients under the age of 18 who had one of these surgical procedures at BKKH or MCH between November 2011 and October 2012. Data on birth date, sex, diagnosis, surgical procedure, and age at time of procedure were abstracted from patient records. Patient selection in Canada was based on the associated Ministry of Health and Long-term Care Schedule of Benefits surgical procedure code rather than primary diagnosis or health state. We excluded all secondary/redo procedures, as well as primary procedures deemed to have been intentionally delayed because of concurrent patient risk factors and/or comorbidities.

1.3. Burden of disease (BoD) analysis

The disability prevented through surgery at each site was estimated using published DALY-based methods [5]. DALYs caused by delayed access to surgery for each procedure were estimated using the formula:

Delayed DALYs = DW × (age at surgery–ideal age at surgery),

where DW is the disability weight recently estimated for each procedure in both countries [20], and the ideal age at surgery was derived from the literature using current clinical practice guidelines where available (Table 1) or by expert consensus in the absence of published guidelines.

Once delayed DALYs per patient were calculated, the total number of delayed DALYs accrued at each site over a period of one year was calculated and compared. Descriptive and comparative statistics, including counts, percentages, means and Student’s t-test were performed using Microsoft Excel®.

2. Results

The study included 1423 surgical procedures in patients younger than 18 years of age within the identified 13 health states. After removing secondary procedures and those intentionally delayed for medical reasons, there were 1208 surgical procedures left. The distribution of the 12 procedures across sites, genders and specialties is detailed in Table 2. The delay in surgical intervention was first estimated in months, and is detailed by procedure in Table 3 and by specialty in Fig. 1. As noted, mean delays in surgery were statistically significantly higher in Kenya for most procedures which included sufficient data points, with the exception of hydrocephalus procedures which had similar mean ages across sites.

The cumulative DALYs resulted from this temporal delay were then estimated and depicted in Fig. 2 by procedure and Fig. 3 by specialty. These cumulative values reflect both the predominance of neurosurgical procedures at BKKH, and the major delays at that site in the care of children primarily with intestinal and urological conditions.

Finally, mean delayed DALYs incurred for each surgical procedure were also estimated by procedure type (Fig. 4) and by specialty (Fig. 5). These figures highlight the impact of delays in the care of children particularly with intestinal, urological, and plastic surgery conditions.

3. Discussion

The current study attempts for the first time to quantify the delayed burden of surgical disease, placing an empirical value on the well-known reality of surgical procedures in low- and middle-income countries (LMICs) often being significantly delayed [21,22]. Moreover, the

**Table 1**

<table>
<thead>
<tr>
<th>Condition</th>
<th>Recommended age at surgery</th>
<th>Publication year</th>
<th>Source</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hypospadias</td>
<td>6–18 months</td>
<td>2009</td>
<td>European Association of Urology (EAU) and the National Guideline Clearinghouse (NGC) <a href="http://www.guideline.gov/content.aspx?id=12594">http://www.guideline.gov/content.aspx?id=12594</a></td>
</tr>
<tr>
<td>Cleft lip with no cleft palate</td>
<td>3 months</td>
<td>2012</td>
<td>NHS <a href="http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx">http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx</a></td>
</tr>
<tr>
<td>Cleft palate with or without cleft lip</td>
<td>6–12 months</td>
<td>2012</td>
<td>NHS <a href="http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx">http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx</a></td>
</tr>
<tr>
<td>Undescended testes</td>
<td>12–18 months</td>
<td>2009</td>
<td>European Association of Urology (EAU) and the National Guideline Clearinghouse (NGC) <a href="http://www.guideline.gov/content.aspx?id=14430">http://www.guideline.gov/content.aspx?id=14430</a></td>
</tr>
<tr>
<td>Hirschsprung’s disease</td>
<td>At diagnosis</td>
<td></td>
<td>IPEG guideline reviewed with no mention of age <a href="http://www.ipeg.org/hirschsprungs/">http://www.ipeg.org/hirschsprungs/</a></td>
</tr>
<tr>
<td>Hydrocephalus</td>
<td>At diagnosis</td>
<td></td>
<td>American Association of Neurological Surgeons <a href="http://www.pediatricsurgerymd.org/AM/Template.cfm?Section=List_Of_Conditions1&amp;template=/CM/ContentDisplay.cfm&amp;ContentID=4289">http://www.pediatricsurgerymd.org/AM/Template.cfm?Section=List_Of_Conditions1&amp;template=/CM/ContentDisplay.cfm&amp;ContentID=4289</a></td>
</tr>
<tr>
<td>Imperforate anus</td>
<td>3–4 weeks of age</td>
<td></td>
<td>APSA <a href="http://www.pediatricsurgerymd.org/AM/Template.cfm?Section=List_Of_Conditions1&amp;template=/CM/ContentDisplay.cfm&amp;ContentID=4289">http://www.pediatricsurgerymd.org/AM/Template.cfm?Section=List_Of_Conditions1&amp;template=/CM/ContentDisplay.cfm&amp;ContentID=4289</a></td>
</tr>
<tr>
<td>Spina bifida</td>
<td>48 hours of life</td>
<td>2005</td>
<td>American Association of Neurological Surgeons</td>
</tr>
<tr>
<td>Intestinal atresia</td>
<td>At birth</td>
<td></td>
<td>Expert consultation</td>
</tr>
<tr>
<td>Abdominal wall defect</td>
<td>At birth</td>
<td></td>
<td>Expert consultation</td>
</tr>
</tbody>
</table>

* “Mild” and “severe” forms of hypospadias, spina bifida and imperforate anus are grouped together in the absence of distinct management protocols.
study compares this delayed burden between two pediatric surgical units in two very different populations, with very different resources. Similarly to our previous report [20], the countries were chosen nonrandomly as sites of clinical activity of the authors, and are re

### Table 2

<table>
<thead>
<tr>
<th>Procedure</th>
<th>MCH F</th>
<th>M</th>
<th>Total</th>
<th>BK KH F</th>
<th>M</th>
<th>Total</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abdominal wall closure</td>
<td>4</td>
<td>3</td>
<td>7</td>
<td>0</td>
<td>2</td>
<td>3</td>
<td>10</td>
</tr>
<tr>
<td>Anoplasty</td>
<td>0</td>
<td>2</td>
<td>2</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Colonal pull-through</td>
<td>3</td>
<td>2</td>
<td>5</td>
<td>1</td>
<td>9</td>
<td>10</td>
<td>15</td>
</tr>
<tr>
<td>Colostomy</td>
<td>0</td>
<td>0</td>
<td>7</td>
<td>15</td>
<td>22</td>
<td></td>
<td>22</td>
</tr>
<tr>
<td>Intestinal atresia repair</td>
<td>1</td>
<td>4</td>
<td>5</td>
<td>0</td>
<td>1</td>
<td>1</td>
<td>6</td>
</tr>
<tr>
<td>Orchidopexy</td>
<td>N/A</td>
<td>163</td>
<td>163</td>
<td>N/A</td>
<td>88</td>
<td>88</td>
<td>251</td>
</tr>
<tr>
<td>PSARP</td>
<td>7</td>
<td>2</td>
<td>9</td>
<td>8</td>
<td>14</td>
<td>22</td>
<td>31</td>
</tr>
<tr>
<td><strong>General surgery total</strong></td>
<td>15</td>
<td>176</td>
<td>191</td>
<td>17</td>
<td>130</td>
<td>147</td>
<td>338</td>
</tr>
<tr>
<td>Spina bifida closure</td>
<td>3</td>
<td>3</td>
<td>6</td>
<td>139</td>
<td>144</td>
<td>283</td>
<td>289</td>
</tr>
<tr>
<td>VPS insertion/ETV</td>
<td>5</td>
<td>4</td>
<td>9</td>
<td>143</td>
<td>180</td>
<td>323</td>
<td>332</td>
</tr>
<tr>
<td><strong>Neurosurgery surgery total</strong></td>
<td>8</td>
<td>7</td>
<td>15</td>
<td>282</td>
<td>324</td>
<td>606</td>
<td>621</td>
</tr>
<tr>
<td>Cleft lip repair</td>
<td>7</td>
<td>11</td>
<td>18</td>
<td>18</td>
<td>30</td>
<td>48</td>
<td>66</td>
</tr>
<tr>
<td>Cleft palate repair</td>
<td>11</td>
<td>8</td>
<td>19</td>
<td>2</td>
<td>6</td>
<td>8</td>
<td>27</td>
</tr>
<tr>
<td><strong>Plastic surgery total</strong></td>
<td>18</td>
<td>19</td>
<td>37</td>
<td>20</td>
<td>36</td>
<td>56</td>
<td>93</td>
</tr>
<tr>
<td>Hypospadias repair</td>
<td>N/A</td>
<td>95</td>
<td>95</td>
<td>N/A</td>
<td>61</td>
<td>61</td>
<td>156</td>
</tr>
<tr>
<td>Urology total</td>
<td>N/A</td>
<td>95</td>
<td>95</td>
<td>N/A</td>
<td>61</td>
<td>61</td>
<td>156</td>
</tr>
<tr>
<td>Grand total</td>
<td>41</td>
<td>287</td>
<td>328</td>
<td>319</td>
<td>551</td>
<td>870</td>
<td>1208</td>
</tr>
</tbody>
</table>

MCH = McMaster Children’s Hospital; BKKH = BethanyKids at Kijabe Hospital; PSARP = posterior sagittal anorectoplasty; VPS = ventriculoperitoneal shunt; ETV = endoscopic third ventriculostomy.

### 3.1. Settings and practice patterns

The two settings compared in this study have been previously described in detail [20], and the inherent differences in their patient populations, referral patterns, and clinical strategies have been highlighted. Such differences account for the obvious discrepancies between numbers of specific procedures noted in Table 1 between the sites. This applies particularly to the notable predominance of neuromuscular procedures in BKKH, a reflection of a unique referral pattern established over the past decade in Kenya.

### 3.2. Delay in surgery

The comparison of mean delays in presentation to surgery between the two sites highlights the delayed access to surgery at BKKH. For instance, the average age of children operated for cleft lip was over 3 years, for hypospadias 4 years, and for orchidopexy and posterior sagittal anorectoplasty (PSARP) 6 years. This is consistent with earlier work by the authors [6,16], and thus believed to be a true reflection of delays in access to surgical care in LMICs, in keeping with other reports [24,25].

Only in the case of hydrocephalus were children in Canada operated at an older age, probably reflecting many instances of mild disease being diagnosed only in later childhood. By contrast, the vast majority of hydrocephalus in the East African setting is either spina bifida-related or follows neonatal encephalitis [26], thus the mean age of four months at time of surgery still represents a significant delay.

It is important to note that the delay observed at both sites in our study includes several components: delay of presentation to medical care, delay of referral for specialized care, and finally delay to actual surgery. Especially in HICs there are currently some major efforts underway to quantify and monitor the latter 2 delays (labeled W1 and W2 in the Canadian Paediatric Surgical Wait Times [CPSWT] Project) [27] for purposes of resource allocation, but in fact in both settings the delay to original presentation to health care facilities remains most important and harder to measure.

### 3.3. Delayed burden of disease

The estimated delayed BoD expands simple wait time by factoring in the DW of each untreated condition. This factor, varying between 0 (full health) and 1 (death), has been estimated for several pediatric surgical conditions [28]. The total delayed DALYs shown in Figs. 2 and 3 are also modulated by institutional surgical volumes, thus reflecting the actual delayed burden encountered in a high- and a low-income setting. The mean delayed DALYs per procedure, as shown in Figs. 4 and 5, eliminate the institutional volume factor and thus highlight the surgical conditions in which children suffered the most while waiting for surgery. This burden was highest within general surgery, particularly for intestinal conditions (imperforate anus and Hirschsprung’s disease), with urological conditions (hypospadias and undescended testes) and cleft palate following. What is the significance of the delayed burden of disease? The concept appears necessary, as the only attempt to bring to light and to measure the years spent by any person suffering from a disabling surgical condition before they are treated. It must be noted that this component is, however, essentially untreatable – it cannot be eliminated through surgery, though it can be prevented through prompt surgical care before it starts accumulating. This may be the reason why the concept didn’t even appear in the classic grouping of surgical burden into met, unmet and unmeetable need [13,14]. As these three components must by definition add up to the total BoD attributed to any condition as estimated by the Global Burden of Disease (GBD) Study [8,13], where does the delayed burden fit in? Our previous work has placed it in either of the three groupings [6,15], as it can be considered met need addressed
once the surgical procedure is done, unmet need waiting for a surgical procedure, or as “lost” unmeetable need that can no longer be gained back even after the surgery is completed. Another way to conceptualize this is to split both met and unmet need into an “incident” or “timely” component representing new congenital cases being born, and a “prevalent” or “delayed” component which is the backlog of unoperated children with that condition awaiting surgery [15]. Within this framework (Fig. 6), the delayed burden fits within the prevalent need, as it results from children who have not been treated in a timely fashion.

Time-based estimates of delayed BoD quantify the magnitude of the access to care issue, but do not necessarily provide insights into the causes of such delays. As addressed by many others [25,29–33], the causes are broad and multifactorial, including geographical challenges (distance to appropriate health care points), educational and cultural challenges (knowledge that a condition is treatable and where to seek help), human resource limitations (sufficient numbers of well-trained surgeons and other health care workers), infrastructure issues (roads for transport, hospitals), financial challenges (cost of care, of transport, and lost income while caring for the child), and technological limitations (such as anesthesia and imaging).

The methodology presented applies itself well to congenital conditions which often, but not always, have clearly set guidelines for timely treatment after birth, but it can also be applied to acquired conditions in which prompt treatment is recommended from the time of presentation (e.g. cancer, acute appendicitis). Moreover, while developed for surgical conditions, the delayed BoD concept can in fact be used for medical conditions as well: it can therefore be applied to the stroke, myocardial infarction, or pneumonia patient suffering while waiting to receive appropriate treatment.

3.4. Implications for LMIC settings

The concepts of met and unmet BoD, access to care, effective coverage and backlog have only recently been further explored and applied to global surgical conditions [15,33–35]. Recent reports highlight the great discrepancy between the unmet and the met BoD for various conditions [32,36,37] and the inadequate effective coverage [33,38]. Early global backlog estimates for isolated surgical conditions are sometimes shocking: a recent report estimated that there are more than 600,000 children with unoperated cleft lip/palate in Africa and South-East Asia alone [16].

The current effort continues this process of exploring health care inequities and needs in LMICs by expanding and quantifying the backlog concept. Not only are there hundreds of thousands of children awaiting surgery in LMICs, but they have been waiting for many years all the while suffering from the BoD or disability of their condition. This type of information adds a human rights component to our clinical activity [37,39], allowing pediatric surgeons and other stakeholders to focus their global involvement efforts, and lobby for additional resources needed to address the issue.

3.5. Implications for HICs settings

The delays in access to surgical care, and associated delayed DALYS, are predictably much lower in HICs – but they still exist. While many of the resource limitations operational in LMICs may not be relevant here, delays typically reflect wait times for referral to surgical care as well as waiting for surgical intervention. Such delays are reflected in the wait list policies introduced in many resource-rich settings, and highlight that such wait times may not only incur risk to life, but engender an associated BoD that we implicitly expect our patients and their families to bear [40]. The burden of waiting one year for a hypospadias repair or orchidopexy is not the same as that of waiting with a

Fig. 2. Total delayed DALYs accrued by procedure and site.

Fig. 3. Total delayed DALYs accrued by specialty and site.

PSARP = posterior sagittal anorectoplasty; VPS = ventriculo-peritoneal shunt; ETV = endoscopic third ventriculostomy
colostomy or a cleft lip/palate — and the methodology presented in this paper allows us to estimate and compare BoD accrued through waiting times. Therefore the efforts underway in HICs to establish, monitor and compare surgical wait lists could benefit from BoD data to allow for more equitable comparisons both within and across surgical specialties.

3.6. Limitations

The current study has several significant limitations. In the first place, the choice of the two institutions was arbitrary, and the ensuing comparison included widely divergent practice and referral patterns at each site. Pediatric surgeons in LMICs have a much broader scope of practice, including urology, plastic surgery, and even neurosurgery or orthopedics. Moreover, the nature of the practice in most institutions in LMICs is vastly predicated by available skills, informal referral networks, funding, geographical and transport realities, and widely uneven distribution of specialist care. While the Canadian institution (MCH) may be fairly representative of other similar-sized pediatric surgical units in the developed world, the Kenyan unit (BKKH) is quite unique in its focus and outputs in the developing world. Therefore the findings of this study cannot be easily generalized to other sites, particularly within LMICs.

The study only presents a snapshot of the delayed BoD from a preselected set of 12 surgical procedures. These procedures address the 13 non–random health states representing the pediatric surgical conditions most prevalent at both sites and for which DW estimates existed. Naturally, the extent of the delayed BoD will change significantly among surgical conditions, institutions, and geographical regions.

As mentioned above, the delays observed in our study must be approached with caution. They all include the time to original presentation to a health care facility, thus are not only a reflection of quality of access to care. Moreover, they are imperfect as it is very difficult to identify those patients in both settings whose procedures were intentionally delayed for legitimate, medical, reasons. Moreover, the age at surgery collected at MCH was only available in years, thus needing conversion into months for delay calculations.

Identifying an ideal time for surgery for any given condition is not straightforward, and clearly depends on the context of care. The authors have been as conservative as possible, selecting the older ideal age whenever a range existed in current literature. This approach also addressed issues of anesthetic safety in LMICs, which often predicate intentional delays in surgery within the neonatal and infancy periods.

Finally, the calculation of DALYs also introduced a significant level of

Fig. 4. Mean delayed DALYs per surgery procedure and site.

Fig. 5. Mean delayed DALYs per surgical specialty by site.

Fig. 6. The conventional and expanded models for components of burden of surgical disease.
uncertainty. This is owing to multiple assumptions and controversies in the DW estimates and age-weighting [10].

4. Conclusions

Despite the above limitations, the current study highlights a hitherto unaddressed issue in global burden of surgical disease research. Any child with a congenital disease apparent from birth who is seen and operated on by a surgeon well within her/his childhood or even teenage years has suffered a real and measurable burden of disease which can no longer be redeemed. An awareness of this reality will hopefully motivate surgeons and policy makers alike to increase their efforts globally, while quantifying the delayed burden will foster informed prioritization of surgical activity in both the global South and North.

The recent World Health Assembly side meeting [41], other global action groups [42–44], and independent reports [14,25,37,39,45,46] have all affirmed the importance of surgery as an essential primary care intervention. Knowing not only the extent of the global need we face, but also the human cost of delaying intervention can guide us as a profession to face the challenges ahead in an informed and committed fashion.

References