

Global Burden of Pediatric Surgical Disease

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Chapter 1

General Introduction and Scope

Over the past two decades global surgery has emerged as a new surgical discipline. Unlike its “international surgery” predecessor, global surgery is specifically defined as “an area for study, research, practice, and advocacy that places priority on improving health outcomes and achieving health equity for all people worldwide who are affected by surgical conditions or have a need for surgical care.”(1) The focus on equity of resources and advocacy establishes it therefore not only as a professional and academic discipline, but also extends it into social activism and health care policy.

In order to achieve its above-stated goals, global surgery requires reliable metrics of burden of disease (BoD) in any given geographical or specialty area and the costs to “repair” the disease, both in the short- and long-run. Since the mid-1990s, primarily through the work of the Global Burden of Disease (GBD) Study Group (ref), the preferred metric has progressively become the disability-adjusted life-year (DALY). The DALY is a “health gap” measure, a summary measure of health incorporating both mortality and morbidity. It is an example of a health-adjusted life years (HALY) measurement, similar but not identical with the quality-adjusted life year (QALY)(2).



Figure 1: DALY infographic (3)

The DALY basic formula is:

$$DALY = YLL + YLD = N * LE + I * LE * DW,$$

where YLL = years of life lost, YLD = years lived with disability, N= number of deaths in population, LE = life expectancy at onset of disease, I = number of incident cases in the population over period of time, and DW = disability weight. This “incidence perspective” was originally adopted in 1999 for the entire formula because of YLL reflecting incident deaths in a population, but was replaced

from the GBD 2010 study onward to a prevalence-based YLD, with the revised formula $YLD = P * DW$, where P = number of prevalent cases of a health condition (4).

Hence the mortality burden is simply expressed as the combined number of years of life lost through disease-related fatalities, while the morbidity burden modulates the combined number of years spent in disability by the disability weight factor, which is both anchored and constrained between 0 (perfect health) and 1 (equivalent of death).

There are several uncertainties and controversies surrounding the factors in this formula, which result in a spectrum of estimated DALYs rather than unique values. The life expectancy (LE) value used is generally based on a global “standard”, originally and commonly still the LE for Japan (5) – although some researchers have opted for country-specific LE values (6). The number of years lost or lived with disability is also prone to debate, as it can be a straight arithmetic value, or be adjusted to include future discounting and / or age weighting (5,7).

The most debatable factor however in the DALY formula is the disability weight (DW), which was first introduced by Chris Murray in 1994 in the form of 6 operationally-defined classes of disability limitations (4). DW values have generally been estimated using various psychometric and econometric methods, including rating / visual analogue scales (VAS), paired comparisons (PC), standard gambles (SG), person trade-off (PTO), and time trade-off (TTO)(5,8,9).

The GBD 2006 study has estimated DW values for 131 conditions health states (corresponding to 291 diseases) across all medical and surgical specialties, using primarily the PTO technique, in which respondents are asked to choose between curing a certain number of individuals with one disease condition versus another number with a different condition (10). In the recently released GBD 2010 study, the number of health state DW values was expanded to 220 (corresponding to 291 diseases), and the primary estimation method was PC (7). The values thus obtained were then used in the standard DALY formula to derive an extensive set of burden of disease data, categorized by global regions, countries, disease categories and specific conditions (7,10).

While the GBD study has successfully provided a full global, regional and national picture of BoD, it has not been easily applied to any given clinical discipline. Thus, for instance, among its 220 published DW values, only 5 actually apply to congenital surgical conditions (cleft lip, cleft palate,

anorectal malformation, neural tube defects, and cardiac defects)(4,10) – despite the broad spectrum of existing conditions. Thus any potential estimation of BoD within this specialty will require the generation of further DW values for the common pediatric surgical conditions.

Besides the above limitations, the original BoD framework as popularized by the GBD study group was not meant to address surgical disease, and especially the impact of surgical intervention. For this reason surgical researchers have developed a unique framework for surgical BoD. This includes the separation of BoD into what was termed *met burden* (burden of surgical disease already addressed through surgical interventions, measured in *averted* DALYs), *unmet burden* (burden of surgical disease remaining to be addressed at any given point in time, measured in *avertable* DALYs), and *unmeetable burden* (surgical disease deemed inoperable even in ideal settings at a given time, measured in *unavertable* DALYs) (11). While unaverted DALYs are simply “generic” DALYs as estimated by their study, averted DALYs take into account not only the direct impact of surgical intervention in DALY reduction, but also estimates of post-operative mortality and morbidity and probability of residual disease. Therefore this framework is not only able to identify the surgical work completed and that remaining to be done, but can evaluate all surgical procedures by the relative BoD that they can each address.

The various stated limitations in the calculation of DALYs have resulted in an ongoing quest for alternative measures of burden of surgical disease. These have included the concepts of effective coverage (EC), surgical backlog, and cost-effectiveness measures. Effective coverage assesses the percentage of surgical interventions in any given area, relative to a standard number of interventions corresponding to ideal care (12,13). Surgical backlog attempts to estimate the number of unoperated patients with a given condition in an area, using either household surveys or age-based cohort calculations (14,15).

Finally, economic valuations typically attempt to quantify the cost effectiveness of surgical interventions by calculating the cost of surgically averting one DALY or QALY (16–21). Such studies have shown that surgical interventions, including in pediatric and neonatal surgery, are quite cost-effective, at par with many medical interventions and often even more effective than them (22,23). The actual financial impact of surgical interventions can also be estimated using econometric methods including the value of a statistical life (VSL) (24). Such studies have been

incorporated in the recent Lancet Commission on Global Surgery (LCoGS) Surgery 2030, which has shown that surgery could restore up to 2.5% of the GNP of many LMICs (25). Unfortunately such cost-effectiveness analyses (CEA) are scarce in pediatric surgery, and no full economic impact studies have been published to date on children's surgery.

Pediatric surgery is defined as “the diagnostic, operative, and postoperative surgical care of patients from prenatal diagnosis through adolescence with congenital and acquired anomalies and diseases, be they developmental, inflammatory, neoplastic, or traumatic. The scope of this discipline is broadly the same as general surgery, focused especially in infancy and childhood but to include the fetus, adolescent, and young adult with special health care needs arising from childhood surgical conditions”(26). Moreover, its spectrum varies significantly with the geographic and particularly the socioeconomic context (27,28). These features thus make BoD considerations in this relatively young subspecialty a useful model for other surgical specialties.

Theme

The recognition of surgical care as an essential component of health care has required evidence of its potential impact in health care systems and cost-effectiveness by comparison with other standard interventions. Such evidence, essential for advocacy for resource allocation in LMICs, is very limited in children. Not only are there few outcomes and cost-effectiveness studies in the specialty, but the required disability weights for most common pediatric surgical conditions are missing, and multidisciplinary long-term follow-up is virtually non-existing, even in high-resource countries (29,30).

The focus of this thesis is therefore two-fold. In **part I** the theoretical framework for surgical burden of disease measurement is reviewed and critiqued, with several alternative metrics usable in pediatric surgery being offered.

Part II includes several empirical studies exploring the implications and applications of the theoretical framework. This includes generating disability weights within pediatric surgery, then establishing the evidence for the burden of surgical disease in children and the cost-effectiveness of its treatment.

The following research questions are identified

1. What are the limitations of the traditional DALY approach in measuring impact of surgical treatment, and how can current metrics be adjusted and adapted for meaningful use in a surgical specialty? (Chapters 2 and 3)
2. Can valid disability weights be generated for a wider set of pediatric surgical conditions? (Chapter 4)
3. What is the total burden of surgical disease in children in LMICs, and the magnitude of the backlog in surgical care? (Chapters 5 and 6)
4. What is the burden of disease averted through surgical care of children in LMICs, compared to HIC settings? (Chapter 7)
5. What is the burden of disease lost through delayed access to surgical care in LMICs? (Chapters 6, 8, and 9)
6. What is the cost effectiveness and economic impact of surgical care of children in LMICs? (chapters 6, 9 and 10)

In chapter 11 the data are discussed and summarized.

References

1. Dare AJ, Grimes CE, Gillies R, Greenberg SLM, Hagander L, Meara JG, et al. Global surgery: defining an emerging global health field. *The Lancet*. 2014;2245–7.
2. Gold MR, Stevenson D, Fryback DG. HALYs and QALYs and DALYs, Oh My: Similarities and Differences in Summary Measures of Population Health. *Annu Rev Public Health* . 2002;23(1):115–34.
3. Wikipedia Contributors. Disability – Adjusted Life Year . Wikipedia, The Free Encyclopedia. 2016 [cited 2016 Mar 9]. Available from: https://en.wikipedia.org/w/index.php?title=Disability-adjusted_life_year&oldid=708810397
4. Vos T, Flaxman AD, Naghavi M, Lozano R, Michaud C, Ezzati M, et al. Years lived with disability (YLDs) for 1160 sequelae of 289 diseases and injuries 1990-2010: a systematic analysis for the Global Burden of Disease Study 2010. *Lancet* . 2012;380(9859):2163–96
5. Murray CJL. Quantifying the burden of disease : the technical basis for disability-adjusted life years. *Bull World Health Organ*. 1994;72(3):429–45.
6. Poenaru D, Lin D, Corlew S. Economic Valuation of the Global Burden of Cleft Disease Averted by a Large Cleft Charity. *World J Surg* . Springer International Publishing; 2015
7. Murray CJL, Vos T, Lozano R, Naghavi M, Flaxman AD, Michaud C, et al. Disability-adjusted life years (DALYs) for 291 diseases and injuries in 21 regions, 1990-2010: A systematic analysis for the Global Burden of Disease Study 2010. *Lancet*. 2012;380(9859):2197–223.
8. Nord E. Methods for quality adjustment of life years. *Soc Sci Med*. 1992;34(5):559–69.
9. Haagsma JA, Polinder S, Cassini A, Colzani E, Havelaar AH. Review of disability weight studies: comparison of methodological choices and values. *Popul Health Metr* . 2014;12(1):20.
10. Lopez A. Global Burden of Disease and Risk Factors . Lopez AD, Mathers CD, Ezzati M, Jamison DT, Murray CJL, editors. New York, NY: Oxford University Press & The World Bank; 2006
11. Bickler S, Ozgediz D, Gosselin R, Weiser T, Spiegel D, Hsia R, et al. Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg* . 2010;34(3):374–80.
12. Ozgediz D, Hsia R, Weiser T, Gosselin R, Spiegel D, Bickler S, et al. Population health metrics for surgery: effective coverage of surgical services in low-income and middle-income countries. *World J Surg* . Springer New York; 2009
13. Shengelia B, Tandon A, Adams OB, Murray CJL. Access, utilization, quality, and effective coverage: an integrated conceptual framework and measurement strategy. *Soc Sci Med* . 2005;61(1):97–109.

14. Habtamu E, Rajak SN, Gebre T, Zerihun M, Genet A, Emerson PM, et al. Clearing the backlog: trichiasis surgeon retention and productivity in northern Ethiopia. Schachter J, editor. PLoS Negl Trop Dis . Public Library of Science; 2011
15. Wall L. Obstetric vesicovaginal fistula as an international public-health problem. Lancet . 2006;368:1201–9.
16. McCord C, Chowdhury Q. A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. Int J Gynaecol Obstet . 2003 Apr;81(1):83–92.
17. Gosselin R a, Heitto M. Cost-effectiveness of a district trauma hospital in Battambang, Cambodia. World J Surg . 2008
18. Gosselin R a, Maldonado A, Elder G. Comparative cost-effectiveness analysis of two MSF surgical trauma centers. World J Surg . 2010 Mar [cited 2011 Aug 6];34(3):415–9.
19. Poley MJ, Stolk EA, Tibboel D, Molenaar JC, Busschbach JJ V. The cost-effectiveness of treatment for congenital diaphragmatic hernia. J Pediatr Surg . 2002;37(9):1245–52.
20. Poley MJ, Stolk EA, Langemeijer RATM, Molenaar JC, Busschbach JJ V. The cost-effectiveness of neonatal surgery and subsequent treatment for congenital anorectal malformations. J Pediatr Surg. 2001;36(10):1471–8.
21. Magee WP, Vander Burg R, Hatcher KW. Cleft lip and palate as a cost-effective health care treatment in the developing world. World J Surg . 2010 Mar [cited 2011 Jul 25];34(3):420–7.
22. Poley MJ, Brouwer WBF, Busschbach JJ V, Hazebroek FWJ, Tibboel D, Rutten FFH, et al. Cost-effectiveness of neonatal surgery: first greeted with scepticism, now increasingly accepted. Pediatr Surg Int . 2008;24(2):119–27.
23. Chao TE, Sharma K, Mandigo M, Hagander L, Resch SC, Weiser TG, et al. Cost-effectiveness of surgery and its policy implications for global health: a systematic review and analysis. Lancet Glob Heal . 2014
24. Corlew D. Economic Modeling of Surgical Disease: A Measure of Public Health Interventions. World J Surg . 2013;37:1478–85.
25. Meara JG, et al. Global Surgery 2030: evidence and solutions for achieving health, welfare, and economic development. Lancet. 2015;in press.
26. American Board of Surgery. Specialty of Pediatric Surgery Defined . ABS Training and Certification. 2016 [cited 2016 Mar 9]. Available from: <http://www.absurgery.org/default.jsp?aboutpediatricsurgerydefined>
27. Nandi B, Mungongo C, Lakhoo K. A comparison of neonatal surgical admissions between two linked surgical departments in Africa and Europe. Pediatr Surg Int . 2008 Aug [cited 2011 Aug 7];24(8):939–42.
28. Poenaru D. The burden of pediatric surgical disease in low-resource settings: Discovering

- it, measuring it, and addressing it. *J Pediatr Surg* . Elsevier Inc.; 2016;51(2):216–20.
29. van der Cammen-van Zijp MHM, Gischler SJ, Mazer P, van Dijk M, Tibboel D, IJsselstijn H. Motor-function and exercise capacity in children with major anatomical congenital anomalies: An evaluation at 5years of age. *Early Hum Dev* . Elsevier Ireland Ltd; 2010;86(8):523–8.
 30. Olieman JF, Penning C, Spoel M, IJsselstijn H, van den Hoonaard TL, Escher JC, et al. Long-term impact of infantile short bowel syndrome on nutritional status and growth. *Br J Nutr*. 2012;107(10):1489–97.

Chapter 2

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A Square Peg in a Round Hole? Challenges with DALY-based “Burden of Disease” Calculations in Surgery and a Call for Alternative Metrics

Gosselin R, Ozgediz D, Poenaru D.

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Abstract

Introduction

In recent years, surgical providers and advocates have engaged in a growing effort to establish metrics to estimate capacity for surgical services as well the burden of surgical diseases in resource-limited settings. The burden of disease (BoD) studies have established the disability-adjusted life year (DALY) as the primary metric to measure both disability and premature mortality. Nonetheless, DALY-based approaches present methodological challenges, some of which are unique to surgical conditions, not fully addressed through the multiple iterations of the BoD studies, including the most recent study.

Methods and Results

This paper examines these challenges in detail, including issues around age-weighting and discounting, and estimates of disability-weights for specific conditions. Surgical burden measurements of specific conditions, or through the assessment of hospital wards as platforms for service delivery, still have unresolved methodological hurdles. The 2010 BoD study addresses some of these issues, but many questions still remain. Other methods estimating surgical prevalence, backlogs in treatment, and disability incurred by delays in care may provide more practical approaches to disease burden that can be useful tools for clinicians and health advocates.

Conclusions

These approaches warrant further exploration in LMICs and these debates require active engagement by surgical providers and advocates globally.

Introduction

Surgical providers working in low and middle-income countries (LMICs) bear witness to a significant volume of preventable death and disability in some of the world’s most vulnerable populations. A primary goal of surgical initiatives in these settings is to reduce this “burden of surgical disease.” To clinical surgeons, the burden manifests in many forms: the injured patient or expectant mother needing emergency care (and possibly surgery) who succumb or remain disabled, the patient with an acute abdominal emergency who may die from irreversible sepsis with or without surgical intervention, or the correctable deformity or enlarging tumor that inexorably progress, causing increasing levels of disability and/or compromising quality of life to the patient, the family, and the community.

While humanitarian surgeons have attempted to address this burden for decades, its accurate measurement has gained attention in recent years. Of course, many other categories of diseases also go untreated or undertreated, and for this reason, the burden of disease (BoD) approach was developed to lend a uniform approach to the measurement of death and disability. This paper reviews the methodology associated with the BoD approach and highlights the challenges associated with this methodology, even in the most recent iterations of the BoD study. Some of these challenges are inherent to the methodology, while others are more specific to surgical conditions.

This discussion is necessary to improve advocacy for surgical conditions, to measure of burden on patients, families, and health care systems, and ultimately, to define impact on public health. These debates may also improve the BoD metrics for non-surgical diseases as well. This paper first describes the BoD model, then details some of the challenges in applying this model to surgical conditions, and the adaptations required in the process. Finally, other conceptual approaches that have been used or may be explored are suggested.

Current model of burden of disease

The burden of disease (BoD) framework was developed to measure, at the population level, the degree of ill health due to fatal and non-fatal conditions, as well as the risk factors.[1] The DALY

metric was developed as a composite metric of both mortality and disability. More formally, the DALY combines years of life lost (YLLs) for fatal conditions with years of life lived with disability (YLDs): $DALY = YLL + YLD$. The YLL is computed as the life expectancy minus the average age of death. The calculation of YLDs requires estimates of disability weights (DWs) for disease severity (explained further in this paper). The DWs have traditionally been estimated through complex panel/expert opinion, as discussed below. They can also be estimated using a table of increasing disability markers as proposed by Murray (1994), who categorized severity of non-fatal outcomes into six classes, based on the impact on function in various spheres of life activity.[2] While the former set of DWs is limited to the diseases covered by successive BoD studies, the latter can be applied to any known condition and are thus particularly useful for specialties (such as surgery) not sufficiently addressed by published BoD studies.

The impact of health intervention can be measured by the BoD averted in DALYs, using the following formula, for fatal conditions:

$$\text{Averted DALYs from one intervention} = LE_{\text{Age at operation}} * RD * PST$$

($LE_{\text{Age at operation}}$ = years of life lost through early death, PST= probability of successful treatment, RD = risk of death without the procedure).

Thus, for instance, the DALYs averted through an appendectomy for acute appendicitis in a 20-year old patient would be calculated thus:

$$60 \text{ (life expectancy in a population at age 20)} * 0.1 \text{ (mortality of acute appendicitis between 5-24\%)} * 1 \text{ (over 95\% chance of permanent cure for appendectomy)} = 6 \text{ DALYs.}$$

For non-fatal conditions, impact can similarly be measured as

$$\text{Averted DALYs from one intervention} = LE_{\text{Age at operation}} * DW * RPD * PST$$

(RPD = risk of permanent disability).

The DW value used here is that for unoperated cases (before treatment), and the residual disability is accounted for by the PST. Thus, for instance, the DALYs averted through a cleft lip repair procedure in a 10-year old girl would be calculated as

70 (life expectancy in a population at age 10) * 0.05 (DW for cleft lip) * 1.0 (cleft is a permanent disability) * 1.0 (over 95% chance of cure for cleft lip repair) = 3.5 DALYs.

Both RPD and PST range from 0 to 1, thus more DALYs are averted through interventions for conditions with higher risk of long-term disability and high possibility of successful treatment. Multiple investigators have used this approach, with modifications, to estimate the burden of disease averted by a range of surgical interventions in hospitals in LMICs.[3, 4]

Proponents of the BOD methodology cite the uniformity in calculation that allows for comparability between diseases and sequelae without the bias that may be introduced by advocates for particular conditions, or by instruments that can only be used for certain categories of conditions.

Problems with current concept

Almost from its inception, the Global Burden of Disease (GBD) framework and its metric, the DALY, were met with strong criticism. The view of Alkire et al resonates with many researchers: the DALY is a useful tool to compare health outcomes, but presented as a concept in isolation, it has little meaning to most audiences.[5] Yet the DALY is, and will remain, the GBD metric for the foreseeable future. A brief review of its drawbacks will help frame our contention that it might not be the best-suited metric to address the burden of surgical conditions.

One of the main purposes of the BoD study was to inform resource allocation. By nature, it was more driven by incidence than by prevalence. Because of the relatively poor quality of data from LMICs, it had to rely on many assumptions concerning effectiveness, quality and coverage.[6] By nature also, the use of the DALY metric implied some value choices: hypothetical life expectancy, age weighting, discounting and the use of disability weights. These choices are intrinsically subjective, and thus vulnerable to critique on ethical, moral, and/or philosophical grounds.

Based on the equity principle, it was decided that an ideal life expectancy (the longest one at the time, found in Japan) should be the universal norm. Although laudable in concept, it soon became apparent that for countries or regions with low life expectancy, the overall burden -

and therefore also the avoidable burden, particularly for lethal conditions -could be overestimated. Using relevant cohort life expectancies that are country-or region-specific, as suggested by Fox-Rushby, invariably reduces projected life expectancies in LMICs, so the “true” burden is always less than that measured using an “ideal” life expectancy.[7] Thus the use of universal life expectancy, in the context of priority setting and resource allocation, may put unrealistic demands and expectations on policy makers and implementers, and actually decrease the effectiveness of the process.[8] Despite these concerns, until now the BoD framework still uses the hypothetical ideal universal life expectancy.

Age weighting reflects the concept that life is valued differently at different ages. The standard DALY, by using an age-weighting modulation parameter, incorporates the value that life is worth less at its extremes than in the middle productive years. Thus the BoD for children and the elderly is underestimated compared to young adults. Discounting is based on the premise that future years have less value than present years. A standard 3% per year discount, common in banking practices, was applied to DALYs. This approach has been largely criticized on the ground that it reflects the values of the GBD framers, but is not universally acceptable. There are thus multiple DALYs: the standard DALY (0.03,1), with discounting and age weighting, DALY(0.03,0) with discounting but no age weighting, and DALY (0,0) without either. The latter has recently become the one most often used in BoD studies, making comparisons with studies using the former, “standard” DALY (0.03, 1) both confusing and difficult.

Disability weighting is probably the area of greatest controversy. A panel of experts arbitrarily assigned universal DWs between 0 (perfect health) and 1 (death) for a comprehensive (but far from exhaustive) set of non-lethal diseases/conditions, using the person trade-off (PTO) approach. DWs are however not stable – they can change in the short vs. long term, and whether the condition is treated or not. It was also soon noted that the impact of disability is highly contextual globally.[9] In addition, many feel that some diseases or groups of diseases associated with poverty (and potentially less familiar to the expert panel) are under-valued.[10] Moreover, a single summary figure is of questionable usefulness for heterogeneous categories, such as intra-cranial injuries, which have a wide array of potential outcomes.[11] Some DWs were actually inconsistent: the DW for thumb amputation was higher than that for arm

amputation, for instance! Most conditions (86% of communicable and 51% of noncommunicable diseases) had the same DW whether they were treated or not, and a few DWs actually increased with treatment (acute glomerulonephritis, for example). The 2004 GBD study includes several flaws: for instance in the Injury group, all of the 12 fractures/dislocations have the same DW with or without treatment, and hip dislocation actually has a DW lower than the one for foot fractures. [12]

Based on all these issues, the appropriateness and usefulness of the initial DWs was questioned. Arnesen and Nord (1999) concluded that there was an underlying implicit inference: that it is more important to save a healthy life than a disabled one.[9] Some, but not all, of the above concerns have been addressed by subsequent GBD updates.

The recently published GBD 2010 has tried to address these criticisms with fundamental revisions of its methodology.[13] YLDs are now calculated in their simplest form: prevalence of condition A * DW, without age weighting or discounting (DALY 0,0). The DWs have themselves been revised: a multi-country community-based survey and an open-access web-based survey have recruited over 30,000 respondents using paired comparison questions in which they had to choose the healthiest of 2 hypothetical health states. DWs thus obtained are now much more “democratic”, no longer just in the realm of expert panels. Of interest also is that, for the first time, YLD estimates are adjusted for comorbidities. Interesting findings include the top 5 causes of disability in men: low back pain, depression, falls, use of drugs and road traffic injuries (RTIs). In fact, falls and RTIs together account for 70% of all injury-related YLDs.

Unfortunately, from a functional standpoint, many of the DWs still do not make much sense: back pain without leg pain has a DW of 0.269 and neck pain 0.221, both many times higher than amputation with treatment of both arms (0.044) or both legs (0.051)! Hip dislocation, with or without treatment, has a DW of 0.017 (more or less the same as hand fracture, 0.016) compare poorly to knee dislocation, with or without treatment, at 0.129. As for spinal cord injury below the neck, it has a DW of 0.440 untreated (still better than recto-vaginal fistula, 0.492), but it dips to 0.047 if treated; severe osteoarthritis is worse (0.171) than severe mental retardation (0.157) -these are just a few examples. It appears clearly that respondents have given more weight to pain and social stigma than to loss of function, maybe in keeping with the fact that

these are “disability”, rather than “impairment”, weights. The newest GBD study will undoubtedly open itself up to a new, different set of criticisms. In addition, the Vision Loss Expert Group was quick to point out that the new disability weights for vision loss and blindness were much lower than in previous iterations, and did not pass the “common-sense test” (blindness given a barely higher DW at 0.195 than moderate skin disfigurement at 0.187, and much lower than mild alcoholism at 0.259. In response, the authors suggested that maybe it was the old weights that were wrong. They also noted that many other expert groups had made similar comments. [14] One obvious consequence is that comparing studies using the new versus old methodology will be very difficult. For example, low back pain, one of the leading causes of YLDs in the current study, is not even mentioned as a separate entity in previous GBD studies.

It remains to be seen if and how this latest iteration of the GBD study will re-define a more effective DALY. It is likely that a single summary measure will never be completely satisfactory. The main advantage of the DALY, ensuring comparability across a wide spectrum of conditions, may also be one of its greatest drawbacks: oversimplification of complex, multi-layered issues. This oversimplification is prone to mislead policy makers into the false comfort of relying on only one number. Using the GBD framework as the main -and often only – health metric to inform priority-setting and resource allocation, has been decried, even opposed, by many. [8, 9, 11, 15]

For surgeons to be able to characterize, measure, and address the growing burden of surgical disease (BoSD), a different paradigm may be necessary. Vos (2009) has recommended evaluation of surgical services rather than surgical conditions.[6] Indeed surgeons, along with other clinicians and policymakers, are concerned not only with how much of a problem there is, but also how much of it can be addressed, and with what measurable impact. If the GBD framework is to be used, as proposed by some (Duda 2008), the recommendations of Fox-Rushby and Hansen (2001) should be considered: use of relevant cohort life expectancies, presentation of DALYs both with (0.03,1) and without (0,0) discounting and age weighting, and always performing a systematic sensitivity analysis.[16, 17] As it stands, the GBD framework

focuses more on disease than on intervention, and this is less than ideal for understanding all determinants of the BoSD.

This distinction was further illustrated by a recent first estimate of the disease burden avertable through surgery.[18] Although, by the authors' own admission, the estimates were based on insufficient data, the attempt to quantify surgical burden across all disease categories highlighted methodological challenges different from those in more vertical clusters. For all the reasons above, estimating avoidable burden from a cross-cutting intervention such as surgery proved difficult within the BoD framework, and the DALY somewhat unyielding for this purpose.

Alternative approaches

In addition to DALY estimates of surgical conditions on surgical wards in LMICs, recent estimates of surgical burden have been through community surveys of operative prevalence and unmet surgical need.[19, 20] Other work has focused on specific conditions such as cleft lip and palate, and vesicovaginal fistula using organization-specific databases and multi-country studies. [21, 22] Economic estimates have also broadened to value/statistical life over costs per DALY averted, widening the spectrum of tools available to estimate the cost-effectiveness of surgical interventions. [23] For nonfatal conditions, estimates of operative backlog may be useful for advocacy and for estimation of resources required to adequately treat patients. Surgical conditions, often curable through a single intervention, are better amenable to this type of assessment, as opposed to chronic diseases requiring ongoing therapy. Another area in further need of scrutiny is the measurement of disability incurred through delays in care. As diseases go untreated, the associated disability increases – yet very few attempts have been made to date to capture this “hidden” disability.

Such non-DALY based BoD metrics may provide the basis for better assessment of surgical systems, and of the public health impact of surgical care. These approaches are likely to require greater academic debate, validation, and refinement in the foreseeable future. Nonetheless, such discussions are critical to advance the science of surgical metrics within global public health and to provide better ways of analyzing the need and the impact for improved surgical

care in resource-limited settings. Only through these efforts can the gaps in care capacity and complications witnessed by surgical providers be translated into practical data.

Conclusions

DALY-based approaches to the burden of disease provide a set of methodological challenges, some of which are unique to surgical conditions, not fully addressed through the multiple iterations of the BoD studies. Even with attempts to quantify burden through specific conditions, or through the assessment of hospital wards as platforms for service delivery, there are methodological hurdles that are difficult to overcome. The 2010 BoD study addresses some of these issues, but many questions still remain. Perhaps more importantly, other methods estimating surgical prevalence, backlog in treatment, and disability incurred by delays in care may provide more practical approaches to disease burden that can be useful tools for clinicians and health advocates. These warrant further exploration in LMICs and these debates require active engagement by surgical providers and advocates.

References

1. Murray, C.J., A.D. Lopez, and D.T. Jamison, The global burden of disease in 1990: summary results, sensitivity analysis and future directions. *Bull World Health Organ*, 1994. 72(3): p. 495-509.
2. Murray, C.J., Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bull World Health Organ*, 1994. 72(3): p. 429-45.
3. McCord, C. and Q. Chowdhury, A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet*, 2003. 81(1): p. 83-92.
4. Gosselin, R.A., A. Thind, and A. Bellardinelli, Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World J Surg*, 2006. 30(4): p. 505-11.
5. Alkire, B., et al., Potential economic benefit of cleft lip and palate repair in sub-Saharan Africa. *World J Surg*, 2011. 35(6): p. 1194-201.
6. Vos, T., Improving the quantitative basis of the surgical burden in low-income countries. *PLoS Med*, 2009. 6(9): p. e1000149.
7. Walker, D. and J. Fox-Rushby, Allowing for uncertainty in economic evaluations: qualitative sensitivity analysis. *Health Policy Plan*, 2001. 16(4): p. 435-43.
8. Mooney, G. and V. Wiseman, Burden of disease and priority setting. *Health Econ*, 2000. 9(5): p. 369-72.
9. Arnesen, T. and E. Nord, The value of DALY life: problems with ethics and validity of disability adjusted life years. *Bmj*, 1999. 319(7222): p. 1423-5.
10. King, C.H. and A.M. Bertino, Asymmetries of poverty: why global burden of disease valuations underestimate the burden of neglected tropical diseases. *PLoS Negl Trop Dis*, 2008. 2(3): p. e209.
11. Haagsma, J.A., et al., Improved and standardized method for assessing years lived with disability after injury. *Bull World Health Organ*, 2012. 90(7): p. 513-21.

12. Salomon, J.A., et al., Common values in assessing health outcomes from disease and injury: disability weights measurement study for the Global Burden of Disease Study 2010. *Lancet*, 2012. 380(9859): p. 2129-43.
13. Murray, C.J., et al., GBD 2010: design, definitions, and metrics. *Lancet*, 2012. 380(9859): p. 20636.
14. Taylor, H.R., et al., Disability weights for vision disorders in Global Burden of Disease study. *Lancet*, 2013. 381(9860): p. 23.
15. Kapiriri, L., T. Arnesen, and O.F. Norheim, Is cost-effectiveness analysis preferred to severity of disease as the main guiding principle in priority setting in resource poor settings? The case of Uganda. *Cost Eff Resour Alloc*, 2004. 2(1): p. 1.
16. Duda, R.B. and A.G. Hill, Surgery in developing countries: should surgery have a role in population-based health care? *Bull Am Coll Surg*, 2007. 92(5): p. 12-8, 35.
17. Fox-Rushby, J.A. and K. Hanson, Calculating and presenting disability adjusted life years (DALYs) in cost-effectiveness analysis. *Health Policy Plan*, 2001. 16(3): p. 326-31.
18. Debas, H., et al., Surgery, in *Disease Control Priorities in Developing Countries*, D. Jamison, Editor 2006, Oxford University Press: New York and Oxford.
19. Petroze, R.T., et al., Estimating operative disease prevalence in a low-income country: Results of a nationwide population survey in Rwanda. *Surgery*, 2012.
20. Groen, R.S., et al., Untreated surgical conditions in Sierra Leone: a cluster randomised, cross-sectional, countrywide survey. *Lancet*, 2012. 380(9847): p. 1082-7.
21. Capes, T., et al., Obstetric fistula in low and middle income countries. *Mt Sinai J Med*, 2011. 78(3): p. 352-61.
22. Poenaru, D., Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease. *World J Surg*, 2012. 23. Corlew, D.S., Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. *World J Surg*, 2010. 34(3): p. 391-6.

Chapter 3

Burden, need, or backlog: A call for improved metrics for the global burden of surgical disease

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Abstract

The global burden of disease (GBD) has been measured primarily through the use of the DALY metric. Using this approach, preliminary estimates were that 11% of the GBD is surgical. However, prior work has questioned specific aspects of the GBD methodology as well as its practicality. This paper refines other conceptual approaches based on met and unmet population need for services by considering incident and prevalent need as well as backlogs for treatment that can inform effective coverage of services. Some of these methods are tested using the example of surgical repair of cleft lip and palate. Measurement of disability incurred by delays in care may also be estimated through these approaches and has not previously been estimated through a validated model. These concepts may provide more practical information for individuals and organizations to advocate for scaling up surgical programs. While many surgical conditions are unique, as a single intervention can lead to cure, these concepts may also prove useful for non-surgical diseases. Further exploration of these approaches is merited in resource-limited settings.

Introduction: Problems with the current burden of disease model

The burden of disease (BoD) framework was developed as a population measure of ill-health. (1) The disability-adjusted life-year (DALY) was a unique innovation because it added disability to mortality in a single metric, and since its inception, has been the primary BoD metric. One of its greatest benefits has been its facilitation of comparisons across disease categories and risk factors, and disparities across regions, enabled by the use a single uniform metric.

Nonetheless, in isolation, the DALY has little meaning to most audiences, and philosophical and methodological criticisms, such as the approach to disability-weighting, have been raised. (2)The most recent BoD study has attempted to address some of these concerns. Other models of disability advocate a social approach over a medical disease-based approach, highlighting social causes and interventions to improve well-being for persons with disabilities. (3) The Katz activities of daily life index and the Washington disability index, for example, provide practical measurement tools of disability burden, while many authors have also called for a novel internationally comparable disability measure.

Surgical conditions cut across all major disease categories (infectious, non-communicable, injuries). This, among other reasons, has made it difficult to estimate surgical burden using the DALY approach compared to more “vertical” disease clusters. Surgical conditions are also unique because unlike many “medical” interventions, the treatment is often curative with a single intervention. Surgical planners are interested in measuring not only the surgical burden, but also the population impact of scaling up surgical interventions. Several studies have attempted to estimate burden averted by surgical wards in rural hospitals, but there remains a need for improved metrics. (4)

Suggestions for change

Several proposals would address the needs highlighted above.

Use “need” rather than “burden”

Although the terms “burden of surgical disease” and “surgical need” have been used interchangeably, they are not identical. While BoD refers to “what is there”, *need* implies “what is missing”. BoD was primarily intended to aggregate health state data and was not designed for subdivision beyond the level of diseases and associated risk factors. *Need*, on the other side, has already been adapted for global surgery by dividing it into met, unmet, and unmeetable need.(5)

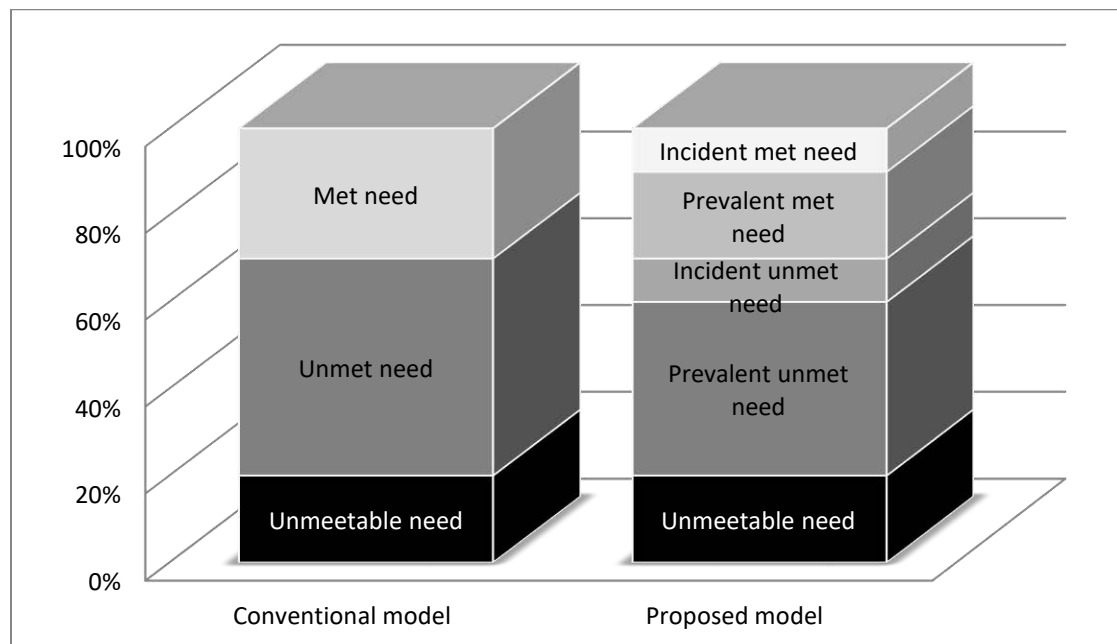
We therefore propose the term “need” to quantify surgical disease and the impact of surgical care. This parallel pathway could allow further development of metrics appropriate to global surgery, without applying concepts of BoD in ways that they were not intended.

Disaggregate BoD into met / unmet, incident and prevalent

The concept of dividing surgical need into met, unmet, and unmeetable need suggested by Bickler et al. has proven extremely useful for global surgery. For any given country, region, or surgical condition, one can theoretically estimate a *total* need (in DALYs) for surgical care, a need met at any given point in time (in “averted DALYs”) through current surgical activity, and an unmet need (in “avertable DALYs”). This is the essence of the “conventional model” depicted in

Figure 1A.

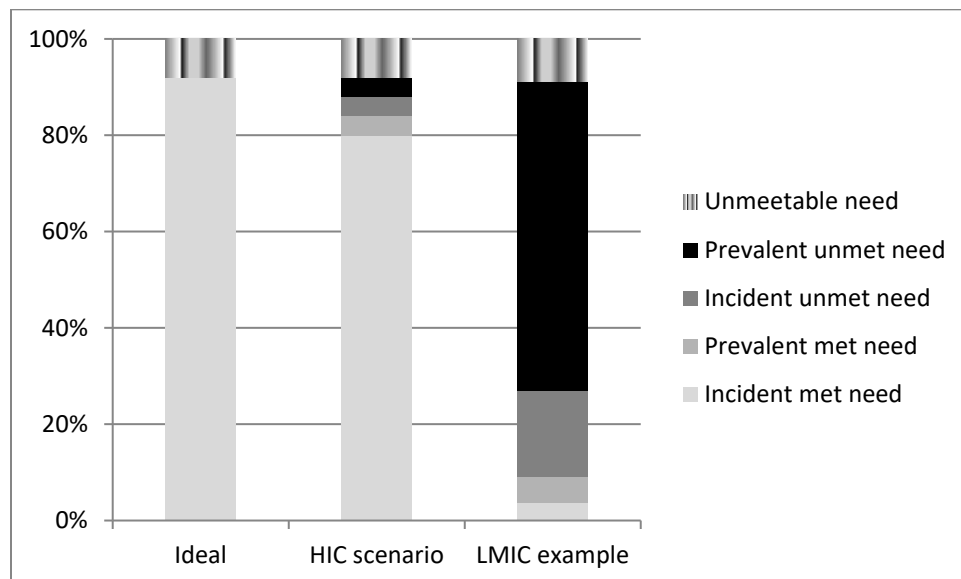
Figure 1: The conventional (A) and proposed (B) models for components of burden of disease



Furthermore, access to surgical in LMICs is frequently delayed. For fatal conditions this results in increased mortality, but for non-fatal conditions, the significant BoD caused by this delay has not been measured to date. Thus when surgical care is provided to a population, the *met* need will be partly “timely met” (or “incident met”, for new cases that are successfully treated on time), and “delayed met” (or “prevalent met”, for cases that are being treated in a delayed time frame). Delayed provision of care results in an actual averted burden (calculated in future DALYs averted), but also finalizes a “lost” unmeetable need (calculated in DALYs lost before the intervention). Similarly, in any given population the unmet surgical need will include not only the new cases emerging during a period of time (the “new / incident unmet need”), but also those who have missed being performed earlier due to ineffective coverage (the “delayed / prevalent unmet need”) (**Fig. 1B**). Appendix 1 provides a real-life example, using cleft lip and palate, and comparing metrics generated through the DALY approach with those generated through alternative metrics proposed here..

It is noteworthy that traditional summary DALY estimates include neither the backlog factor nor the lost unmeetable need. **Figure 2** depicts a hypothetical typical disaggregation of surgical need in ideal settings, in high-income countries (HICs), and in our LMIC calculation above. Besides truly unmeetable needs, only in the ideal setting is 100% of the need met promptly, at the “incident” stage. In the “real world” there will always be some delayed access resulting in needs met at the “prevalent” stage, as well as some unmet and unmeetable needs.

Figure 2: Model disaggregation of surgical need in an ideal setting, in a high-income countries (HIC) scenario, and in the current calculation for low-and-middle-income countries (LMIC)



Furthermore, the disability associated with untreated surgical disease can increase over time, and render delayed surgical intervention less effective. This reality continuously inflates the unmet need at the expense of the met need. As a result, care can be delayed to the point of becoming so difficult to provide that it becomes ineffective, futile, or even undesirable (hence “realistically unmeetable”). Examples of such futility of intervention include unrepaired cleft palate beyond childhood, late-presenting tumors, and delayed presentation of gross hydrocephalus.

Start using backlog as key metric

The concept of “backlog”—i.e. the number of individuals waiting for a specific intervention—for primarily non-fatal conditions, is a clear measure of *unmet* surgical need. This has been difficult to estimate in LMICs with any accuracy – estimates exist only for a few selected surgical conditions (**Table 1**) - but not for dozens of other chronic and debilitating conditions.(6-10)

Table 1: Estimated Global Surgical Backlog for Selected Non-Fatal Conditions, compared to HIV Care

Cataracts	50 million eyes
Trichiasis	8.2 million people
Obstetric fistula	2 million women
Cleft lip/palate	1.1 million people
HIV care	9 million people

Tabin et al (2008), Habtamu et. al (2012), Capes et. al (2008), Poenaru (2012), UNAIDS (2012)

In the absence of national or regional wait lists, the only option for backlog estimates would be extensive (and expensive) community surveys. For example, a recent survey in Sierra Leone showed that 25% of household members reported a surgical condition needing attention, while in Rwanda, a 41% lifetime prevalence of an operative condition was estimated using the same instrument.(11, 12) Piloting the use of such metrics may require prioritized measurement of a few surgical conditions for validation, as proposed recently.(13)

There is however another surrogate measure of backlog: the mean age delay between time of onset of a surgical condition and the time of corrective surgery. At a population level, each year of delayed intervention results in a new cohort of untreated patients, equal (assuming again no condition-related mortality) to the number of new cases with that condition appearing in the population. Thus a mean delay of 5 years in an intervention would generate 5 cohorts of untreated patients, as exemplified in the calculation above. In the case of congenital conditions, the cohorts equal the incidence of the condition in any given population, and the delay is the difference between the age at intervention and the ideal age of treatment. Any condition-related mortality would naturally decrease the size of successive cohorts. **Figure 3** adds change in backlog to a plot of the sample cleft palate calculation over a 10-year period.

As expected from the above assertions, provision of care in a population in excess of the yearly incidence of new cases would gradually result in a decrease in the mean age at treatment, reflecting a decrease in backlog.(9, 14, 15) Once the backlog for a specific condition is cleared in any given region, the unmet need becomes equal to the yearly incidence in the population, and all patients are treated at their ideal age.

Combine backlog with effective coverage (EC) of populations

EC is defined as the fraction of potential health gained that is actually realized for a health intervention. At the national level, the best example of the use of EC was in Mexico, where effective coverage was estimated for essential health interventions in each region.(16)

EC was explored as a surgical metric in trauma and obstetrics.(17) In obstetrics, studies of unmet need have used emergency obstetric care indicators, thus limiting their applicability to other conditions. (18) Mock et al. recently estimated that approximately 2 million deaths could be averted globally in severely injured adults with improved trauma systems, though that study did not estimate actual EC.(19)

In its basic form, $EC = \text{met need} / (\text{met} + \text{unmet need})$. Therefore, in an ideal scenario with no unmet need, this ratio would be one; in contrast, with minimal needs met, the ratio would be closer to zero. Moreover, EC integrates need, use, and quality of care for an intervention - and the aforementioned Mexican study measures of each of these dimensions with validated tools. (16)

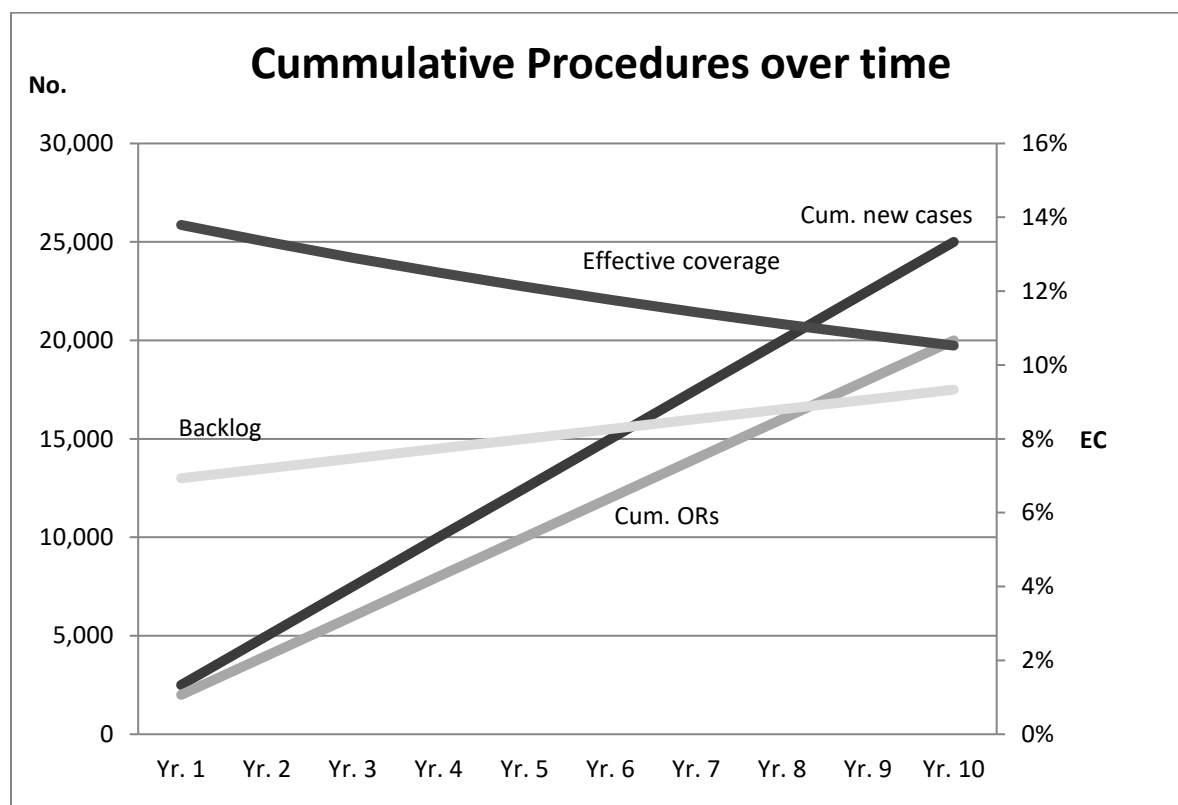
Backlog calculations, as discussed in the previous section, may provide a practical method to calculate EC for priority non-fatal surgical conditions. Backlog corresponds to unmet need, while met need can be calculated from health facilities providing surgical services. Countries and regions with large surgical backlogs would thus have low rates of EC. Regionally, such estimates could allow for improved planning and measurement of disparities both within regions and countries. The concepts of incident and prevalent met and unmet need as discussed previously could be incorporated into EC, but to our knowledge this has not been done.

Using the hypothetical model calculation above, the EC would be 80% in the absence of a backlog. A more realistic estimate is derived however by taking into account the backlog:

$$EC = \text{met need} / \text{total need} = (\text{total need} - \text{backlog}) / \text{total need} = 1 - \text{backlog} / \text{total need}.$$

This EC, actually decreasing over time in our sample calculation, is plotted in Figure 3.

Figure 3: Relationship between effective coverage, timely and delayed intervention, and backlog



ORs: surgical interventions

Limitations of the EC concept for non-fatal conditions include challenges in measuring use and quality of care, especially in settings where a majority of patients do not interact with the health care system and where outcome studies are limited. In addition, the proportion of fatal vs. non-fatal burden of surgical conditions is unknown. Interventions for emergency conditions that are not 100% effective may still avert significant burden and preserve function. On the other hand, for non-fatal disabling conditions, the effectiveness of intervention may need to be greater to avert a comparable burden. (17) This may lead planners to prioritize emergency care over the care of more prevalent surgical conditions.

Overall, however, basic data to estimate backlogs and coverage for surgical conditions should be available in most LMICs if surgical data can be collected from health facilities and for conditions where the population incidence can be reasonably estimated. This may work best initially for some conditions, such as congenital anomalies, where the population incidence may be more

constant across populations, compared to acquired surgical conditions that may exhibit greater geographic variation.

Conclusions

Current measurements of the BoD are still primarily based on the DALY, which ensured comparability through a single metric combining morbidity and mortality, but has also has some disadvantages as a sole burden metric. Disability incurred during delays in care, for example, is not captured through this model. Alternative, potentially more practical approaches to surgical burden include estimates of backlogs for prevalent non-fatal conditions requiring surgical treatment. This may also facilitate estimates of incident and prevalent met and unmet need at the population level, as well as effective coverage for surgical conditions. Further work is needed to explore the usefulness of these approaches for surgical care in LMICs. Initial approaches could be made through estimates for surgical congenital anomalies, where incidence across populations may be more constant. The metrics proposed here may also prove useful for non-surgical areas of health service delivery.

References

1. Murray C, Lopez A. The Global Burden of Disease: Volume 1. Cambridge, MA: Harvard University Press; 1996.
2. Arnesen T, Nord E. The value of DALY life: problems with ethics and validity of disability adjusted life years. *BMJ (Clinical research ed)*. 1999 Nov 27;319(7222):1423-5.
3. Palmer M, Harley D. Models and measurement in disability: an international review. *Health policy and planning*. 2012 Aug;27(5):357-64.
4. Gosselin RA, Thind A, Bellardinelli A. Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World journal of surgery*. 2006 Apr;30(4):505-11.
5. Bickler S, Ozgediz D, Gosselin R, Weiser T, Spiegel D, Hsia R, et al. Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World journal of surgery*. 2010 Mar;34(3):374-80.
6. Tabin G, Chen M, Espandar L. Cataract surgery for the developing world. *Current opinion in ophthalmology*. 2008 Jan;19(1):55-9.
7. Habtamu E, Rajak SN, Gebre T, Zerihun M, Genet A, Emerson PM, et al. Clearing the backlog: trichiasis surgeon retention and productivity in northern Ethiopia. *PLoS neglected tropical diseases*. 2011;5(4):e1014.
8. Capes T, Ascher-Walsh C, Abdoulaye I, Brodman M. Obstetric fistula in low and middle income countries. *The Mount Sinai journal of medicine, New York*. 2011 May-Jun;78(3):352-61.
9. Poenaru D. Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease. *World journal of surgery*. 2012 Dec 12.
10. UNAIDS. 2012 [updated 2012; cited 2012 October 6]; Available from: <http://www.unaids.org/en/targetsandcommitments/15millionaccessingtreatment/>.
11. Groen RS, Samai M, Stewart KA, Cassidy LD, Kamara TB, Yambasu SE, et al. Untreated surgical conditions in Sierra Leone: a cluster randomised, cross-sectional, countrywide survey. *Lancet*. 2012 Sep 22;380(9847):1082-7.

12. Petroze RT, Groen RS, Niyonkuru F, Mallory M, Ntaganda E, Joharifard S, et al. Estimating operative disease prevalence in a low-income country: Results of a nationwide population survey in Rwanda. *Surgery*. 2012 Dec 17.
13. Mock C, Cherian M, Juillard C, Donkor P, Bickler S, Jamison D, et al. Developing priorities for addressing surgical conditions globally: furthering the link between surgery and public health policy. *World journal of surgery*. 2010 Mar;34(3):381-5.
14. Magee WP, Jr., Vander Burg R, Hatcher KW. Cleft lip and palate as a cost-effective health care treatment in the developing world. *World journal of surgery*. 2010 Mar;34(3):420-7.
15. Singh SK. Smile Train: The ascendancy of cleft care in India. *Indian journal of plastic surgery : official publication of the Association of Plastic Surgeons of India*. 2009 Oct;42 Suppl:S192-8.
16. Lozano R, Soliz P, Gakidou E, Abbott-Klafter J, Feehan DM, Vidal C, et al. Benchmarking of performance of Mexican states with effective coverage. *Lancet*. 2006 Nov 11;368(9548):1729-41.
17. Ozgediz D, Hsia R, Weiser T, Gosselin R, Spiegel D, Bickler S, et al. Population health metrics for surgery: effective coverage of surgical services in low-income and middle-income countries. *World journal of surgery*. 2009 Jan;33(1):1-5.
18. Tackling unmet need for major obstetric interventions. *Unmet Obstetric Need Network; 2008* [updated 2008; cited 2012 October 6]; Available from: <http://www.uonn.org/pdf/Guide1.pdf>
19. Mock C, Joshipura M, Arreola-Risa C, Quansah R. An estimate of the number of lives that could be saved through improvements in trauma care globally. *World journal of surgery*. 2012 May;36(5):959-63.

Chapter 4

Establishing disability weights for congenital paediatric surgical conditions: a prospective, cross-sectional, multi-modal approach

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Abstract

Background

Burden of disease (BoD) as measured by Disability-Adjusted Life Years (DALYs) is one of the criteria for priority-setting in health care resource allocation. DALYs incorporate disability weights (DWs), which are currently expert-derived estimates or non-existent for most pediatric surgical conditions. The objective of this study is to establish DWs for a subset of key pediatric congenital anomalies using a range of health valuation metrics with caregivers in both high and low-resource settings.

Methods

We described 15 health states to health professionals (physicians, nurses, social workers, and therapists) and community caregivers in Kenya and Canada. The health states summaries were expert- and community-derived, consisting of a narrated description of the disease and a functional profile described in EQ-5D-5L style. DWs for each health state were elicited using four health valuation exercises (preference ranking, visual analogue scale (VAS), paired comparison (PC), and time trade-off (TTO)). The PC data were anchored internally to the TTO and externally to existing data to yield DWs for each health state on a scale from 0 (health) to 1 (dead). Any differences in DWs between the two countries were analyzed.

Findings

In total, 154 participants, matched by profession, were recruited from Kijabe, Kenya (n=78) and Hamilton, Canada (n=76). Overall calculated DWs for 15 health states ranged from 0.13 to 0.77, with little difference between countries (intra-class coefficient 0.97). However, DWs generated in Kenya for severe hypospadias and undescended testes were higher than Canadian-derived DWs ($p=0.04$ and $p<0.003$, respectively).

Interpretation

We have derived country-specific DWs for pediatric congenital anomalies using several low-cost methods and inter-professional and community caregivers. The TTO-anchored

PC method appears best suited for future use. The majority of DWs do not appear to differ significantly between the two cultural contexts and could be used to inform further work on estimating the burden of global pediatric surgical disease. Care should be taken in comparing the DWs obtained in the current study to the existent list of DWs because methodological differences may impact on their compatibility.

Background

The global health data provided through the Global Burden of Disease (GBD) study¹ using the Disability-Adjusted Life Year (DALY) metric has been a key component in the development of health policy, especially in low- and middle-income countries (LMICs). In such settings, in the absence of available primary data, GBD data have been proposed and used for broad health care initiatives, such as the Lancet Commission on Global Surgery². Within specialty areas, this necessitates a level of granularity that has not been originally intended or provided. Such is the case of pediatric surgery, where concerted efforts to improve access to care and quality of care lack data support.

In 2006, Debas conservatively estimated that 11% of the GBD was being attributed to “surgical disease”³, i.e. health conditions primarily treated through surgical intervention. More recently, Shrimpe et al placed this percentage to as high as 30%⁴, though the methodology used to derive this figure is not clear. Disproportionately carrying this surgical burden are children in LMICs, who have garnered increased attention in the global surgery community⁵. Congenital anomalies, one of the largest subsets of pediatric surgical conditions, are believed to account for 1.9% of the GBD⁶, although this is likely to be an underestimate due to the limited number of conditions studied and the difficulty associated with capturing BoD data⁷. The primary objective of this study was thus to enable the estimation of DALYs for a subset of key pediatric congenital anomalies.

The DALY is a widely used metric in LMICs developed to quantify BoD and inform global priority-setting and resource allocation^{8,9}. It encompasses both mortality and morbidity

by combining the number of years lost due to premature mortality (Years of Life Lost, YLLs) and the Years Lived with Disability (YLDs). Calculating the latter requires a disease-specific disability weight (DW), which is an empirically determined factor reflecting the health decline associated with each health state, ranging between 0 (perfect health) and 1 (death)^{7,10}. Estimation methodologies for DWs are wide-ranging and potentially contentious¹¹. All valuation methods are by definition judgmental tasks solved by participants at the moment of the exercise and results about their compatibility are mixed as best⁸. There is no reason to expect the same results across all methods, yet comparability with other DWs remains a key requirement in the GBD context to prevent methodological differences impacting the outcomes of burden of disease comparisons across countries and diseases areas. A preferred option therefore is to construct new DWs using similar estimation methods and assumptions as used in the GBD context, although this has been somewhat of a moving target.

Many different methods for DW estimation have developed over time. DWs may for instance be elicited through various psychometric exercises¹² or by trade-off methods¹³. The former include ranking exercises, magnitude estimation, visual analogue scaling (VAS), and pairwise comparison (PC) or rank ordering tasks. The latter comprise the standard gamble, time trade-off (TTO), and person trade-off (PTO)⁸ methods. The earliest DALY version appeared in a 1993 World Development Report, assigning conditions to various degrees of perceived disability¹⁴. In the second DALY version by Murray and Lopez, published in 1996 as part of the 1990 GBD study¹⁵, medical expert decisions-makers valued a subset of 22 indicator disease-oriented scenarios using the PTO method, then used the rating scale generated for the entire set of 131 conditions. The Dutch Disability Weights Project¹⁶ expanded the available weights by eliciting PTO values for another set of conditions described using the EQ-5D and an additional cognition dimension¹⁷. Subsequent modeling of those Dutch data by the Australian BoD team further expanded the set of available DWs¹⁸. In the most recent GBD update¹⁹, the methodology was significantly changed to a world-wide survey of over 30,000 household- and web-based PCs covering 220 unique health states. The

results of the PCs were then anchored on a subset of 30 health states for which population health equivalence choices were elicited through one of the four used web-based surveys. Other parallel efforts in North America include the US National Institutes of Health DALY study⁸, and the Public Health Agency of Canada's Classification and Measurement System of Functional Health (the CLAMES system)²⁰. Haagsma et al offers a comprehensive review of DW methods and studies published through 2012²¹.

The methods have clearly advanced with the scope of DW investigation. The methods used in the most recent GBD update are flexible and generate a high level of granularity by adopting the PC method while minimizing complexity of respondents' task through a limited number of complex population health equivalence choices¹⁹. Despite the above efforts, DW values for many surgical conditions, particularly within subspecialties, are missing²², thus rendering the quantification of surgical BoD challenging. Moreover, the original and subsequent GBD studies have summarized health states and their sequelae by age groups, regions and countries, rather than analyzing them by (sub)specialty. As a result, the burden of surgical conditions affecting children, especially in LMICs, has not been formally estimated, and their DWs are conspicuously missing³. In fact the 2006 extensive volume on the GBD Study only included DW values for 7 congenital surgical conditions in four disciplines, themselves pulled from the original GBD 1990 study⁷, and there were none in GBD 2010. In their absence, surgical specialty literature has used DW proxies, estimated by expert opinion using ballpark disability descriptions^{23, 24, 25, 26}.

This study intends to address the above gaps by investigating DWs for 15 congenital pediatric surgical conditions. Given the controversy surrounding the influence of cultural factors on the DW process²⁶, this study's DWs were derived in both Canada and Kenya. In developing our strategy we acknowledged the GBD Study viewpoint that achieving comparability of DWs across countries, time periods and – in our case- disease areas is of utmost importance. While the possibilities to perfectly achieve this are inherently limited because data will necessarily be collected at a different moment in time and in different resource contexts, we attempted to broaden our methodology while

maintaining as high a comparability of assumptions and methods with the original data estimates as possible.

Methods

Study Design and Participants

Data was collected for this study in Kijabe, Kenya and Hamilton, Canada between March and August 2012. Research ethics approval was obtained at both institutions (AIC Kijabe Hospital and Hamilton Integrated Research Ethics Board [11-328]) and written consent obtained from all participants. Total sample size was based on feasibility of recruitment at both centers.

Focus groups at both sites were conducted primarily in English, with Kenyan community groups in Swahili and then translated. Participants were selected based on experience with pediatric congenital anomalies (balancing experienced and non-experienced) and were recruited to match roles (i.e., physician, nurse, social worker, therapist, community participant) between the two sites. Data were collected in Kenya over 2 weeks at AIC Kijabe Hospital and in a community setting in Nairobi, and in Canada over 3 months at McMaster Children's Hospital. Each participant completed all study instruments in a single three-hour session. Focus groups were facilitated by a local research assistant and the research coordinator, and comprised 5-15 participants based on individual availability.

Health State Descriptions

We developed a set of lay descriptive handouts as suggested by Rehm and Frick⁸ for each of 15 health states (mild / severe hypospadias, undescended testis, cleft lip, cleft palate, mild / severe imperforate anus, Hirschsprung's Disease before / after colostomy, mild / severe spina bifida, mild / severe abdominal wall defect, hydrocephalus, and intestinal atresia). An example of a handout is shown in **Figure 1**. These health states were chosen based on a ranking of the most prevalent congenital pediatric surgical

conditions encountered at both sites. The handouts were circulated amongst an expert panel for face validity of the lay descriptions of functional health status and symptoms of each state; diagrams were included to improve understanding. Each handout comprised a disability profile description on 8 domains, including the five EQ-5D dimensions (mobility, self-care, usual activities, pain, mood)¹⁴, and three additional domains: “cognitive functioning”, “evacuation problems”, and “social stigma”. The three additional domains were informed by the CLAMES study¹⁷, the Dutch Disability Weights project¹³, and from our qualitative community-based focus groups with Kenyan caregivers of children with neural tube defects exploring culturally-based social stigma²⁷ as suggested by Kipiriri et al²⁸.

Figure 1: health state information example

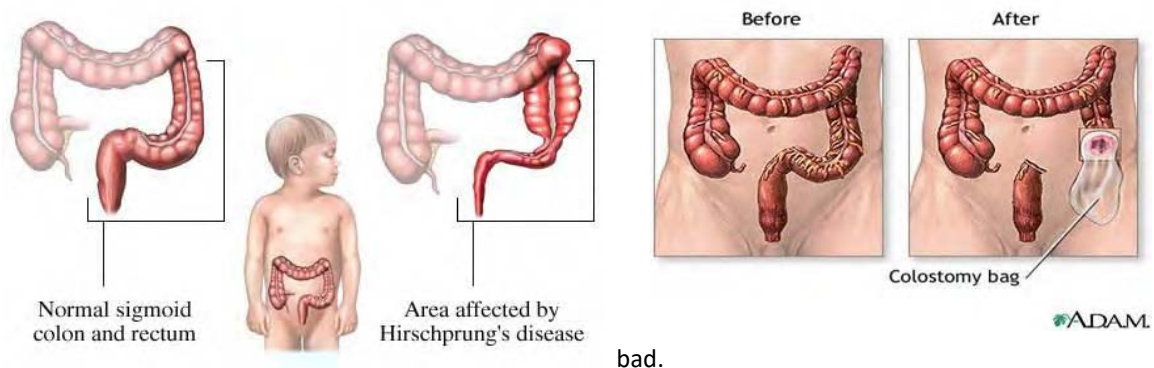
Hirschsprung's Disease 1

Background:

Hirschsprung's disease is a condition of the intestine in which the muscle cells of the bowel can't push the stool through, leading to blockage. The intestine becomes very large and may also get infected.

Children with this condition may stop having bowel movements, vomit, be very constipated, not gain weight properly, or even have severe diarrhea. These problems can happen right after birth or anytime in childhood.

The condition requires surgery, where the part of the intestine which doesn't function well is cut out. Often children also require a colostomy (an opening in their skin on their side where their stool comes out, instead of from their anus). A bag is attached (stuck) to their side around this opening to collect their stool. This bag needs to be cleaned, changed and washed regularly. Colostomies can sometimes smell



Hirschsprung's disease is divided in the following stages:

- 1) Before colostomy
- 2) After colostomy

We now ask you to value the following health state:

Children with Hirschsprung's disease who have not yet had a colostomy

Description:

- No problems with moving about
- No problems with washing and dressing self
- Some problems with performing usual activities
- Some / severe pain and discomfort
- Moderately anxious or depressed
- No / some social stigma
- No thinking problems
- Severe bowel problems (e.g. constipation, blockage, soiling, leakage)

Based on severity and surgical management, some conditions were divided into two distinct health states (e.g., Hirschsprung's before and after colostomy). All valuations applied only to the health states before definitive treatment (*untreated*) – thus a state such as “Hirschsprung's after colostomy” referred to a temporary procedure still requiring a definitive surgery. Five health states also had DWs derived by the GBD 1990 study¹³ and were used as the gold standard and were compared against our newly derived DWs.

Valuation Tasks

Standard protocols were developed for research staff training and participant explanations. Prior to data collection at each site, a pilot focus group with a representative sample was conducted to assess understanding and language for each exercise and for the lay description handouts using a series of Likert scales.

All participants completed four health valuation exercises for each health state, including Preference Ranking (PR), Visual Analogue Scales (VAS), Paired Comparisons (PC) and Time Trade-Off exercises (TTO)²⁹. Participants were asked to complete the exercises in the following order: PR, VAS, PC, TTO. The PR task was introduced to familiarize participants with the various health states and obtain an understanding of their relative severity. The VAS task was then introduced to introduce the concept of health valuation and to ensure their understanding of the health state descriptions and the purpose of the exercises. After these simpler tasks, the participants then completed the more complicated PC and TTO exercises which were used as the primary data for this study. The PC method was specifically chosen for consistency with GBD methodology. PC data, however, are generated on a latent scale, and these values need anchoring to the full health-dead scale in any of several ways. In our case, we were able to harmonize the results with the GBD scale by using the values for overlapping conditions as anchor points for the PC results. Alternatively, the TTO values might be used to identify how the PC-derived latent values relate to the full health-dead scale as

shown by Rowen et al³⁰. The use of TTO additionally enables comparison of our DW values with those published in the wider health-related quality of life (HRQoL) literature where TTO is a preferred valuation method. Having these two options was considered relevant as the feasibility and validity of the option of anchoring the new DWs to existing GBD data depends on the level of congruence across the new and existing DWs.

To complete the PR, each participant was given a set of 15 health state index cards in random order, and asked to rank each from least to most severe. Next, participants completed a VAS using a 100-point line anchored by death and perfect health with 5-point increments demarcated on the line. Participants were instructed to mark an exact point on the line for each health state in terms of severity. Additional instructions included placing similar health states closer together and vice-versa. Participants then completed a series of PCs that directly compared each health state to every other one, choosing which state was more severe. This resulted in 105 pairwise comparisons ($15 * 14 / 2$) for each participant.

In the TTO exercise participants were instructed to trade off years of healthy life for years of life lived in the specific health state, as if they were the parent of a child with the condition, and as if they were trading years of their child's life. The TTO adopted a time frame (T) of 60 years (derived from WHO standard life expectancy rates), and a smallest tradeable unit of 10 years. For example a participant could choose between living for 60 years in a particular health state or living 10, 20, 30, etc. years in perfect health. The TTO exercise was aimed at determining the number of years t in perfect health that would make the two options equally attractive (i.e. the indifference point), so that the value of a life year could be computed as t/T .

Statistical Analysis

All participants' individual responses from each valuation measure were included. DWs were calculated for each health state, by each exercise, for each country. We summarized the data from the PR task by averaging the rank order for each health

state and transforming the data to a continuous number between 0 and 1. Of note, these scores reflect how good or bad all health states are relative to the value of the best and the worst state in the set, but not relative to health states that were not included in the choice tasks - e.g. dead and full health. Therefore these scores cannot be used as DWs. For the VAS, direct measurements from the VAS scale were obtained and averaged amongst participants. In the PC exercise the proportion of the number of times each health state was chosen over its comparator was calculated for each condition, and using the normal curve, the proportions were transformed into Z-scores. The scores associated with each health state were then summed and averaged to yield an overall Z-score corresponding with the probability of a health state being chosen over all others³¹. The resulting score is a DW that is estimated on a latent scale (the resulting values are not yet anchored on the full health-dead scale). Addition of the magnitude of the most negative score and subsequent division by the highest score was applied to all values to yield a set of weights spanning a range of 0 to 1. Finally, DWs were calculated from the TTO exercise with the formula “utility = time in full health / time in disease state”.

The final analytical step involved anchoring of the PC-derived values onto the full health-dead scale. There are several ways to achieve this. Relying solely on data collected in this study, the PC scores obtained on latent scales were anchored to mean TTO values (PC-TTO) through linear regression, as suggested by Stouthard et al¹⁵ and Rowen et al³¹. Alternatively, the PC-derived values may be anchored on the full health-dead scale by using previously reported DWs, i.e. those from GBD 1990, for the 5 health states included in both data sets. The Intraclass Correlation Coefficient (ICC) assuming a one-way random model for average measures was used to analyze the agreement between the PC-derived values obtained in our study and the TTO and GBD values.

Formal quantitative data comparisons between sites were analyzed using SPSS v20.0 with a 5% significance level and Z scores computed in an Excel[®] spreadsheet. Results were presented using summative descriptive statistics with means, standard deviations,

and 95% confidence intervals where appropriate. Differences between groups were assessed using either the Fisher Exact Test or the Mann Whitney U test, depending on normalcy of the data. All DW data was first explored graphically for trends at each site, as well as descriptively between sites.

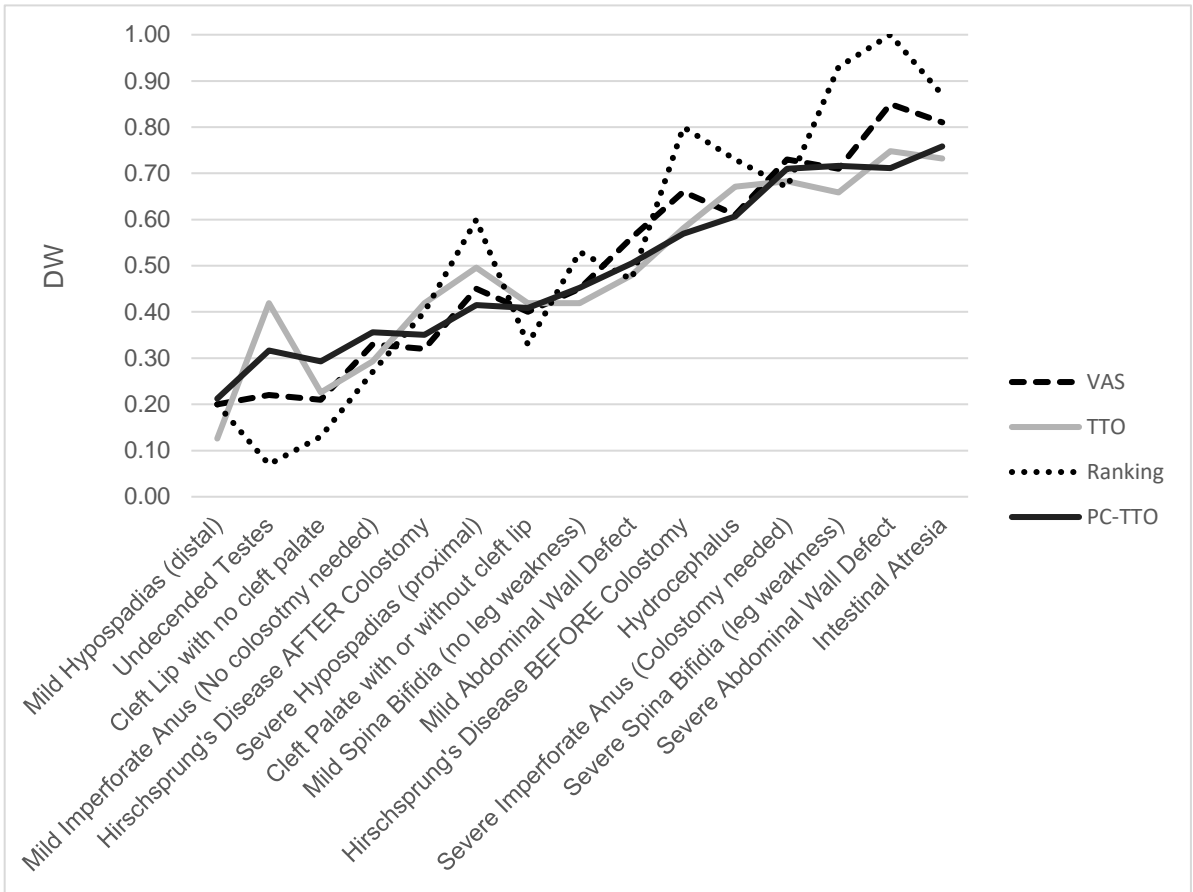
Results

In total 154 participants were recruited, 78 from Kenya and 76 from Canada (**Table 1**). DWs obtained from each of the four exercises (using internally derived PC-TTO values) is depicted in **Figures 2a and 2b** for Kenya and Canada, respectively.

Table 1: Study Participant Characteristics (n=154)

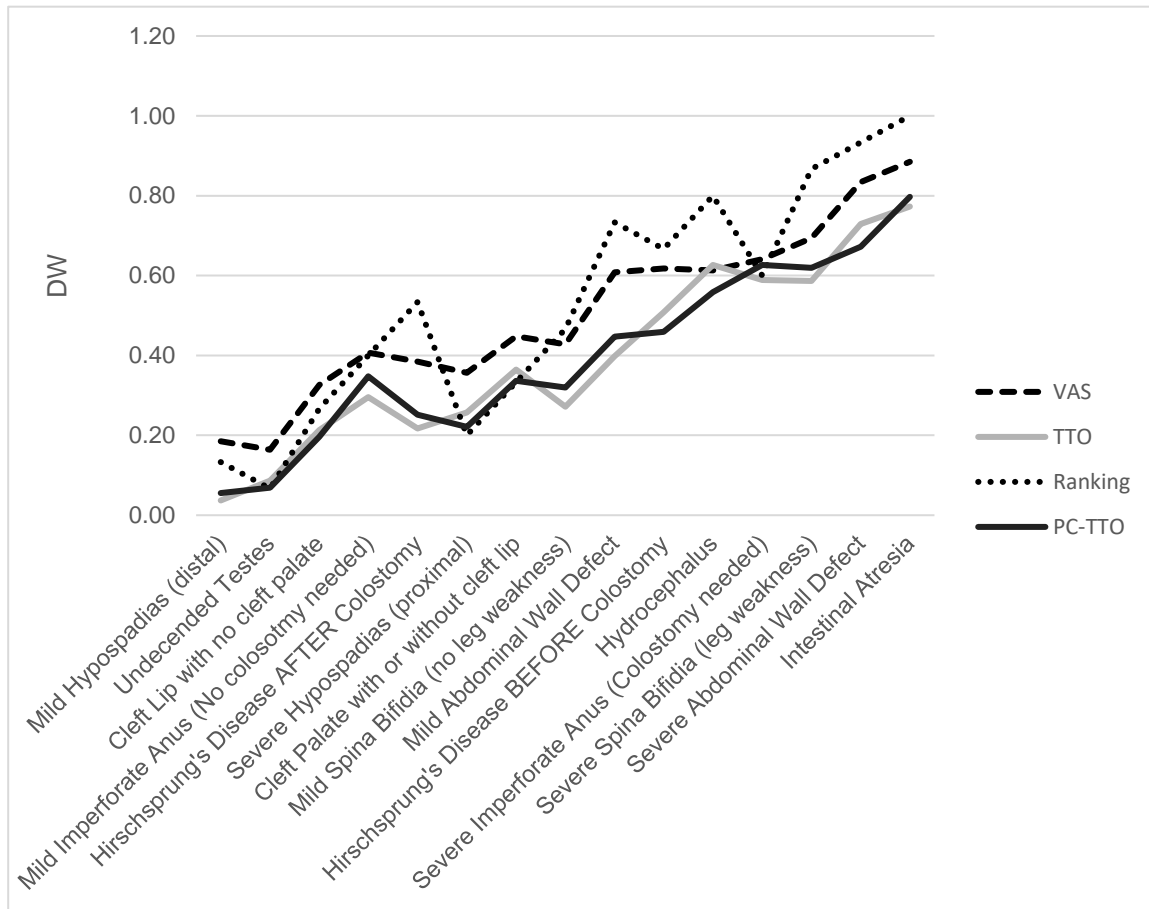
Participants	Kenya	Canada
<i>Healthcare Professionals</i>	<i>60</i>	<i>64</i>
Physicians	20	25
Nurses	35	32
Allied health professionals	5	7
<i>Community Members</i>	<i>18</i>	<i>12</i>
Total	78	76

Figure 2a. Kenyan Disability Weights per Exercise



DW = disability weight; VAS = visual analog scale; TTO = time trade-off method; PC-TTO = TTO-anchored paired comparisons method.

Figure 2b. Canadian Disability Weights per Exercise



DW = disability weight; VAS = visual analog scale; TTO = time trade-off method; PC-TTO = TTO-anchored paired comparisons method.

Tables 2a and 2b detail the DW values obtained at each site by all methods, including both internal (TTO) and external (GBD) anchoring of PC values.

Comparison of results across the 2 sites is shown using both TTO-anchored and GBD-anchored PC values in **Table 3**, and overall values obtained by the 2 anchoring methods are depicted in **Figure 3**.

Table 2a: Kenyan Disability Weights per Tool (n=78)

Health State	Ran king	VAS	TTO	PC-TTO	PC-GBD
Mild Hypospadias	0.2	0.2	0.126	0.212	0
Undescended Testes	0.07	0.22	0.419	0.317	0.115
Cleft Lip with no cleft palate	0.13	0.21	0.226	0.293	0.076
Mild Imperforate Anus	0.27	0.33	0.293	0.356	0.179
Hirschsprung's Disease AFTER Colostomy	0.4	0.32	0.419	0.351	0.170
Severe Hypospadias	0.6	0.45	0.496	0.415	0.275
Cleft Palate with/without cleft lip	0.33	0.4	0.419	0.408	0.264
Mild Spina Bifida	0.53	0.45	0.419	0.452	0.336
Mild Abdominal Wall Defect	0.47	0.56	0.479	0.505	0.421
Hirschsprung's Disease BEFORE Colostomy	0.8	0.66	0.581	0.569	0.526
Hydrocephalus	0.73	0.61	0.671	0.607	0.587
Severe Imperforate Anus	0.67	0.73	0.683	0.710	0.756
Severe Spina Bifida	0.93	0.71	0.659	0.716	0.765
Severe Abdominal Wall Defect	1	0.85	0.748	0.711	0.758
Intestinal Atresia	0.87	0.81	0.732	0.758	0.834

VAS = visual analog scale; TTO = time trade-off method; X-rank = TTO/VAS anchored ranking method; X-PC = TTO/VAS anchored paired comparisons method.

Table 2b: Canadian Disability Weights per Tool (n=78)

Health State	Ranking	VAS	TTO	PC-TTO	PC-GBD
Mild Hypospadias (distal)	0.133	0.185	0.037	0.055	0
Undescended Testes	0.067	0.164	0.087	0.069	0
Cleft Lip with no cleft palate	0.267	0.326	0.213	0.196	0.058
Mild Imperforate Anus	0.4	0.407	0.296	0.348	0.298
Hirschsprung's Disease AFTER Colostomy	0.533	0.385	0.217	0.252	0.146
Severe Hypospadias (proximal)	0.2	0.357	0.257	0.221	0.098
Cleft Palate with or without cleft lip	0.333	0.448	0.365	0.336	0.280
Mild Spina Bifida	0.467	0.428	0.272	0.320	0.254
Mild Abdominal Wall Defect	0.733	0.608	0.398	0.447	0.455
Hirschsprung's Disease BEFORE Colostomy	0.667	0.618	0.508	0.460	0.475
Hydrocephalus	0.8	0.613	0.627	0.559	0.632
Severe Imperforate Anus	0.6	0.641	0.589	0.627	0.740
Severe Spina Bifida	0.867	0.694	0.587	0.620	0.729
Severe Abdominal Wall Defect	0.933	0.834	0.729	0.672	0.812
Intestinal Atresia	1	0.885	0.773	0.797	1

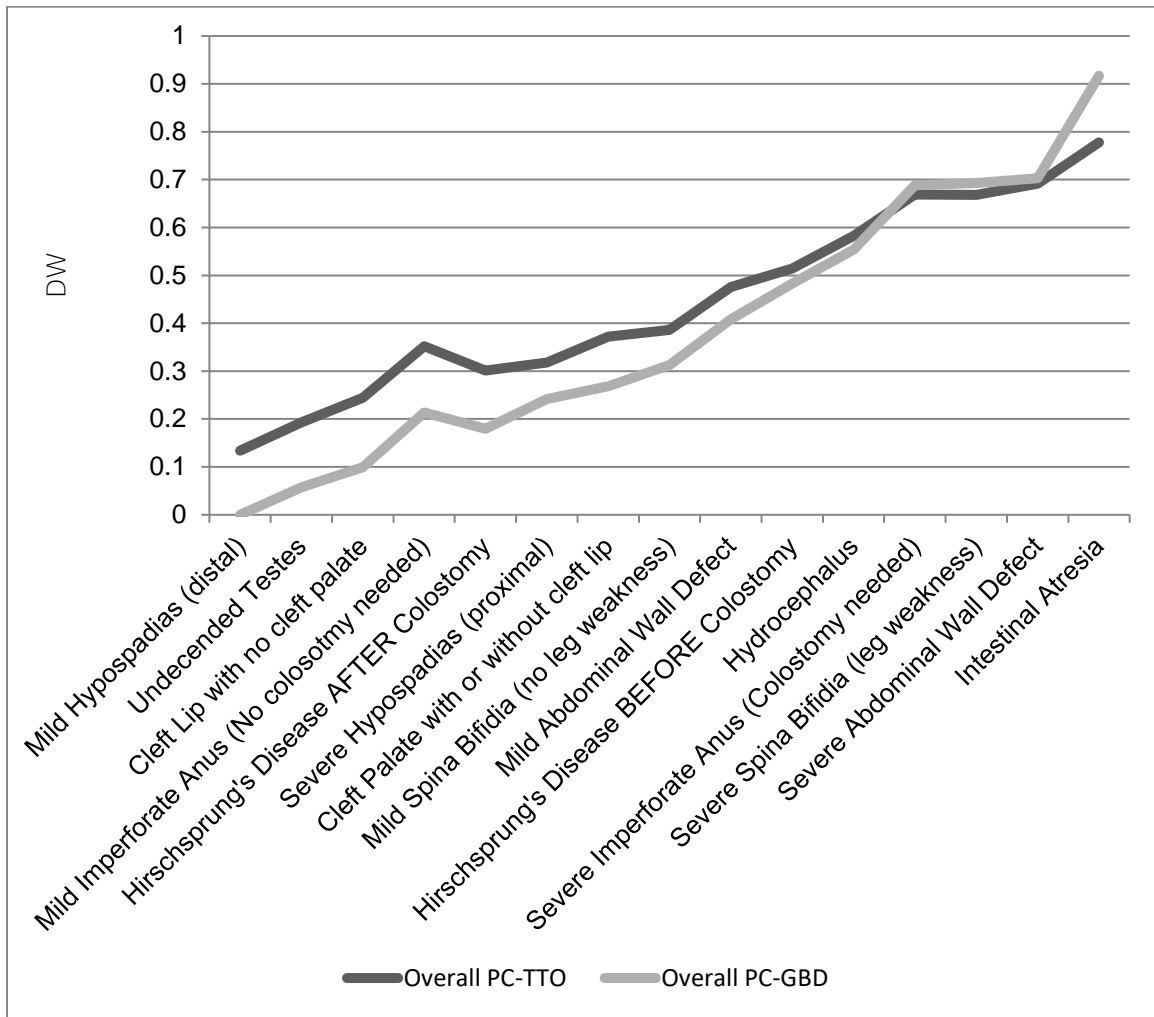
VAS = visual analog scale; TTO = time trade-off method; X-rank = TTO/VAS anchored ranking method; X-PC = TTO/VAS anchored paired comparisons method.

Table 3: Multi-National Disability Weight Comparison

Health State	KE PC-TTO	CAN PC-TTO	KE PC-GBD	CAN PC-GBD
Mild Hypospadias (distal)	0.212	0.055	0	0
Undescended Testes	0.317	0.069	0.115	0
Cleft Lip with no cleft palate	0.292	0.196	0.076	0.123
Mild Imperforate Anus	0.356	0.348	0.179	0.249
Hirschsprung's Disease AFTER Colostomy	0.351	0.252	0.170	0.189
Severe Hypospadias (proximal)	0.415	0.221	0.275	0.209
Cleft Palate with or without cleft lip	0.408	0.336	0.264	0.273
Mild Spina Bifida	0.452	0.320	0.336	0.289
Mild Abdominal Wall Defect	0.505	0.447	0.421	0.394
Hirschsprung's Disease BEFORE Colostomy	0.569	0.459	0.526	0.439
Hydrocephalus	0.607	0.559	0.5870	0.520
Severe Imperforate Anus	0.710	0.627	0.7555	0.621
Severe Spina Bifida	0.716	0.619	0.765	0.620
Severe Abdominal Wall Defect	0.711	0.672	0.758	0.648
Intestinal Atresia	0.758	0.797	0.834	1

KE = Kenya; CAN = Canada; VAS = visual analog scale; TTO = time trade-off method; X-rank = TTO/VAS anchored ranking method; X-PC = TTO/VAS anchored paired comparisons method.

Figure 3. Disability Weights by internal and external anchoring methods



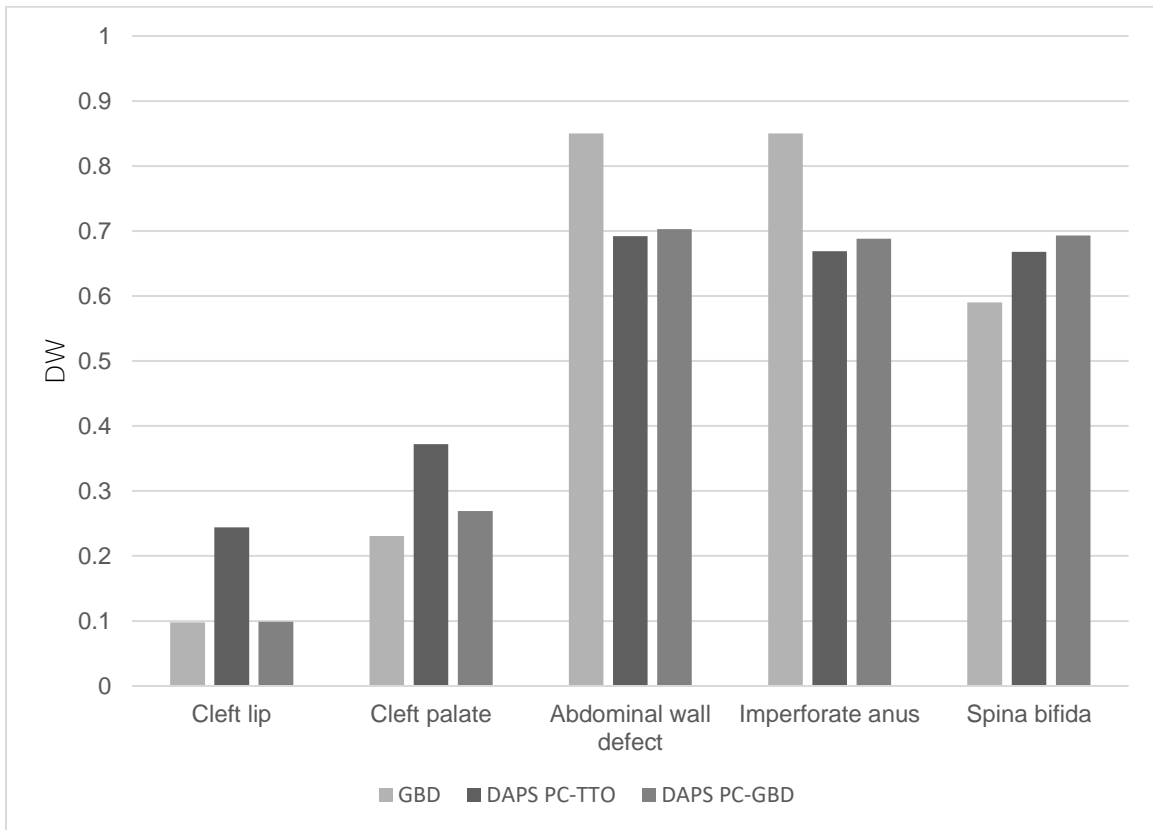
DW = disability weight; PC-TTO = TTO-anchored paired comparisons method; PC-GBD = GBD-anchored paired comparisons method.

In general, discrepancies entailed higher estimated DW values in Kenya, and for severe hypospadias and undescended testes these values were statistically significantly higher than Canadian-derived DWs ($p=0.04$ and $p<0.003$, respectively).

Disability weights for the common health states included both in our study and the GBD 1990 study are compared in **Figure 4**. While values were generally similar for several health states, discrepancies were noted particularly for cleft lip and palate, and these discrepancies were reduced when our PC values were anchored externally to the GBD study.

The ICC showed high levels of reliability between the DW data calculated for both Kenya and Canada (ICC 0.97, 95%CI: 0.93-0.99), as well as between the GBD values, TTO-anchored and GBD-anchored DW values for the 5 health conditions common to both studies (ICC=0.97; 95%CI 0.83-1.0).

Figure 4. Global Disability Weights for common conditions in GBD 1990 and current study



DW = disability weight; GBD = GBD 1990 study; DAPS = current study; PC-TTO = TTO-anchored paired comparisons method; PC-GBD = GBD-anchored paired comparisons method.

Discussion

The GBD effort over the past two decades has been instrumental in quantifying health burden, needs and factors both geographically and by broad sets of conditions, thus providing an invaluable body of information to policy-makers and healthcare professionals. The GBD project and its wide adoption by the World Health Organization, World Bank, and several national bodies^{32, 33, 34, 35} has also been essential in establishing DALYs as the preferred metric globally in disease burden measurement. While the GBD project has been extremely comprehensive, its stated global and all-inclusive purpose has resulted in limited granularity within specific medical and surgical specialty areas. In particular in LMICs, in the absence of direct population data, efforts to estimate DALYs are constrained by the available DW values, which are frequently very sparse. The current study has aimed to generate DW values for a set of congenital pediatric surgical conditions as a way to start filling that gap.

The task was successfully accomplished in both study settings. DW values generated by VAS, ranking, PC & TTO for the 15 health states spanned the full health-dead spectrum and were generally comparable. Latent scale PC values were alternatively anchored both internally to the TTO and externally to the GBD scale, generating again similar results. With a few exceptions (discussed below), inter-country results showed significant similarity, as documented by the ICC values. Internally generated PC-TTO values correlated well with GBD values for common conditions, and anchoring to these values naturally improved the correlation.

DW Values

In the absence of previous studies within the subspecialty, and faced with a wide choice of valuation methods available, each with its own benefits and shortcomings, the authors chose to start with four different methods, both psychometric and econometric, and compare the results obtained by these broad inputs for each health state. This strategy produced a large number of data points without over-burdening the participants, and allowed inter-method comparisons as well as both *a priori* and *post-*

hoc suggestions for preferred methods. Yet, we also faced the complex question of how to deal with potential discrepancies across the methods. Variation in DW estimates could result from participants' different health states interpretation, their risk aversion and time preference, but also to differences in valuations between exercises for the same health state, and overall distributional concerns such as VAS distortion³⁶.

An anticipated strength of the chosen set of methods was its ability to generate different types of data: PC values being derived on a latent scale they can complement other methods while avoiding potential conflicts of scale when other methods are paired (e.g. VAS and TTO). This strategy is increasingly popular^{31,36,37}. The adoption of PC in the recent version of the GBD Study also strongly mitigates in favor of its use on the latent scale. Pooling of values obtained across the other methods has to the best of our knowledge not been done – instead a choice for either one is made based on pros and cons of each. Similarly, while rank data could be used to provide values on a latent scale like PC, the latter is favored for its greater reliability, with rank data often used just as a “warm-up” exercise³⁸. Nevertheless presence of values derived from multiple methods remains beneficial, allows at least to assess convergence across methods and demonstrate validity. Against that background we were pleased with the high level of agreement, as shown in the high ICC values, that was achieved across the methods.

The DW values generated for the 15 health states across the two sites were generally similar based on high ICC values, leading support to the assertion that DWs are stable cross-nationally and cross-culturally^{39,16}. Two notable exceptions to this purported DW stability were encountered in the current study. Severe (i.e. proximal) hypospadias and undescended testes were assigned higher DW values in Kenya. This discrepancy may be explained culturally: both health states include the possibility of infertility in their descriptions, a state associated with significant stigma in many non-Western cultures^{40,41}.

Limited possibilities exist for external validation of the DW estimates generated in this study. The GBD 1990 study, already used in our study for external anchoring, included

DWs for 7 congenital surgical conditions (cleft lip, cleft palate, abdominal wall defects, imperforate anus, cardiac defects, esophageal atresia, and spina bifida)⁷, and later DW studies globally did not expand this list. Moreover, only the cleft lip and palate states include both untreated and treated values, a significant limitation to the use of other published DW values in surgical arenas. Within the limitation of slightly different methodologies used, the current study had the dual opportunity of using the DW values of the common health states for both external validation (of PC-TTO values) and external anchoring (as in PC-GBD values). We consider the PC-TTO as our primary “take-home” results as they are internally derived and not dependent on overlapping health states with other studies. Moreover, the 2 methods generally generated similar DW values, well within the same order of magnitude. Of note however, cleft lip and palate received however significantly lower values in the GBD study. This may be due to disability from cleft lip/palate being artificially limited to the first 5 years of life in the GBD study, a constraint not reflecting the reality of older children living with this untreated condition in LMICs⁴².

Comparison to GBD 2010 Study and Advantages

With the recent publication of the GBD 2010 study, any parallel attempt at deriving DWs must be justifiable, valid, and comparable. The primary justification for the current study is simply the necessity to obtain a wider set of DW values within a given specialty, for the purpose of generating relevant specialty-specific BoD data that can inform policy decision making in this area. Yet in order to offer valid inter-specialty comparisons, the methodology of such parallel studies must be sufficiently similar to that of the GBD gold standard. Without the benefit of the latest iteration of the GBD study at the time of study design, and using a much smaller study sample, the authors chose a panel of valuation methods which allowed the comparison of commonly used methods in the literature. In light of the current results, the use of paired comparisons appears justified and probably sufficient, in conjunction with a method of anchoring the results to the health-dead scale. This process resembles the GBD Study in its use of PCs, though differing from it in the anchoring method. Other strengths of the current study include

standardized and explicit health state descriptions, and input from both health care workers and families familiar with the conditions investigated. But caveats remain, such as the different approach to describing the health states, and uncertainty whether the same health state DW values will be obtained in PCs if more or less health states are included in the experiment. The checklist for any such future efforts must include clear, consistent health state descriptions and a single psychometric valuation such as PC, anchored firmly to the disability scale. Moreover, studying health states spanning a wide range of severity would facilitate robust data generation.

The main limitation of the study pertains to the underlying concept of the DALY and of the disability weighting which it requires. In the first place, it is extremely difficult to harmonize universal DW values with the widely divergent sociocultural and economic contexts where they are derived⁴³. Furthermore, there are multiple controversial value decisions in the computations of DALYs which can significantly impact the ultimate BoD conclusions drawn from them^{44,45}, as well as limitations inherent within the specific health valuation exercises themselves. Finally, DALYs seem to underestimate several specialty areas, such as neglected tropical diseases⁴⁶ and surgical conditions¹⁸.

Conclusions and Applications

The current study has successfully generated a set of DW values for pediatric congenital anomalies, making these values available for all necessary future studies⁴⁷. The process involved in generating such a limited DW set was relatively straightforward and inexpensive, and, within the confines of the above limitations, the results were robust and comparable to those generated by large global studies. The DWs do not appear to differ significantly across divergent socio-cultural contexts and can be used to calculate both the met and the unmet burden of global pediatric surgical disease⁴⁸.

While the extensive global and national BoD studies will remain the basis for global policy decisions, the study suggests that DW sets may be expanded and refined within a surgical specialty. While waiting for future studies to show whether other specialties may be equally successful in the process, a cautious, well-guided advance is

recommended in this novel field in order to ultimately generate practical knowledge in global health.

Acknowledgment

The results of this study have been presented in the form of a poster at the Global Health Metrics and Evaluation meeting in Seattle, USA, under the title “Establishing Disability Weights for Congenital Pediatric Surgical Disease: A cross-sectional, multi-modal study”.

References

- ¹ GBD 2015 Disease and Injury Incidence and Prevalence Collaborators. Global, regional, and national incidence, prevalence, and years lived with disability for 301 acute and chronic diseases and injuries in 188 countries, 1990–2013: a systematic analysis for the Global Burden of Disease Study 2013. *Lancet* 2015;386:743–800.
- ² Meara JG, Leather AJM, Hagander L., Alkire BC, Alonso N, Ameh E, et al. Global Surgery 2030: evidence and solutions for achieving health, welfare, and economic development. *Lancet* 2015;6736:569–624.
- ³ Debas H et al. Surgery. In: Jamison D, ed. *Disease control priorities in developing countries*. 2nd edn. New York: Oxford University Press; 2006.
- ⁴ Shrimpe MG, Bickler SW, Alkire BC, Mock C. Global burden of surgical disease: an estimation from the provider perspective. *Lancet Glob Health* 2015;3 Suppl S8–9.
- ⁵ Ozgediz D, Poenaru D. The burden of pediatric surgical conditions in low and middle income countries: A call to action. *Journal of Pediatric Surgery* 2012;47:2305–2311.
- ⁶ World Health Organization (WHO). *Global Estimates 2014 Summary Tables: DALY by Cause, Age, and Sex for 2000-2012*. World Health Organization Jun 2014. Geneva, Switzerland. http://www.who.int/healthinfo/global_burden_disease/en/. Accessed 1 July 2015.
- ⁷ Debas H et al. *Disease Control Priorities, Third Ed. Volume 1. Essential Surgery*. World Bank. 2015 Washington, DC. <https://openknowledge.worldbank.org/handle/10986/21568>. Accessed 1 July 2015.
- ⁸ Murray CJL. Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bulletin of the World Health Organization* 1994;72:429-445.

⁹ Lopez AD, Mathers CD, Ezzati M, Jamison DT, Murray CJL, eds. Global Burden of Disease and Risk Factors. Disease Control Priorities Project. Washington (DC):World Bank; 2006.

¹⁰ Rehm, J, Frick, U. Valuation of health states in the US study to establish disability weights: lessons from the literature. *Int J Methods Psych Res* 2010;19:18–33.

¹¹ Mont D. Measuring health and disability. *Lancet* 2007;369:1658–1663.

¹² Revicki D, Kaplan R. Relationship between psychometric and utility-based approaches to the measurement of health-related quality of life. *Qual Life Res* 1993;2:477–487.

¹³ Dolan P. The measurement of health-related quality of life for use in resource allocation decisions in health care. In Culyer A, Newhouse J, editors. *Handbook of Health Economics*. Elsevier; 2000. p. 1724–1760.

¹⁴ World Bank. *World development report 1993; investing in health*. Oxford: Oxford University Press; 1993.

¹⁵ Murray CJL, Lopez AD (1996) A comprehensive assessment of mortality and disability from disease, injuries and risk factors in 1990 and projected to 2020. In: *Global burden of disease*. Cambridge: Harvard University Press; 1996.

¹⁶ Stouthard M, Essink-Bot ML, Bonsel GJ. *Disability Weights for Diseases in the Netherlands*. Erasmus University Rotterdam; 1997.

¹⁷ Brooks R, EuroQoL Group. EuroQoL: The current state of play. *Health Policy* 1996;37:53–72.

¹⁸ Mathers CD, Vos T, Stevenson C. *The burden of disease and injury in Australia Summary report*. Canberra, Australia; 1999.

-
- ¹⁹ Salomon J, Vos T, Hogan D R, Gagnon M, Naghavi M, Mokdad A, et al. Common values in assessing health outcomes from disease and injury: disability weights measurement study for the Global Burden of Disease Study 2010. *Lancet* 2012;380:2129–43.
- ²⁰ McIntosh CN, Connor Gorber S, Bernier J, Berthelot JM. Eliciting Canadian population preferences for health states using the Classification and Measurement System of Functional Health (CLAMES). *Chronic Dis Canada* 2007;28:29–41.
- ²¹ Haagsma JA, Polinder S, Cassini A, Colzani E, Havelaar AH. Review of disability weight studies: comparison of methodological choices and values. *Pop Health Metrics* 2014;12:1.
- ²² Gosselin R, Ozgediz D, Poenaru D. A Square Peg in a Round Hole? Challenges with DALY-based “Burden of Disease” Calculations in Surgery and a Call for Alternative Metrics. *World J Surg* 2013;37:2507-2511.
- ²³ McCord C, Chowdhury Q. A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obs* 2003;81:83–92.
- ²⁴ Gosselin R, Thind A, Bellardinelli A. Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World J Surg* 2006;30:505–511.
- ²⁵ Gosselin R, Maldonado A, Elder G. Comparative cost- effectiveness analysis of two MSF surgical trauma centers. *World J Surg* 2010;34:415–419.
- ²⁶ Poenaru, D. (2013). Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease. *World J Surg* 2013;37:1562–1570.
- ²⁷ Frankfurter C, Pemberton J, Cameron BH, Poenaru D. Understanding the burden of surgical congenital anomalies in Kenya: an international mixed-methods approach.

Poster at the Consortium of Universities for Global Health (CUGH). Washington, DC.
2014-05-20

²⁸ Kapiriri L, Frithjof Norheim O. Whose priorities count? Comparison of community-identified health problems and Burden-of-Disease-assessed health priorities in a district in Uganda. *Health Expect* 2002;5:55–62.

²⁹ Brazier J. *Measuring and valuing health benefits for economic evaluation*. Oxford:Oxford University Press; 2007.

³⁰ Rowen D, Brazier J, Van Hout B. A Comparison of Methods for Converting DCE Values onto the Full Health-Dead QALY Scale. *Med Decis Making* 2015;35:328–40.

³¹ Streiner DL, Norman GR. *Health measurement scales: a practical guide to their development and use*. Oxford: Oxford University Press; 2008.

³² Stouthard MEA, Essink-Bot ML, Bonsle GJ et al. Disability weights for diseases. *Europ J Public Health* 2000;10:24–30.

³³ Essink-Bot ML, Pereira J, Packer C, Schwarzingler M, Burstrom K. Cross-national comparability of burden of disease estimates: the European Disability Weights Project. *Bull World Health Organ* 2002;80:644–52.

³⁴ McKenna MT, Michaud CM, Murray CJ, Marks JS. Assessing the burden of disease in the United States using disability-adjusted life years. *Am J Prev Med* 2005;28:415–23.

³⁵ Yoon SJ, Bae SC, Lee SJ, et al. Measuring the burden of disease in Korea. *J Korean Med Sci* 2007;22:518–23.

³⁶ Salomon, JA, Murray CJL. Estimating health state valuations using a multiple-method protocol. In *Summary Measures of Population Health Concepts, Ethics, Measurement and Applications*. Geneva: World Health Organization; 2002. p. 487–499.

-
- ³⁷ Feng Y, Devlin N, Shah K, Mulhern B, van Hout B. New methods for modelling EQ-5D-5L value sets: an application to English data. *Health Economics & Decision Science (HEDS) Discussion Paper Series*, Jan. 2016.
- ³⁸ Louviere JJ, Hensher DA, Swait JD. *Stated Choice Methods: Analysis and Applications*. Cambridge: Cambridge University Press; 2000.
- ³⁹ Schwarzingler M, Stouthard MEA, Burström K. Cross-national agreement on disability weights : the European Disability Weights Project. *Pop Health Metrics* 2003;1:1–11.
- ⁴⁰ Dyer, Silke J. The value of children in African countries-Insights from studies on infertility. *J Psychosomatic Obs Gynecol* 2007;28:69-77.
- ⁴¹ Inhorn MC. "The Worms Are Weak" Male Infertility and Patriarchal Paradoxes in Egypt." *Men Masculinities* 2003;5:236-256.
- ⁴² Magee WP, Vander Burg R, Hatcher KW. Cleft lip and palate as a cost-effective health care treatment in the developing world. *World J Surg* 2010;34:420–7.
- ⁴³ Reidpath DD. Measuring health in a vacuum: examining the disability weight of the DALY. *Health Policy Planning* 2003;18:351–356.
- ⁴⁴ Anand S, Hanson K. Disability-adjusted life years: a critical review. *J Health Econ* 1997;16:685–702.
- ⁴⁵ Arnesen T, Kipiriri L. Can the value choices in DALYs influence global priority-setting? *Health Policy* 2004;70:137–49.
- ⁴⁶ King CH, Bertino AM. Asymmetries of poverty: why global burden of disease valuations underestimate the burden of neglected tropical diseases. *PLoS Neglected Tropical Dis* 2008;2:e209.
- ⁴⁷ Poenaru D, Pemberton J, Frankfurter C, Cameron BH. Quantifying the Disability Averted through Pediatric Surgery: a cross-sectional comparison of a pediatric surgical unit in Kenya and Canada. *World J Surg* 2015;39:2198-2206.

⁴⁸ Bickler S, Ozgediz D, Gosselin R, Weiser T, Spiegel D, et al. Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg* 2010;34:374–80.

Chapter 5

Burden of Surgical Congenital Anomalies in Kenya: A Population-Based Study

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Abstract

Background

Efforts to estimate the burden of pediatric surgical disease in Africa are limited by the absence of population-based data.

Methods

Community volunteers randomly surveyed households at sites across Kenya. Caretakers were asked to identify on a photographic portfolio several visible congenital malformations present among the children in their household. Disability-adjusted life years (DALYs) were calculated based on life expectancy tables and published and estimated disability weights for the conditions encountered.

Results

Caregivers of 5,559 children (54% female) were surveyed in 1,909 households. Overall prevalence of congenital malformations was 6.3 / 1000, amounting to 54 - 120 DALYs per 1000 depending on the life tables used. The most prevalent condition in the survey was club foot, while spina bifida had the highest burden of disease.

Discussion

This study is the first to document the prevalence of selected surgical congenital malformations among children in Kenya and the burden of disease associated with them.

Introduction

Congenital malformations are a significant cause of morbidity and mortality, especially in low- and middle-income countries (LMICs). Debas *et al* [1] estimate that congenital malformations account for 9% of the surgical burden of disease (BoD), contributing to the disability experienced by 150 million children around the world, with disabilities being more common among children in LMICs [2]. These disabilities represent a burden placed on child development and family responsibilities, and are both the cause and the result of poor socioeconomic status [2,3].

The burden of childhood surgical disease from congenital malformations is significant in sub-Saharan African settings. In Gambia, such conditions represented the second highest proportion of children presenting for surgical care, preceded only by injuries [4,5]. Surgical procedures to address congenital malformations comprised 40% of all operations performed at a major teaching hospital in northern Nigeria [6], and nearly 1 in 4 children presenting at a major sub-Saharan hospital suffered from congenital malformations [4,5]. In studies that focused on specific malformations and/or geographic regions, the incidence of congenital malformations in LMICs was calculated to be between 3.9 and 11.8 per 1,000 live births [7,8]. However, in such studies congenital malformations are likely to be underreported due to stigmatization or fear associated with being rejected by the community [9,10].

Based on the premise that about half of all congenital malformations are surgical [1], an obvious method of decreasing the burden of childhood disease is the prevention and early treatment of neonatal surgical conditions, many of which result in disability or death when left untreated [11]. Surgical care can have a significant impact on the health burden of congenital malformations, which account for 3% of pediatric deaths and 24 million disability-adjusted life years (DALYs) worldwide [12]. DALYs are a widely accepted metric of disease burden, as they take into account both the years of life lost and the reduced quality of life resulting from disability.

There is limited data on the extent and impact of pediatric disabilities in LMICs. In response to the increased recognition of disabilities as a major global health concern, a comprehensive review of three large databases was undertaken to identify publications focused on pediatric intellectual, hearing, speech, vision, motor, and neurological disabilities in LMICs [9]. The reviewers noted a paucity of information on most pediatric disabilities with the exception of intellectual and hearing impairments, and few LMICs have epidemiological information on pediatric disabilities in their region. The Kenya National Survey for Persons with Disabilities recently estimated a 2.4% overall prevalence of disability among Kenyan children aged 0 to 14 years [13]. There is a need however for more detailed data on specific disabilities in order to identify the most effective health care interventions for alleviating the morbidity and mortality of pediatric congenital disease. Moreover, quantification of the disease burden of congenital malformations potentially avertable by surgery allows for comparisons with other health interventions competing for limited resources. We are not aware of any population-based study quantifying the burden of surgically treatable congenital malformations in an African country. The objective of this study therefore was to estimate the prevalence and burden of selected surgical congenital malformations among children in Kenya.

Methods

Design

The study is a population-based prevalence survey. It entailed field workers surveying households and looking for family members 16 years of age or younger who have any of a set of 8 visible congenital malformations. The field workers were recruited by liaising with the local community health office at each designated site. The workers were already volunteering at, or employed by, the health office and were known in the community and familiar with visible disabilities. They were trained on site by one of 3 study coordinators, who would then follow them on their initial home visits to ensure adequate performance. The study sample consisted of caretakers – parents and other guardians – from a sample of 10 urban and rural locations in

Kenya. These locations were selected to match the population distribution in Kenya by province.

Within each location, interviews were conducted by the field worker in a random selection of households, using a method of the World Health Organization Expanded Programme on Immunization as described by Durkin *et al* [14]. According to this method, in each study community the field worker began in a central location (such as the community's local government health center or chief's office), and walked in one of four possible directions (north, south, east, west). The field worker, through a random process, then selected a household to interview which was

- 1) along the left or right side of the road (by throwing a dice, with 1 to 3 = left, 4 to 6 = right); and
- 2) between 1 and 6 houses away from his/her starting location (e.g. for a 4, go to the 4th house on the side of the road that was decided in step 1).

A household was defined as a family, including its caretakers/servants, who shared a single front door within a residential property that borders the side of the road. The field worker continued in the direction of travel using the above process of sampling until the community boundary was reached. This process was then repeated in the 3 other remaining directions. In the event that the direction of travel was no longer possible (such as at a T-junction), the field worker used a random process to decide a new direction to travel (left or right) until the earliest opportunity to resume the original direction. In the event of no children living in the selected household, or if nobody was at home, the next closest household was approached.

A standard portfolio of photographs of congenital malformations was provided to each community worker as a screening tool to help caretakers identify the conditions of interest (cleft lip, club foot, spina bifida / encephalocele, hydrocephalus, hypospadias, bladder exstrophy, and imperforated anus). These conditions were chosen because they are easily identifiable, visible deformities for which surgical treatment is available in the country. Caretakers from each interviewed household were presented with the screening tool and

shown additional pictures if there was confusion in the identification of the condition. Caretakers were also asked to provide additional information, including education level, household income and their child(ren)'s place of birth¹. Similar door-to-door interview techniques, along with the use of photographic albums, have been previously used to determine the community-based prevalence rate of pediatric disabilities [7,15].

Sample size

The overall prevalence of pediatric disabilities in community-based studies in LMICs varies between 0.4% - 12.7% [3,15,16,17,18,19,20], and the prevalence of neurological and motor disabilities among children in LMICs varies between 0.2% - 6.1% and 0.2% - 2.8%, respectively [17,17,18,18,21,21,22]. Using a point estimate of 4% as the prevalence of all the disabilities that we seek to identify in our study, and applying standard statistical formulae for calculating 95% confidence intervals from the binomial distribution, we estimated that a survey of 5,000 children would provide 95% confidence intervals of 3.5 - 4.6%. We therefore intended to survey an average of 500 children within each of the 10 communities sampled. Assuming an average household size of 5.3 [23], we anticipated on average interviewing the caretakers of 3 children per household, 167 households per community, and 1,670 households in total. The sample size calculation was performed using a standard online statistical calculator [24].

DALY Calculation

Burden of disease (BoD) estimates were calculated using previously established methods [25] and universal and country-specific life tables provided by the GBD Study [26] and the World Health Organization [27]. DALYs are calculated as the sum of

¹ The survey and the photographic portfolio are available for download from <http://dl.dropbox.com/u/9254603/KIDS%20study/KIDS%20survey.docx> and <http://dl.dropbox.com/u/9254603/KIDS%20study/KIDS%20photo%20portfolio.pdf> , respectively.

life years lost due to premature mortality (YLL) and years lived with disability (YLD), where YLD is the product of the life expectancy at disease onset and the disability weight (DW). In light of the fact that the current study only attempted to identify prevalent conditions among living children and that the study assumed that these conditions do not affect mortality rates in these children, the DALY calculation included only the YLD. As the GBD Study provides DWs for only a few selected pediatric surgical conditions, DWs for the remainder was estimated using the scale endorsed by the World Health Organization [28] and shown in **Table 1**.

Table 1: Disability weights used in disability-adjusted life year (DALY) calculations [28]

Description	Weight
Limited ability to perform at least one activity in one of the following areas: recreation, education, procreation or occupation	0.1
Limited ability to perform most activities in one of the following areas: recreation, education, procreation or occupation	0.2
Limited ability to perform activities in two or more of the following areas: recreation, education, procreation or occupation	0.4
Limited ability to perform most activities in all of the following areas: recreation, education, procreation, or occupation	0.6
Needs assistance with instrumental activities of daily living such as meal preparation, shopping or housework	0.8
Needs assistance with activities of daily living such as eating, personal hygiene or toilet use	0.9

Age weighting and discounting [28,29,30] are common practices in GBD studies, but they remain somewhat controversial, especially when applied to children [29,30,31]. The authors therefore chose to do a sensitivity analysis on life expectancy tables, using 3 different tables: Kenya standard uniform, undiscounted tables (so-called YLL(0,0)), Kenya standard age-weighted, 3% future discounted tables (YLL(3,1)), and universal standard undiscounted tables [28].

Statistical analysis

Descriptive statistics were applied to the demographic variables and DALYs, using Microsoft Excel® software.

Ethics

This study was approved by the Research Ethics Committee of AIC Kijabe Hospital in Kijabe, Kenya. Before entering each locale, the community chiefs were first approached for written consent to undertake this study in their jurisdiction. Verbal consent was also obtained from the household caretakers before proceeding with the interviews.

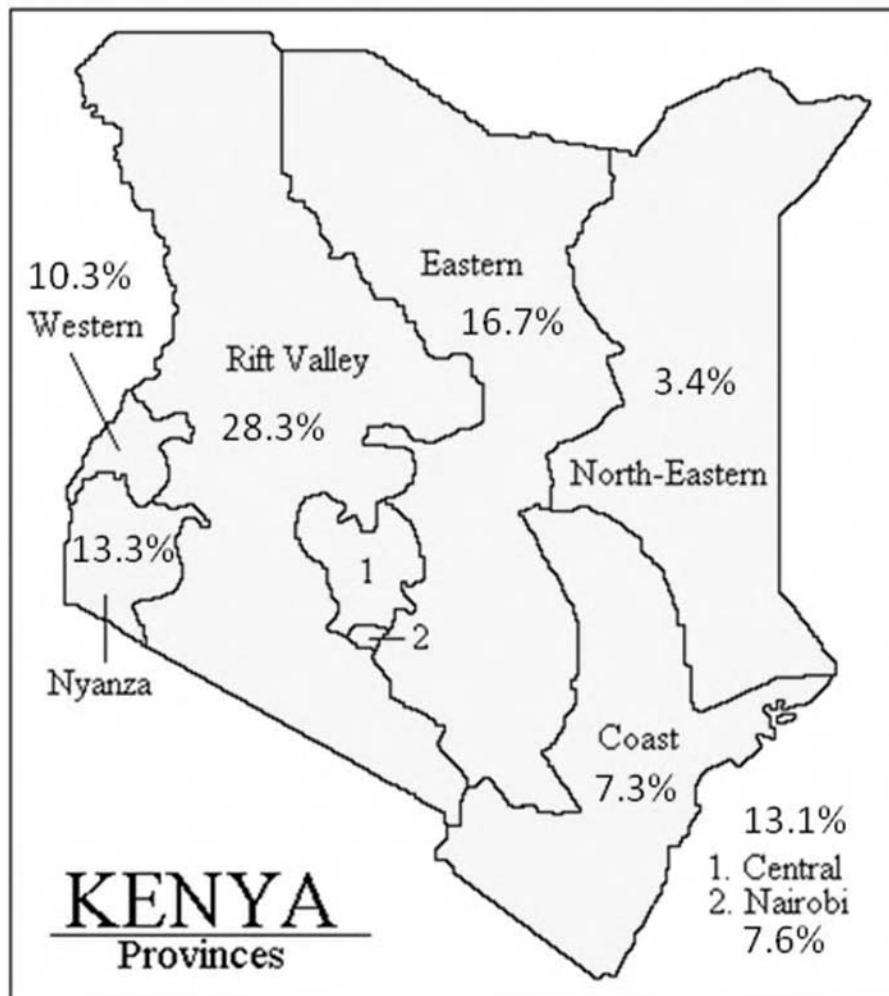
Results

Between July 2009 and March 2010 the caregivers of a total of 5,559 children were surveyed in 1,909 households. All 8 provinces were sampled at or above their intended target levels. There were no non-responders in the study: of all caretakers that were invited to participate in the study, none refused to participate. The distribution of respondents by location is shown in **Table 2**. **Figure 1** depicts the location of the homesteads surveyed - most children resided in the population-rich Rift Valley and Eastern provinces. Fifty-five percent of the households were in rural areas, and there were 2.9 eligible children on average per interviewed household.

Table 2: Distribution of households and children interviewed, by province Percentage is in parenthesis.

	Rift Valley	Eastern	Nyanza	Western	Central	Coast	Nairobi	North Eastern	Total
Homesteads	540 (28.3)	319 (16.7)	254 (13.3)	196 (10.3)	250 (13.1)	140 (7.3)	145 (7.6)	65 (3.4)	1909 (100.0)
Children	2114 (38.0)	787 (14.2)	747 (13.4)	451 (8.1)	620 (11.2)	336 (6.0)	253 (4.6)	251 (4.5)	5559 (100.0)

Figure 1: Location of surveyed homesteads by province



Additional demographic characteristics of the caretakers are presented in **Table 3**. The highest education level among the majority of caretakers was primary school or no formal education at all, with fathers more likely to attain higher education. Almost half of the households (45%) included children who were born in the home rather than at a clinic or hospital. The median household income was 5,000 KSh / month (range, 0 to 600,000) or approximately US\$65 / month. These demographics are fairly consonant with published socio-economic studies for Kenya [32].

Table 3: Demographic characteristics of caretakers interviewed

	Number	Percent
Household location		
Urban	232	12.2
Suburban	634	33.2
Rural	1043	54.6
Child(ren)'s place of birth		
Home	454	23.8
Local clinic	963	50.4
Both	411	21.5
No children / Declined to answer	81	4.2
Income distribution		
	Average monthly income per household (KES)	Percent share of wealth
Poorest 20%	1,101	2.9
Second 20%	3,036	8.0
Middle 20%	4,770	12.5
Fourth 20%	7,449	19.5
Highest 20%	21,775	57.1
Declined to answer: 562 households		
Education level		
	Mother (%)	Father (%)
Did not attend school	233 (12.2)	183 (9.6)
Primary	869 (45.5)	631 (33.1)
Secondary	546 (28.6)	608 (31.8)
College	166 (8.7)	201 (10.5)
University	39 (2.0)	69 (3.6)
Not applicable / Declined to answer	56 (2.9)	217 (11.4)

There were 35 malformations encountered in the 8 selected conditions sought by the current study, 19 males and 16 females (Table 4). Forty-nine percent of the children identified were under 5 years of age, 31% were between 5 and 10 years, and 20% were over 10 years old.

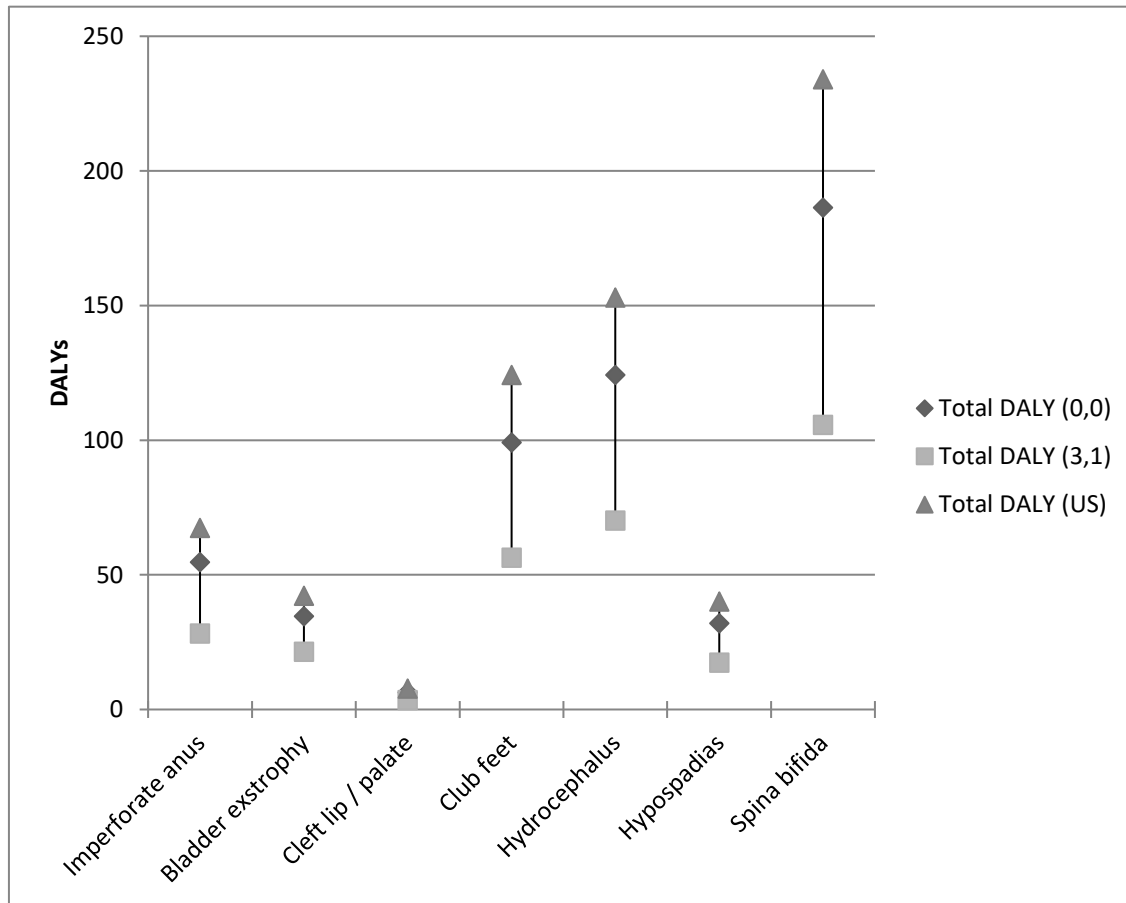
The population prevalence of these malformations and associated disability weights is shown in **Table 4**. The overall rate of congenital malformations was 6.3 per 1,000 children. Club foot was the most prevalent malformation encountered (2.9 per 1,000 children) both in males and females .

Table 4: Disability weight, number and prevalence of congenital malformations

Condition	Disability Weight	Number	Prevalence (per 1,000)	
			Total	95% CI
Bladder exstrophy	0.6	1	0.2	0.0-1.0
Cleft lip	0.05	2	0.4	0.0-1.3
Club foot	0.1	16	2.9	1.6-4.7
Encephalocele	0.6	2	0.4	0.0-3.7
Hydrocephalus	0.4	5	0.9	0.3-2.1
Hypospadias	0.1	5	0.9	0.3-2.1
Imperforate anus	0.85	1	0.2	0.0-1.0
Spina bifida	0.6	3	0.5	0.1-1.6
TOTAL		35	6.3	4.4-8.7

Figure 2 depicts the spread of estimated DALYs from each malformation by 3 different calculation methods. Spina bifida carried the highest BoD by all methods (106-234 DALYs in the sample), while cleft lip the lowest (3.5-7.7 DALYs). The lowest estimate of total burden of surgical disease in the sample was 303 DALYs (54 DALYs / 1,000 children) when age-weighted, discounted Kenya YLL values were used, and 669 DALYs (120 DALYs / 1,000 children) when using universal, unweighted and undiscounted YLL values. The standard deviation of the DALYs per 1,000 estimated by all methods was 29.

Figure 2: Sensitivity analysis for burden of disease (in DALYs) from selected congenital malformations

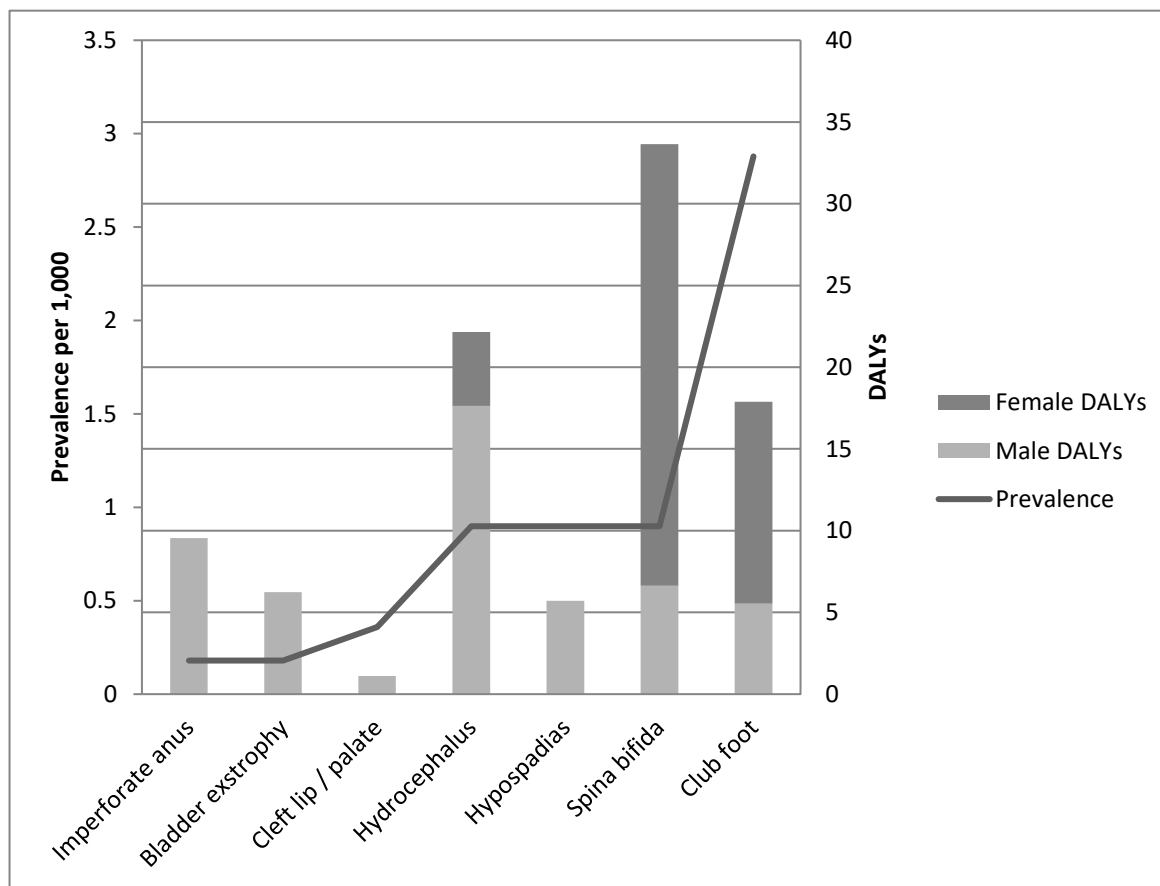


DALY (0,0): DALYs using standard undiscounted Kenya life expectancy table
 DALY (3,1): DALYs using age-weighted and 3% discounted Kenya life expectancy table
 DALY (US): DALYs using universal standard undiscounted life expectancy table

Figure 3 compares the prevalence of the selected malformations with the mean BoD (as calculated by the 3 methods above), by gender, associated with them – both per 1,000 children. While the prevalence is plotted in increasing order, the corresponding BoD varies widely in order.

Spina bifida resulted in the greatest BoD, with rates of 19 - 42 DALYs per 1,000 children. When stratified for gender, hydrocephalus represented the greatest BoD among males (10 - 20.7 DALYs per 1,000 children) and spina bifida for females (15.1 - 32.2 DALYs per 1,000 children).

Figure 3: Prevalence and mean burden of disease from selected congenital malformations, by gender



Discussion

To our knowledge, this is the first population-based study to estimate the prevalence and burden of congenital surgical malformations in Kenya. Population-based estimates of congenital malformations and pediatric disabilities in LMICs are scarce.

One reason for the scarcity of data is the challenge of reliably selecting a random, non-biased sample in LMIC populations. Many similar surveys are based on hospital-based birth data [33], which naturally miss the significant percentage of births in LMICs which occur outside health care facilities. Moreover, surveys are often done in urban areas, because of the challenges of rural surveying. Address registries, village maps, and telephone directories were not available for any of the communities targeted, which led us to the sampling method described above.

This simple method allowed us to quickly and inexpensively train community workers to administer the survey. Moreover, the use of photographic portfolios in interviews has been shown to be an effective way of helping respondents identify various congenital malformations [7,15] - especially important in resource-poor settings where illiteracy may be an issue.

The significance of the study lies in its ability to derive population-based data for common visible congenital anomalies. BoD metrics hold advantages over traditional ones such as prevalence, as demonstrated by Figure 2. Depending on the DW of each condition and the age of each child, some conditions (e.g., imperforate anus, spina bifida and hydrocephalus) carry a disproportionately higher BoD than implied by their prevalence - while the opposite is true for less disabling conditions such as club foot, cleft lip and hypospadias.

The nature of the survey questions resulted in prevalence rates for these conditions, which may differ from their corresponding incidence rates. While incidence rates are more generally accepted as the metric in epidemiological studies of congenital conditions, prevalence estimates are needed for BoD calculations [14,15,16,16]. Prevalence directly allows the estimation of surgical backlog in any area, as well as unmet need or avertable DALYs [25].

Although comparisons with other studies should be made with caution, the prevalence rates obtained in this study seem to be lower than in other published reports [7,8,34]. This may be in part due to the survey method, as detailed below. Moreover, the study generated prevalence, rather than incidence, rates. In situations when the congenital malformation carries a low mortality (e.g. club foot, hypospadias or cleft lip), the prevalence rate should be similar to the yearly incidence rate. For instance club foot was estimated in our study at 2.9 per 1,000, twice the rate recently found recently in the Uganda Sustainable Clubfoot Care Project [33]. Given however that most malformations still carry with them some risk of death, our results would therefore tend to underestimate the true incidence of the congenital malformations of interest. This approach offers therefore *minimum* prevalence and BoD estimates for the population in question.

The DALY calculations were significantly dependent on the life expectancy tables used - although all values were reassuringly well within the same order of magnitude. The age-weighted life tables slightly increased the DALY values, while future discounting decreased the values by roughly 50%. As discussed earlier, the choice of age weighting and discounting is debatable, carrying a fair amount of social value judgment and therefore uncertainty [35]. In the absence of a clear consensus, sensitivity analysis at least presents a spectrum of plausible results [35,36]. The use of universal rather than country-specific life tables is also not standardized. While the majority of surgical studies simply followed the universal tables introduced in the original GBD Study [28], the availability of country-specific tables allows us and others to be more specific in the calculations [37].

The BoD associated with congenital malformations is very significant by comparison to adult surgical or medical conditions. This is due to a unique mix of features of congenital malformations: young age, typically infant (thus maximum number of years potentially to be lived with a disability); often severe nature (thus associated with either premature death or high DW); stable, chronic condition (unlike trauma or other surgical emergencies, in the absence of treatment the disability simply persists for decades); and frequently some measure of residual life-long disability even after successful treatment (for instance impaired mobility in spina bifida, speech problems in cleft palate, or incontinence in imperforate anus). The accurate measurement of the BoD associated with congenital malformations can therefore play a significant role in the efforts to document the importance of surgical intervention for congenital malformations. In as much as the GBD study set out “to disentangle epidemiology from advocacy in order to produce objective, independent and demographically plausible assessments of the burdens of particular conditions and diseases” [35], such objective assessments as can be generated must be put to use for the sake of improved health care for those in need [38].

Ultimately, an area-wide survey of congenital malformations achieves much more than help derive some prevalence and BoD values. As Pirani *et al* found out in their country-wide Ugandan study, their survey raised awareness of congenital malformations within the

population, and provided a unique opportunity for mass education on congenital disabilities and their treatment [33].

Limitations

There are several limitations to this study. In the first place, we based our sample size calculation on a point estimate that is greater than the true prevalence calculated at the end of the study. Upon data analysis, this produced confidence intervals that were skewed around their point estimates. Such values should be interpreted with some caution, as they do not precisely mirror the binomial distribution that the sample size was based upon.

Secondly, the results of this study may be underestimated due to the caretakers' difficulty in disclosing their children's health status. In our setting the sampling requirements and literacy considerations resulted in the natural use of community workers for interviewing caretakers. The use of interviewer-administered surveys however for potentially sensitive topics may lead to less truthful information gathered when compared to other methods such as paper-based surveys [39]. In fact, we noticed caretakers commonly responding that they have seen children in their community who had conditions mentioned in the portfolio. This may indicate parents' willingness to participate in the survey, but an unwillingness to disclose when the child with the malformation in question was theirs [9,10].

Thirdly, the photographic portfolio was very useful for most, but not all, conditions surveyed. In the case of genitourinary and anorectal conditions, cultural sensitivity required that photographs be only used if requested by the respondent, which may have led to some inaccuracy in the responses.

Finally, the disability weights used in this study are simple estimates using the rather coarse WHO tool. The fact is that the GBD Study has not produced many disability weights within surgery, especially pediatric surgery. Currently there are published DW values for only 6 pediatric surgical conditions (abdominal wall defects, anorectal atresia, cleft lip and palate, esophageal atresia, congenital heart anomalies, and spina bifida)[26]. It is obvious that an accurate measurement of the burden of pediatric surgical disease will require the formal

establishment of disability weights associated with a large variety of pediatric surgical conditions.

Future considerations

This study provided an important first step in establishing prevalence rates and disease burden for surgically correctible congenital malformations in Kenya, a topic that has received little previous attention. The project reinforced the feasibility of low-scale population-based BoD studies in resource-limited settings. Future studies may include surveys assessing both the incidence and prevalence of congenital malformations, as well as larger sample sizes. Further studies also need to cover a broader range of both congenital and acquired malformations — this study focused on relatively common, externally visible congenital anomalies — in order to provide a more comprehensive scope of the burden of congenital disease and disability that can potentially be treated by surgery.

The recognition of surgery as an effective intervention in LMICs has been hindered by the perception that it is a non-cost-effective luxury [40]. In recent years however surgery has gained attention as a cost-effective way of addressing the global BoD [11]. Similarly to the existing cost effectiveness analysis (CEA) studies in general and trauma surgery [41,42] in LMICs, CEA studies in pediatric surgery are essential tools in the global advocacy effort towards appropriate resource allocation for the surgical treatment and long-term rehabilitation of children with congenital malformations.

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REFERENCES

1. Debas H, Gosselin R, McCord C, et al Chapter 67 – Surgery (2006) In: Disease Control Priorities in Developing Countries, p. 1245-1259
2. Disability - a Global Picture (2011) In: World Report on Disability. World Health Organization, Geneva, p. 19-46
3. Natale JE, Joseph JG, Bergen R, et al (1992) Prevalence of Childhood Disability in a Southern Indian City: Independent Effect of Small Differences in Social Status. *Int J Epidemiol* 21:367-372
4. Bickler SW, Sanno-Duanda B (2000) Epidemiology of paediatric surgical admissions to a government referral hospital in the Gambia. *Bull World Health Organ* 78:1330-1336
5. Bickler SW, Telfer, ML, Sanno-Duanda B (2003) Need for paediatric surgery care in an urban area of The Gambia. *Trop Doct* 33:91-94
6. Ameh EA, Chirdan LB (2001) Paediatric surgery in the rural setting: prospect and feasibility. *West Afr J Med* 201:52-55
7. Cherian A, Seena S, Bullock RK, et al (2005) Incidence of neural tube defects in the least-developed area of India: a population-based study. *Lancet* 366:930-931
8. Delpont SD, Christianson AL, van den Berg HJS, et al (1999) Congenital anomalies in black South African liveborn neonates at an urban academic hospital. *S Afr Med J* 85:11-15
9. Maulik PK, Darmstadt GL (2007) Childhood Disability in Low- and Middle-Income Countries: Overview of Screening, prevention, services, Legislation, and Epidemiology. *Pediatrics* 120:S1-S55
10. Kirpalani HM, Parkin PC, Willan AR, et al (2000) Quality of life in spina bifida: importance of parental hope. *Arch Dis Child* 83:293-297

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11. Azzie G, Bickler S, Farmer D, et al (2008) Partnerships for developing pediatric surgical care in low-income countries. *J Pediatr Surg* 43:2273-2274
 12. World Health Organization (2009) Mortality and burden of disease estimates for WHO member states in 2004. http://www.who.int/healthinfo/global_burden_disease/estimates_country/en/index.html
Accessed January 12, 2012
 13. Kenya National Survey for Persons with Disabilities - Preliminary Report (2008) National Coordinating Agency for Population and Development, Nairobi
 14. Durkin MS, Hasan ZM, Hasan KZ (1998) Prevalence and correlates of mental retardation among children in Karachi, Pakistan. *Am J Epidemiol* 147:281-288
 15. Sauvey S, Osrin D, Manandhar, DS, et al (2005) Prevalence of Childhood and Adolescent Disabilities in Rural Nepal. *Indian Pediatr* 42:697-702
 16. Al-Ansari A (1989) Prevalence estimates of physical disability in Bahrain: a household survey. *Int Disabil Stud* 11:21-24
 17. Biritwum RB, Debres JP, Ofosu-Amaah S, et al (2001) Prevalence of children with disabilities in Central Region, Ghana. *West Afr J Med* 20:249-255
 18. Paul TJ, Desai P, Thomburn MJ (1992) The prevalence of childhood disability and related medical diagnosis in Clarendon, Jamaica. *West Indian Med J* 23:8-11
 19. Couper J (2002) Prevalence of childhood disability in rural Kwazulu-Natal. *S Afr Med J* 92:549-552
 20. Chen J, Simeonsson RJ (1993). Prevention of childhood disability in the People's Republic of China. *Child Care Health Dev* 19:71-88

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21. Mung'ala-Odera V, Meehan R, Njuguna P, et al (2006) Prevalence and risk factors of neurological disability and impairment in children living in rural Kenya. *Int J Epidemiol* 35:683-688
 22. Thornburn M, Desai P, Paul TJ, et al (1992) Identification of childhood disability in Jamaica: the ten question screen. *Int J Rehabil Res* 15:115-127
 23. Bongaarts J (2001) Household size and composition in the developing world in the 1990s. *Population Studies* 55:263-279
 - 24 . Power, Sample Size and Experimental Design Calculations. <http://www.statpages.org/#Power>. Accessed January 24, 2012
 25. Bickler S, Ozgediz D, Gosselin R, et al (2010) Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg*, 34:374-380
 26. Lopez AD, Mathers CD, Ezzati M, et al eds. (2006) *Global Burden of Disease and Risk Factors*. Oxford University Press & The World Bank, New York
 - 27 . World Health Organization National Tools: Standard Life Table. <http://www.who.int/entity/healthinfo/bodreferencestandardlifetable.xls>. Accessed January 23, 2012
 28. Murray CJ (1994) Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bull World Health Organ* 72:429-445
 29. Keren R, Pati S, Feudtner C (2004) The generation gap: differences between children and adults pertinent to economic evaluations of health interventions. *Pharmacoeconomics* 22:71-81
 30. Anand S, Hanson K (1997) Disability-adjusted life years: a critical review. *J Health Economics* 16:685-702
 31. Corlew DS (2010) Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. *World J Surg* 34:391-396

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32. Suri T, Tschirley D, Irungu C, Gitau R, Kariuki D (2009) Rural incomes, inequality and poverty dynamics in Kenya. Tegemeo Institute of Agricultural Policy and Development, Egerton University. WPS 30/2008, p. 12
 33. Pirani S, Naddumba E, Mathias R, et al. Towards effective Ponseti clubfoot care: the Uganda Sustainable Clubfoot Care Project (2009) *Clin Orthopaed Rel Res* 467:1154-1163
 34. Venter PA, Christianson AL, Hutamo CM, et al (1995) Congenital anomalies in rural black South African neonates--a silent epidemic? *S Afr Med J* 85:15-20.
 35. Salomon JA, Mathers CD, Murray CJL, et al (2001) Methods for life expectancy and healthy life expectancy uncertainty analysis. Global programme on evidence for health policy working paper 10, p. 1-18
 36. The global burden of disease: a comprehensive assessment of mortality and disability from diseases, injuries and risk factors in 1990 and projected to 2020 (1996) In: Murray CJL, Lopez, AD (eds.) *Global Burden of Disease and Injury Series, Vol. 1*. Harvard University Press, Cambridge, p. 1-51
 37. Stein CE, Birmingham M, Kurian M, et al (2003) The global burden of measles in the year 2000--a model that uses country-specific indicators. *J Infectious Dis* 187:S8-14
 38. Lopez AD (2005) The evolution of the Global Burden of Disease framework for disease, injury and risk factor quantification: developing the evidence base for national, regional and global public health action. *Globalization and health* 1:5
 39. MacMillan HL, Wathen CN, Jamieson E, et al (2006) Approaches to screening for intimate partner violence in health care settings: a randomized trial. *JAMA* 296:530-536
 40. Farmer P, Kim J (2008) Surgery and global health: a view from beyond the OR. *World J Surg* 32:533-536
 41. Gosselin R, Thind A, Bellardinelli A (2006) Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World J Surg* 30:505-511

42. Gosselin R, Maldonado A, Elder G (2010) Comparative cost-effectiveness analysis of two MSF surgical trauma centers. *World J Surg* 34:415-419

Chapter 6

Revised from:

Burden of Surgically Correctable Disabilities Among Children in the Dadaab Refugee Camp

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Abstract

Background

Surgery is increasingly recognized as a means to reduce the morbidity and mortality of disabling impairments in resource-limited environments. We sought to estimate the burden of surgically correctable disabling impairments and the cost-effectiveness of their treatment among children in a large refugee camp.

Methods

This is a chart review of all patients aged 0 to 18 years from Dadaab Refugee Camp (Kenya) treated at a facility primarily responsible for providing pediatric surgical care in the region. Total disability-adjusted life years (DALYs) averted were calculated using life expectancy tables and established or estimated disability weights. A sensitivity analysis was performed using various life expectancy tables. Delayed averted DALYs caused by delay in care were also estimated. Inpatient costs were collected to perform a cost-effectiveness analysis.

Results

Between 2005 and 2011 a total of 640 procedures were performed on 341 patients. The median age at surgery was 4.6 years, and 33% of children treated were female. Only 13.5% of surgeries estimated as required for common congenital surgical conditions were actually performed. The total number of DALYs averted was 4,136 – 9,529 (6.4 -14.8 per patient), depending on the calculation method used. Cost-effectiveness analysis resulted in values of \$40 - \$88 per DALY.

Conclusions

The burden of pediatric surgical disabling impairments in refugee camps is substantial. Surgical intervention to address this burden is both feasible and cost-effective. Such intervention can significantly decrease the burden of disability among children affected by armed conflicts.

Introduction

Childhood disability is a significant cause of morbidity, especially in low- and middle-income countries (LMICs). The World Health Organization (WHO) estimates that 150 million children suffer from some type of disability globally, the majority of whom reside in LMICs with limited access to medical care [1]. Multiple studies have shown that children with functional disabilities have substantially decreased quality of life (QOL) when compared to non-disabled children [2,3,4]. Some of this decrease in QOL in LMICs results from the marginalization and stigma, both in the home and in the community [5,6]. Evidence exists that childhood disability is a major risk factor for limitations in daily activities into adulthood [7].

The Dadaab Refugee Camp

Shortly after the outbreak of the civil war in Somalia, the Dadaab camp was opened in 1992 in North-Eastern Kenya. Originally built for less than 90,000 inhabitants, it is currently home to over 460,000 predominantly Somali refugees, with others originating in other unstable neighboring countries [8,9]. Health care in the camp is provided by several humanitarian organizations under the umbrella of the United Nations High Commission for Refugees (UNHCR). As a result of the ongoing conflict in the horn of Africa, the already limited health care resources in the camp are continuously stretched by increasing numbers of refugees (typically over 1,000 new arrivals daily). Disabilities among the refugees are frequent and significant [10], due to the larger proportion of vulnerable persons seeking refuge, as well as the extremely limited health care provision for the past 20 years in Somalia. This results in a significant backlog of care, with patients of all ages frequently presenting with untreated chronic surgical conditions and disabilities, both congenital and acquired.

Burden of surgical disease

Surgery plays an important role in reducing the burden of disabilities in LMICs. Congenital abnormalities represent an important cause of pediatric disability, and approximately half of such abnormalities require surgical intervention [11]. Moreover, in LMICs trauma is frequently the commonest cause for children's surgery [12], and when injuries are left untreated or are improperly treated, chronic disability often ensues [7,13]. Finally, it is estimated that only 3.5%

of all surgical procedures are performed in the poorest third of the world, despite a burden of disease (BoD) greater than in the rest of the world [14].

Disability adjusted life years (DALYs), a health gap measure combining the morbidity and mortality associated with a disease, have become a standard metric to quantify BoD [15]. DALYs often highlight diseases with a chronic component: while only 3% of all pediatric deaths are attributable to congenital malformations, these account for a staggering 24 million DALYs worldwide [15]. Within global surgery, BoD has been divided into *met need* (surgical care already provided, or DALYs averted), *unmet need* (potentially avoidable/treatable disability and death due to surgical conditions, or DALYs avertable), and *unmeetable need* (disability and premature death that is unavoidable, even with the best surgical care) [16,17].

DALYs lend themselves to cost-effectiveness analysis (CEA), by estimating the cost required to offset one DALY through any given health care intervention. Interestingly, surgical treatment has been shown to cost not more – and in fact often less – in \$/DALY than proven medical interventions such as immunizations and anti-retroviral therapy [18,19,20].

While health care services to displaced people have been emphasized by the global health community [21,22], surgical services are rarely mentioned. Moreover, the BoD stemming from conflict-related population migration has never been estimated to our knowledge, neither globally nor locally. The objective of this study is to calculate the burden of surgically averted disease among children in the world's largest refugee camp, and estimate the cost-effectiveness of surgical intervention in this group. Such information, especially when extended to populations, will be important for resources advocacy in the care of these vulnerable groups.

Methods

We reviewed the hospital records of all patients aged 0 to 18 years residing in the Dadaab Refugee Camp and treated at BethanyKids at Kijabe Hospital, the primary pediatric surgical provider for the camp. *BethanyKids* is an international faith-based organization dedicated to the holistic care of children with surgical conditions. The *BethanyKids* unit in Kijabe, Kenya has 65 beds, 2 operating rooms, and a full surgical and rehabilitation staff. Based on a long-lasting

relationship with the UNHCR, *BethanyKids* surgeons have visited the refugee camp for patient screening and follow-up every 2 months since 2004.

Patient records were abstracted for gender, birth date, diagnosis, and surgical interventions; outpatient records were excluded. Mean age of refugee patients undergoing common congenital procedures was compared to that of non-refugee patients in the *BethanyKids* database during the same time period (8,642 patients).

Estimates of the number of children in the camp with several common congenital conditions were derived as the sum of the yearly number of new cases expected in the camp for each condition (calculated using average global incidence rates per 1,000 and yearly birth estimates in the camp) plus the yearly number of refugee children expected to enter the camp with each condition. The latter value assumes (based on consistent observations over the study period) that no child born with the index conditions received adequate care before reaching the camp. An example of such a calculation follows.

cleft lip cases expected in 2010 = 300,000 (average camp population in 2010 [23]) * 43/1,000 (Somalia birth rate) * 1/1,000 (average cleft lip incidence in Africa) + 13,000 (new refugees arrived in Dadaab in 2010) * 0.62 (percentage of children in Dadaab [23]) * 1/1,000 (average cleft lip prevalence) = 20.96.

DALY calculation

BoD estimates were calculated using previously established methods and country-specific life tables provided by the Global Burden of Disease Study [24]. DALYs were calculated as the product of the years lived with disability (YLD) and the disability weight (DW). Both universal and specific life expectancy tables for the sampled population (Somali) were used [25,26]. Years of Life Lost (YLL) were estimated for each patient based on their life expectancy at time of treatment. Age weighting and future discounting [24,25] are common methods in global burden of disease (GBD) studies, but remain somewhat controversial, especially when applied to children [27,28,29]. A sensitivity analysis was therefore performed on the life expectancy tables, using both “plain” values without any age weighting or discounting (so-called YLL(0,0)) and standard tables (YLL(3,1), denoting 3% discounting of future years and applied age weighting) [26].

Averted DALYs (representing *met* need) were defined as the number of DALYs potentially gained through each surgical procedure by averting death [YLL] and decreasing morbidity from condition for the rest of the child’s expected life (YLD) [16]. The YLD estimate includes the DW, the potentially less than unitary risk of permanent disability without surgery (RPD), and the less than unitary probability of a fully successful surgery (PST). The latter factor aims to account both for the possibility of post-operative complications and the need for further procedures. While DWs exist for a few pediatric surgical conditions [15], for the remaining conditions they were estimated by the authors using the functional scale provided by Murray [25]. Averted DALYs through any intervention are thus calculated as previously described [20,30] using the formula:

$$LE_{\text{Age at operation}} * DW * RPD * PST,$$

where the last 3 terms are constrained between 0 and 1.

In this study all interventions were elective and thus performed on chronic surgical disability states, thus we conservatively assumed that there were no actual deaths (or YLL) prevented through our interventions. The final averted DALYs in this study were therefore based on YLD only. Following is a graphic representation of the impact of surgical intervention on a patient:



Averted DALY values can be calculated either per patient, or as a cohort / population total. For instance, a 6 year-old boy with hydrocephalus treated by ventriculo-peritoneal shunt insertion would have 71.05 ($LE_{\text{Age at operation}}$) * 0.4 (DW) * 1 (RPD>95%) * 0.7 (PST between 50% - 95%) = 19.9 DALYs averted.

We also estimated the *delayed* averted BoD caused by the observed delays in provision of care (backlog). This was done by multiplying the DW with the delay in care (age of actual operation minus ideal age of operation). In the example above, the delayed averted surgical burden was $(6 - 0.2) * 0.4$ (DW) = 2.3 DALYs. Because of inherent challenges in accurate history of acquired conditions, this measurement was restricted to congenital malformations.

Cost-effectiveness analysis

Inpatient costs associated with surgical procedures were extracted from the patient database. This included actual patient bills for the operating room, hospital stay, medications and supplies, staff salaries, and administrative charges. While staff salaries were included in the hospital bill, the surgeons volunteered their time and some surgical supplies were donated. The cost-effectiveness ratio was derived from the aggregate cost of all surgical admissions divided by the total DALYs averted through the procedures performed.

Ethics

This study was approved by the Research Ethics Committee of BethanyKids at Kijabe Hospital.

Results

Between March 2005 and June 2011, 640 surgical procedures were performed at BethanyKids of Kijabe Hospital, Kenya on 341 children (under 18 years) from the Dadaab Refugee Camp. Median age of the children was 4.6 years and 66% were male. Forty-three percent of procedures were performed for congenital anomalies and 57% for acquired conditions.

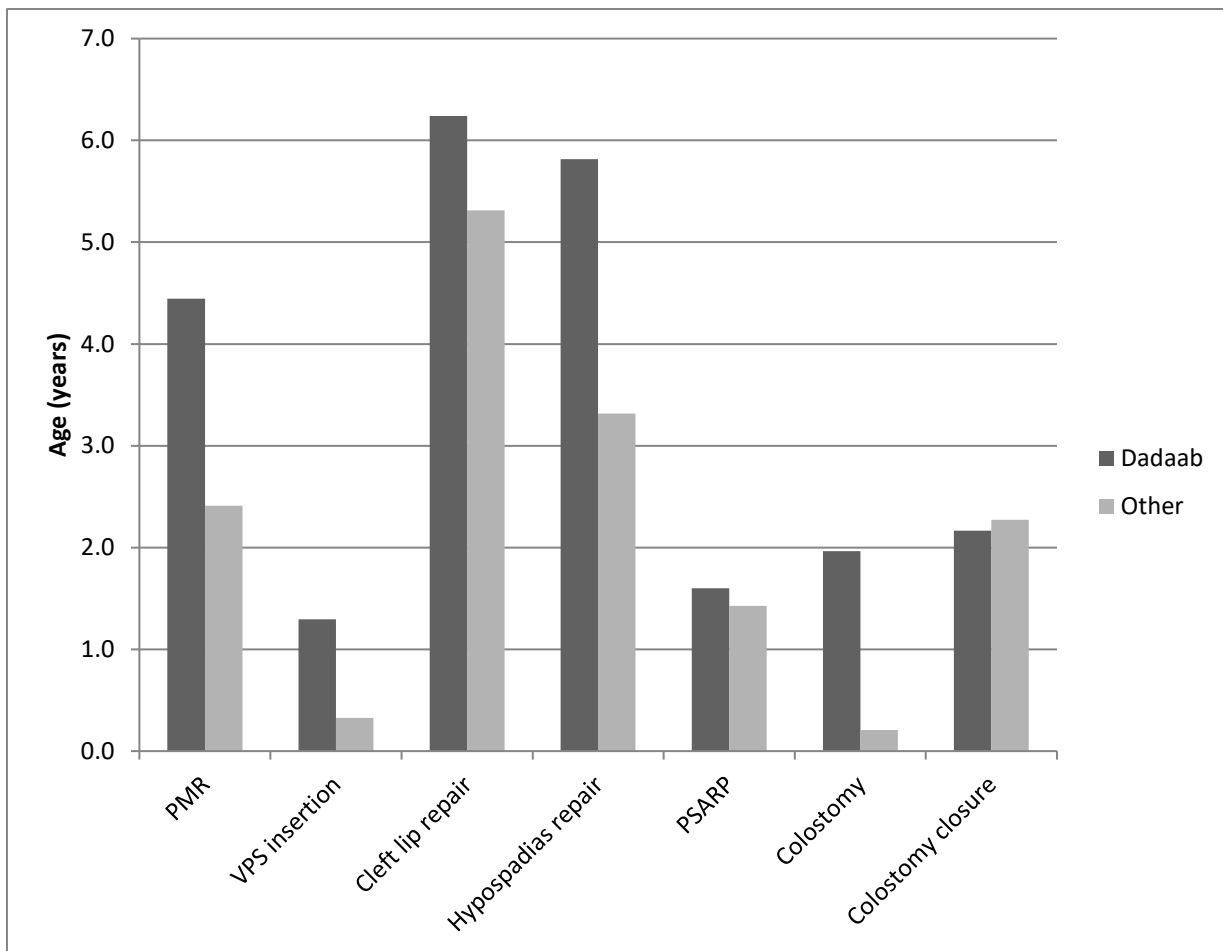
The commonest congenital and acquired procedures performed are listed in **Table 1**, with the median age at operation. **Figure 1** compares the age at surgery for the commonest congenital procedures on refugee children with a similar sample of non-displaced patients during the same time period. Cleft lip repair and burn contracture release were the commonest procedures performed for congenital and acquired conditions, respectively.

Table 1. Most common congenital and acquired surgical procedures with their median age

Procedure	Number	Median age [yrs]
Congenital		
Posterior medial release for club foot	34	4.4
VP shunt insertion	31	1.3
Cleft lip repair	40	6.2
Hypospadias repair	26	5.8
PSARP for imperforate anus	17	1.6
Colostomy	15	2.0
Colostomy closure	14	2.2
Acquired		
Dressing change	111	3.7
Contracture release	66	5.9
Debridement	20	2.2
Cast change	14	8.6
Biopsy	12	8.1

¹ Club foot was treated surgically rather than with serial casting because of the advanced age of the patients.

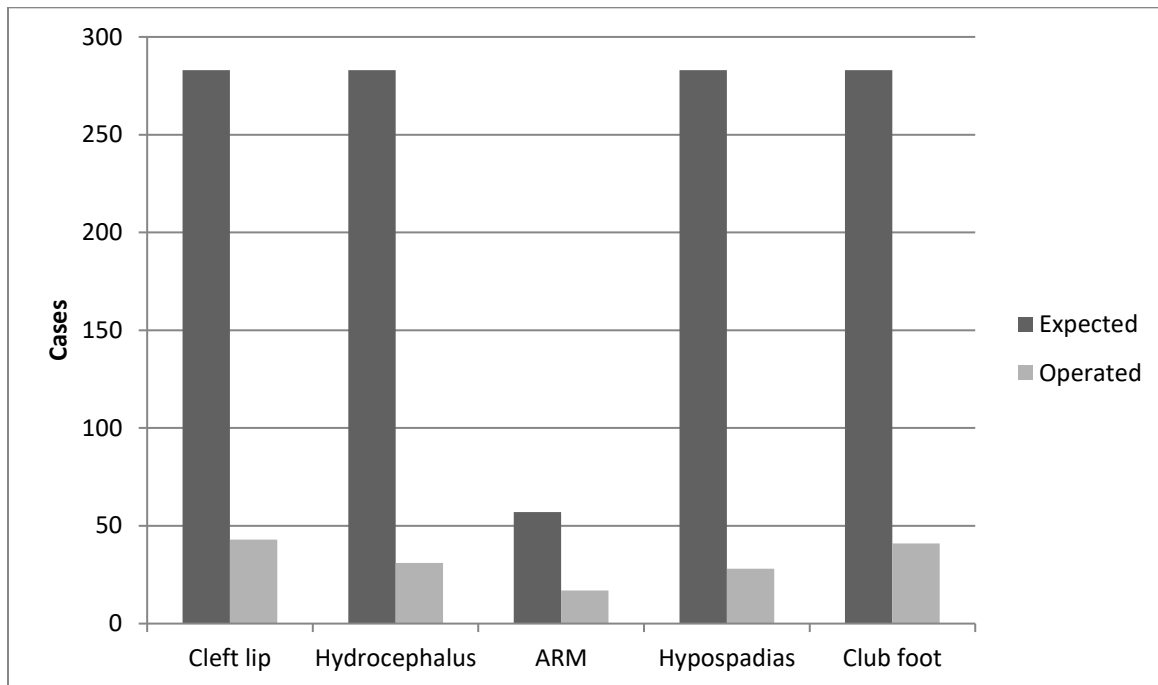
Figure 1: Median age of various surgical procedures for refugee vs. non-refugee patients



PMR = posterior median release; VPS = ventriculoperitoneal shunt; PSARP = posterior sagittal anorectoplasty

Figure 2 compares the expected cases of the 5 commonest congenital conditions over the study period with the actual number of procedures performed for those conditions. The ratio of surgeries performed over those expected was 13% (range, 10 – 30%).

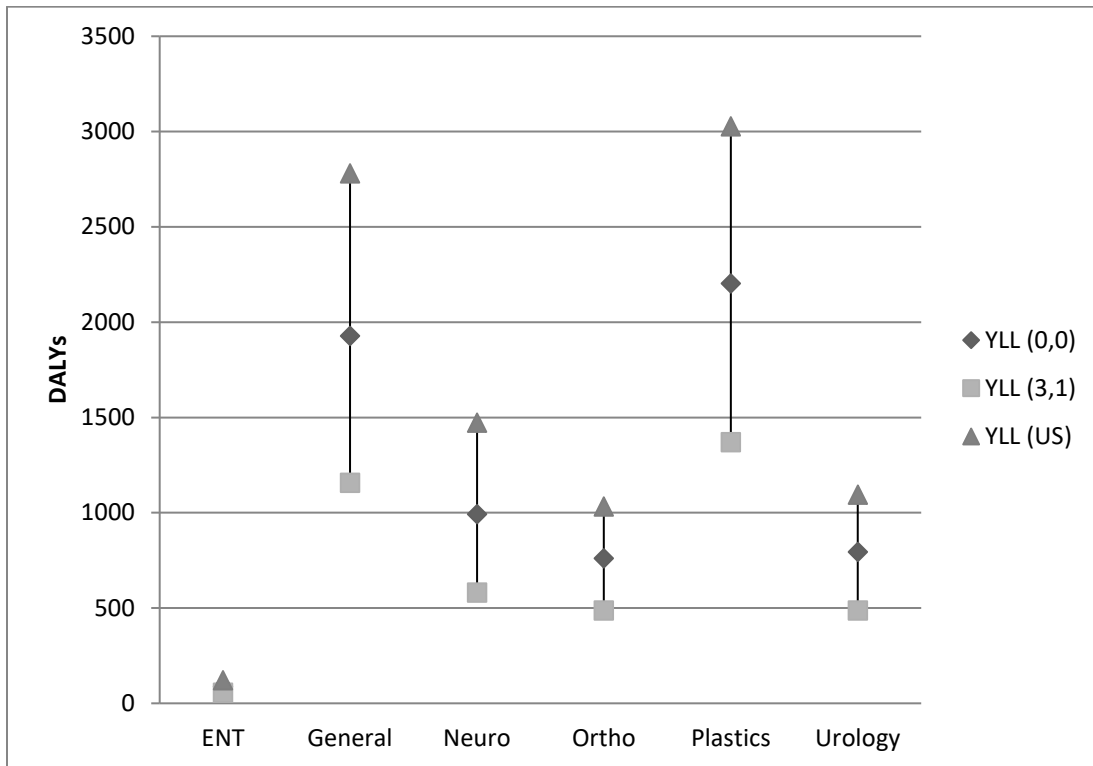
Figure 2: Number of children with 5 most common congenital conditions operated in Dadaab vs. estimated cases over study period



ARM = anorectal malformation

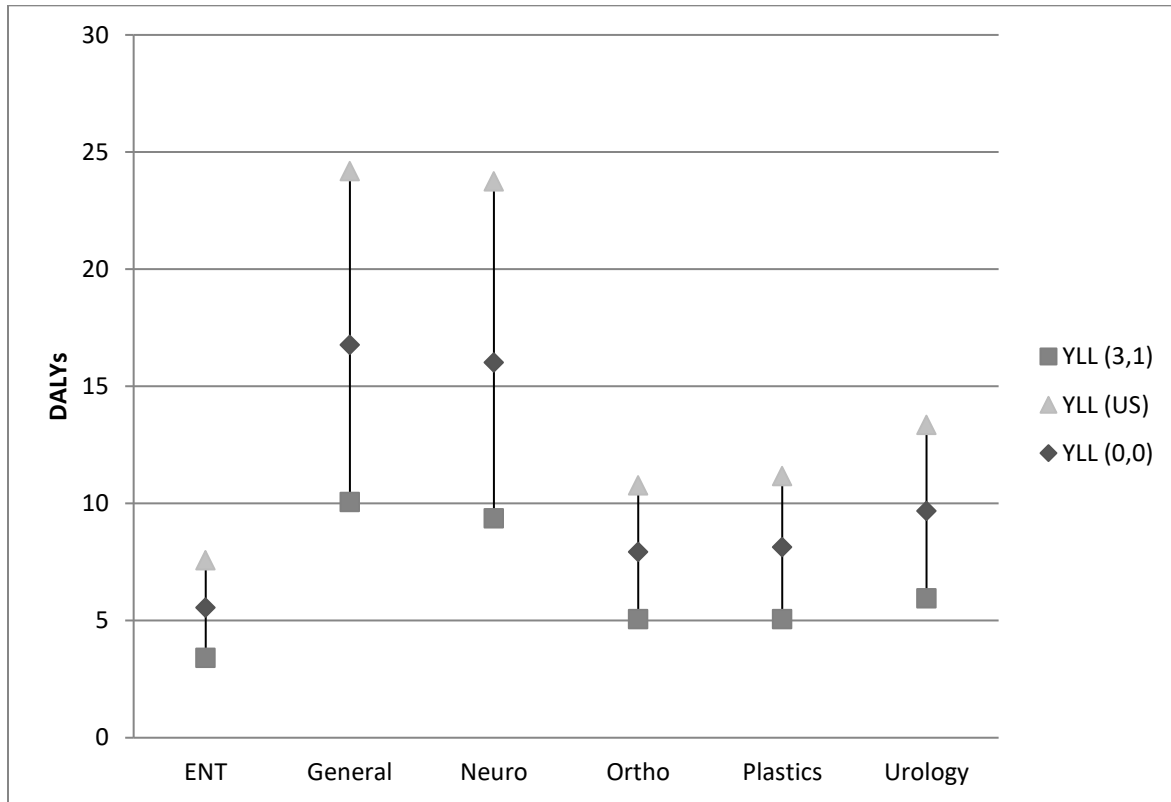
Estimated total and per procedure averted DALYs are presented in Figures 3 and 4, respectively, while delayed averted DALYs are shown in Figure 5. Sensitivity analysis resulted in a total of 4,136 – 9,529 DALYs averted over the study period (corresponding to 6.4 – 14.8 DALYs per patient), evenly distributed between congenital (totaling 2,065 – 4,911 DALYs, or 7.4 – 17.6 DALYs per patient) and acquired conditions (totaling 2,071 – 4,618 DALYs, or 5.7 – 12.7 DALYs per patient). The total delayed averted surgical BoD for congenital malformations in this study cohort was estimated at 300 DALYs over the study period, representing 1.1 DALYs per patient.

Figure 3: Sensitivity analysis for total burden of disease (in DALYs) averted by surgical specialty



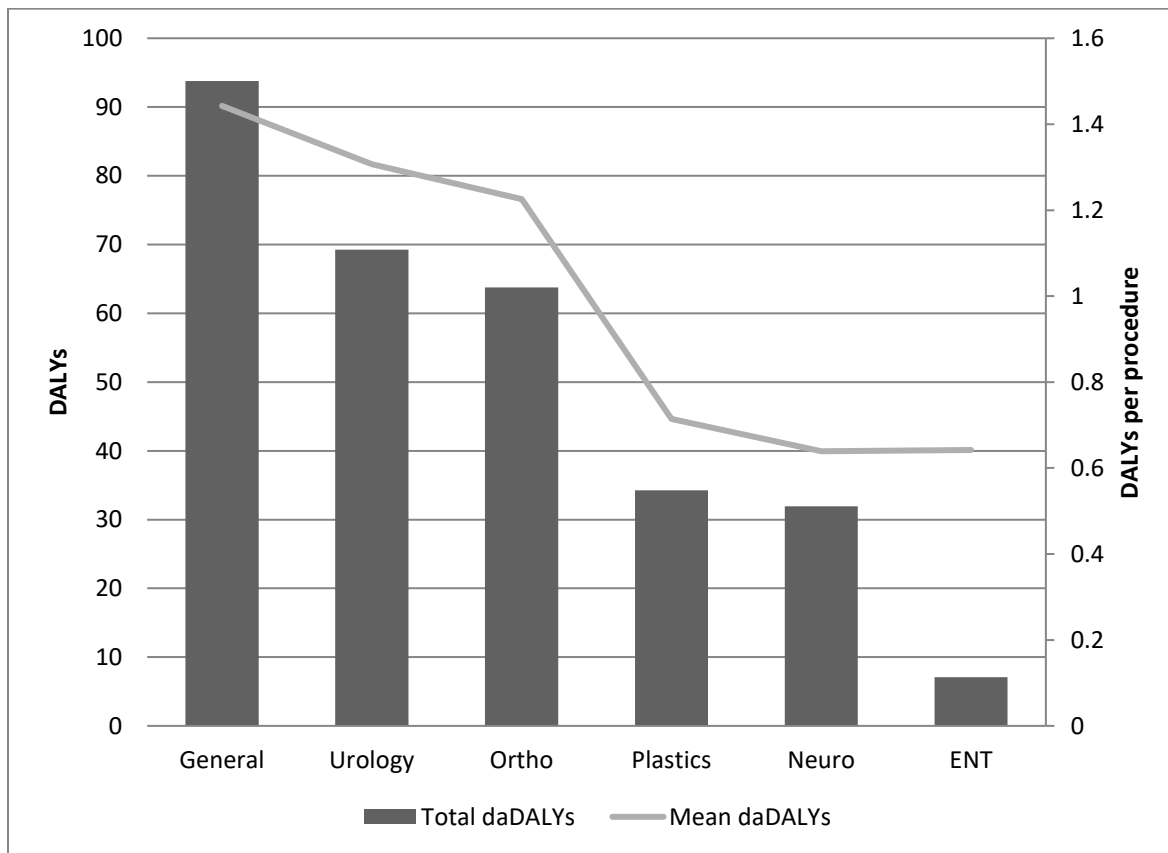
DALY (0,0): DALYs using standard undiscounted Kenya life expectancy table
 DALY (3,1): DALYs using age-weighted and 3% discounted Kenya life expectancy table
 DALY (US): DALYs using universal standard undiscounted life expectancy table

Figure 4: Sensitivity analysis for burden of disease (in DALYs) averted per surgical procedure by surgical specialty



DALY (0,0): DALYs using standard undiscounted Kenya life expectancy table
 DALY (3,1): DALYs using age-weighted and 3% discounted Kenya life expectancy table
 DALY (US): DALYs using universal standard undiscounted life expectancy table

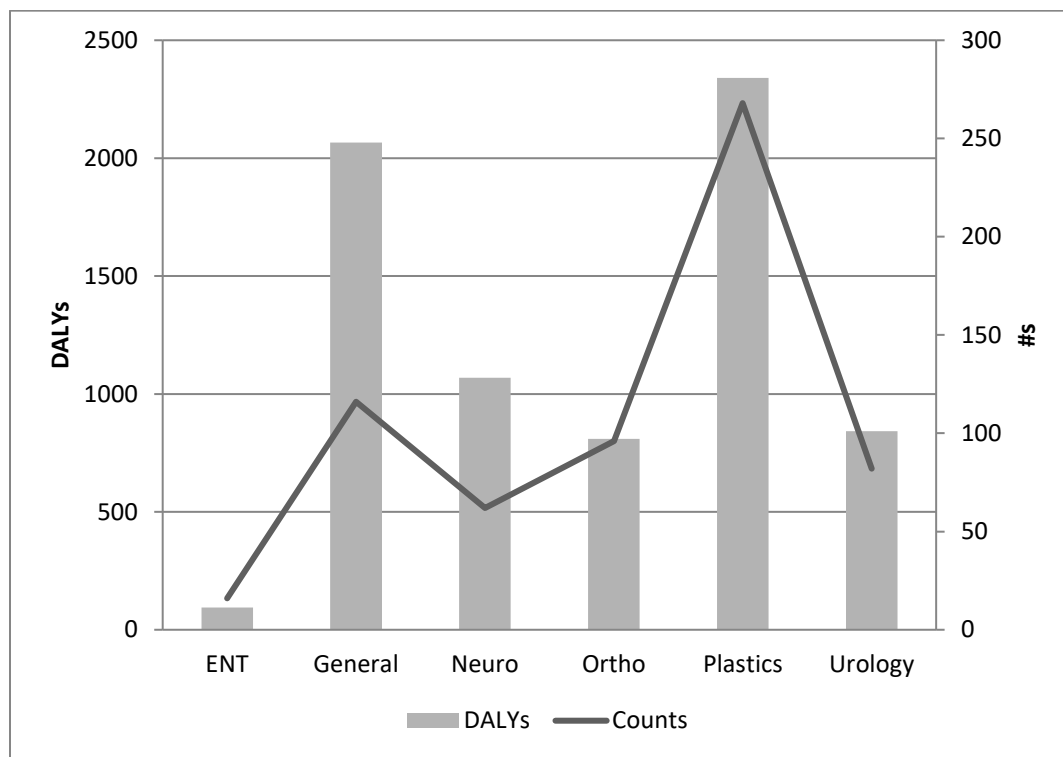
Figure 5: Total and average delayed averted DALYs by surgical specialty



daDALY = delayed averted DALY

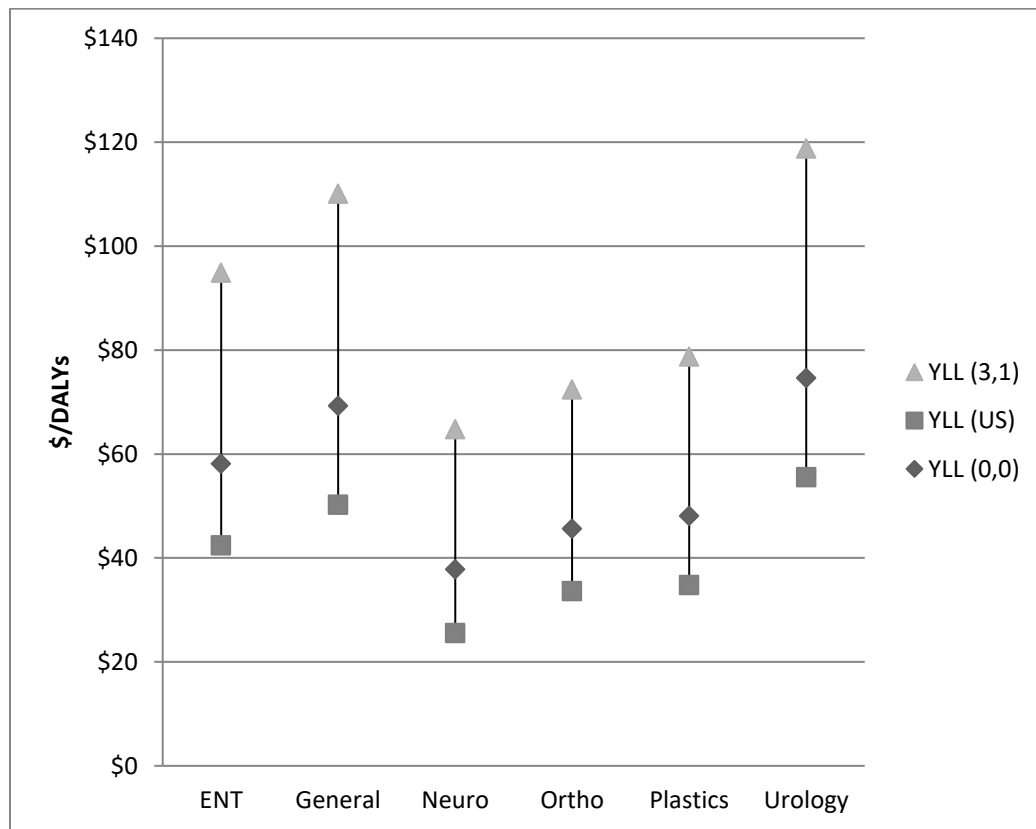
The number of surgical procedures by specialty is compared to the respective averted BoD in Figure 6. Plastic and general surgical procedures accounted for the most *total* attributable DALYs averted, while general and neurosurgical procedures resulted in the largest number of averted DALYs *per patient*. General surgery was also associated with the highest proportion of *delayed* averted DALYs (93.8).

Figure 6: Number of surgical procedures and corresponding DALYs in each specialty



Accurate hospital bills were available for 289 surgical admissions (45% of total). The total costs incurred from the surgical treatment of these children was \$141,675. This figure yielded cost-effectiveness ratios between \$40 - \$88 per DALY. Cost-effectiveness ratios for each specialty are presented in Figure 7, showing neurosurgical procedures to have the lowest cost per DALY (\$26 - \$65).

Figure 7: Sensitivity analysis on cost per averted DALY in each surgical specialty



DALY (0,0): DALYs using standard undiscounted Kenya life expectancy table
 DALY (3,1): DALYs using age-weighted and 3% discounted Kenya life expectancy table
 DALY (US): DALYs using universal standard undiscounted life expectancy table

Discussion

Displaced persons are a significant vulnerable group globally, and children are among the most vulnerable in most limited-resource settings – yet the surgical needs of children in refugee camp settings have been ignored in the literature. This is the first study to quantify the burden of surgically correctable disabilities among children in a large refugee camp. While burden of surgical disease is generally neglected across Sub-Saharan Africa [31], we have found in Dadaab this burden to be very significant. With the increasing backlog of persons seeking refugee registration in the camp, there is a compelling case to highlight the magnitude of the surgical work already performed, as well as still required, in this setting.

The demographics of the children cared for over the 6-year study period are not representative of any other known population group, as they reflect a multitude of forces affecting migrant populations in areas of armed conflict. Although there is no evidence that surgical disease is more common among refugees, vulnerable groups such as the young and the ill will be preferentially evacuated from conflict areas, thus appearing in refugee camps more frequently than in the general population [23]. Limited data from Dadaab however reveals that many disabilities are not recognized: in 2009 only 2.6% of the population were registered by the rehabilitation agencies [32]. Of these persons with disabilities, only 450 were under 18 years of age and had a physical disability. While the total number of surgical procedures performed by BethanyKids over 5 years compares well with this figure, it is obvious that 450 is a gross underestimate of pediatric physical disability in the camp, as many new children with surgical disabilities entered the camp over the study period. Figure 2 highlights the inadequacy of pediatric surgical care in Dadaab: on average only 13.5% of the estimated children with congenital surgical disabilities received treatment. While this figure is fraught with many uncertainties and assumptions, it does provide a rough idea of the magnitude of the surgical need in this displaced population.

The male preponderance of the operated children mirrors the 60% male refugee ratio found in several camp surveys [23]. Other explanations may include issues of gender-based access to care and a higher prevalence of acquired injury-related surgical disabilities in males.

The unique variety of surgical conditions, both congenital and acquired, presenting in the refugee camp reflects a population virtually devoid of previous access to surgical care. Thus it is expected that most, if not all, fatal conditions have died, while non-fatal conditions generally survived, first presenting for care at whichever age refugees reached the camp health care system (Table 1, Fig. 1). This extreme scenario virtually transforms urgent pediatric surgical conditions into chronic elective disabilities – as witnessed by teenagers and even adults with unrepaired open bladder exstrophy, cleft lip, or recto-vestibular fistula.

This unique spectrum of disease, quite different from that of “traditional” pediatric surgery, requires a corresponding unique skill set for any organization attempting to serve such populations. At *BethanyKids* we have developed over the years such expertise, and are passing it

on to national pediatric surgeons through a disability-focused accredited pediatric surgical training program.

While DALYs may not provide the most accurate reflection of the true perception of a disease state by the individual [25], they have rapidly become the most common metric for BoD. The total BoD in any population includes the met, unmet and unmeetable components [16]. In the context of a refugee camp, the unmet need continuously fluctuates based on the demographics and health status of the incoming and outgoing refugees. Due however to the fact that *BethanyKids* was the sole provider of pediatric surgical care in the camps over several years, we had the unique opportunity to estimate the magnitude of the met need. This magnitude depends on the life tables used and the inherent assumptions regarding age weighting, future discounting, and use of country-specific vs. universal tables discussed above. Generally the lowest DALY values were generated by the country-specific, discounted tables - while undiscounted, universal tables produced the highest values. While each of these options has its inherent pros and cons, it is reassuring that all DALY estimates were well within one order of magnitude.

The ability of DALYs to capture the burden of chronic disability is highlighted by the disparity between numbers of surgical procedures in each specialty and their corresponding BoD. General surgical and neurosurgical procedures appear to be associated with the greatest BoD, likely because of the higher DW frequently characterizing congenital malformations in these specialties (such as imperforate anus and spina bifida).

Besides the *met* surgical need attributable to surgical interventions, our data allows the calculation of the *delayed* averted burden, caused by the years lived with disability (YLD) preceding the surgical treatment. This previously unidentified factor is likely a sub-component of the *unmet* need, although its irrecoverable nature makes it behave more like an *unmeetable* need. Not only is past suffering not recoverable, but, in the case of many pediatric surgical conditions, delay in care may be associated with more challenging surgical procedures and a higher likelihood of complications. A 20-year old patient with unoperated cleft palate will not only have suffered for 20 years from the sequelae of her condition, but her surgery will be more challenging than in a 1 year-old, and her speech deficits likely irreversible. The advanced age at surgery for congenital malformations in the displaced children compared to non-displaced ones

(Fig. 1) highlights an even greater surgical backlog in the refugee camps than in the surrounding, already severely resource-limited, LMICs - and the higher delayed averted burden in this most vulnerable population. The tremendous challenge caused by armed conflict and population displacement is further increased by the inadequacy of the current pediatric surgical care provided (evidenced by less than 25% of children with congenital surgical conditions receiving treatment). Ongoing efforts by the UNHCR and its implementing partner agencies to improve health care in the camps are hindered by the constant increase in refugee numbers and the security concerns of the ongoing armed conflict in the region [8].

Our study draws upon previously established CEA methods for surgical interventions [19,20,2828]. The current cost-effectiveness ratios of \$40 - \$88 per DALY compare favorably with other studies showing surgical intervention to be a cost-effective method of decreasing BoD. Debas *et al* [11] estimated a cost per averted DALY in LMICs between \$33 and \$94, while Gosselin *et al* arrived at CEA values of \$172 / DALY in a Nigerian hospital and \$223 per DALY in Haiti [30]. Our cost per DALY may have been artificially decreased by the exclusion of “fixed” costs like the depreciation of hospital equipment, and the reliance on donated surgeon time and equipment. More likely however, even highly specialized surgical procedures on children are likely to be cost-effective because of the long life expectancy following the procedure, and hence the many years of disability averted. This favorable CEA supports the inclusion of pediatric surgical care in the package of services offered to displaced populations.

The most cost-effective procedures proved to be neurosurgical ones (due likely to the impact of relatively simple procedures like ventriculo-peritoneal shunts on high-DW conditions), while urological operations were the most expensive (the majority being hypospadias repairs, with a relatively low DW).

Limitations

Our study has several limitations. Despite the apparently comprehensive nature of the surgical care provided by the *BethanyKids* facility, the type of procedures performed reflected in part the type of surgical expertise available during that time. Moreover, with waiting times up to one year

for most procedures, our data cannot be used to estimate true BoD in the camp over a given period of time – this remains a task for future studies.

While we have attempted to capture the uncertainty factor intrinsic to the various life tables [33] in the sensitivity analysis, the 3 estimated YLL modifiers (DW, RPD, and PST) each carry significant additional uncertainty. As most DWs for the conditions included in the study were not available in the literature, the study needed to rely on much coarser estimates using published methods. As for the RPD and PST, they are both not only crude but very context-dependent: for many surgical conditions, delay in treatment can increase both the risk of permanent disability (such as speech problems in cleft palate) and lower the probability of successful treatment (such as urinary continence in bladder exstrophy closure). Moreover, the PST depends significantly on the quality of the surgical facilities, equipment and expertise available, as well as on a multitude of other patient-related factors (such as nutritional status, anemia, and other co-morbid conditions) which are difficult to quantify. Also as stated in the Methods, we have conservatively assumed that no actual deaths were averted by our elective procedure, as the vast majority of deaths in our patient population from their conditions would have occurred before they had access to surgery.

Finally, all CEA inherently carries multiple assumptions and a large degree of uncertainty [28]. While some limitations were evident in our case (surgeons' salaries, donated equipment, fixed costs), dollar values associated with surgical work remain difficult to assign with any certainty.

Conclusion

This first estimate of burden of surgical disease averted in the Dadaab Refugee Camp highlights the significant impact that surgical intervention can have in such a setting, and its cost-effectiveness. Given the magnitude of the number of persons displaced by armed conflicts across the globe, such data can inform appropriate resource allocation for the health care needs of these vulnerable populations.

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References

1. World Health Organization (2011) Health Statistics and health information systems: Life tables for WHO Member States. World Health Organization
<http://www.who.int/entity/healthinfo/statistics/LT199020002009whs2011.zip>. Retrieved January 16, 2012
2. Natale JE, Joseph JG, Bergen R, et al (1992) Prevalence of Childhood Disability in a Southern Indian City: Independent Effect of Small Differences in Social Status. *Int J Epidemiol* 21:367-372
3. Schalamon J, Bismark S, Schober PH, et al (2003). Multiple trauma in pediatric patients. *Pediatr Surg Int* 19:417-423
4. Davey TM, Aitken LM, Kassulke D, et al (2005) Long-term outcomes of seriously injured children: A study using the Child Health Questionnaire. *J Paediatr Child Health*, 41:278-283
5. Kirpalani HM, Parkin PC, Willan AR, et al (2000) Quality of life in spina bifida: importance of parental hope. *Arch Dis Child* 83:293-297
6. Maulik PK, Darmstadt, GL (2007) Childhood Disability in Low- and Middle-Income Countries: Overview of Screening, Prevention, Services, Legislation, and Epidemiology. *Pediatrics* 120:S1-S55
7. Power C, Leah L, Manor O (2000) A prospective study of limiting longstanding illness in early adulthood. *Int J Epidemiol* 29:131-139
8. Aguilar S (2011) Dadaab: Walking the fine line between helping refugees and risking lives. UNHCR - The UN Refugee Agency. <http://www.unhcr.org/4ed3bc316.html> Retrieved January 16, 2012
9. UNHCR (2012) 2012 UNHCR country operations profile - Kenya. UNHCR - The UN Refugee Agency. <http://www.unhcr.org/cgi-bin/txis/vtx/page?page=49e483a16&submit=GO> Retrieved January 16, 2012
10. Reilly R (2008) Disabilities among refugees and conflict-affected populations. *Forced*

Migration Review 35:8–10

11. Debas HT, Gosselin R, McCord C, et al Surgery (2009) In: Jamison DT, Breman JG, Measham AR, et al (eds) Disease Control Priorities in Developing Countries, 2nd ed. The World Bank, Washington, p. 1245-1260
12. Bickler SW, Sanno-Duanda B (2000) Epidemiology of paediatric surgical admissions to a government referral hospital in the Gambia. *Bull World Health Organ* 78:1330-1336
13. Albertyn R, Bickler SW, van As AB, et al (2003) The effects of war on children in Africa. *Pediatr Surg Int* 19:227-232
14. Azzie G, Bickler S, Farmer D, et al (2008) Partnerships for developing pediatric surgical care in low-income countries. *J Pediatr Surg* 43:2273-2274
15. World Health Organization (2008) Global Burden of Disease: 2004 Update. World Health Organization, Geneva
16. Bickler S, Ozgediz D, Gosselin R, et al (2010) Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg* 34:374-80
17. Vos T (2009) Improving the quantitative basis of the surgical burden in low-income countries. *PLoS Medicine* 6:e1000149
18. Edejer TT, Baltussen R, Adam T, et al (eds) (2003) WHO Guide to cost-effectiveness analysis. World Health Organization, Geneva
19. Gosselin, R. & Heitto, M (2008) Cost-effectiveness of a district trauma hospital in Battambang, Cambodia. *World J Surg*, 32:2450-2453
20. McCord C, Chowdhury Q (2003) A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet* 81:83-92
21. Toole MJ, Waldman RJ. The public health aspects of complex emergencies and refugee situations (1997) *Pub Health* 18:283-312
22. Shears P, Berry AM, Murphy R, et al (1987) Epidemiological assessment of the health and nutrition of Ethiopian refugees in emergency camps in Sudan, 1985. *BMJ* 295:314-318

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23. UNHCR (2009) Dadaab Statistics at a glance. *Pop Trends* 2009:1-2.
 24. Lopez AD, Mathers CD, Ezzati M, et al eds. (2006) *Global Burden of Disease and Risk Factors*. Oxford University Press & The World Bank, New York, NY, p.400-402
 25. Murray CJL (1994) Quantifying the burden of disease : the technical basis for disability-adjusted life years. *Bull WHO* 72:429-445
 26. World Health Organization (2011) *Health Statistics and health information systems: Life tables for WHO Member States*
<http://www.who.int/entity/healthinfo/statistics/LT199020002009whs2011.zip> Retrieved January 16, 2012
 27. Keren R, Pati S, Feudtner C (2004) The generation gap: differences between children and adults pertinent to economic evaluations of health interventions. *Pharmacoeconomics* 22:71-81
 28. Corlew DS (2010) Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. *World J Surg* 34:391-396
 29. Anand S, Hanson K (1997) Disability-adjusted life years: a critical review. *J Health Econ* 16:685-702
 30. Gosselin R, Maldonado A, Elder G (2010) Comparative cost-effectiveness analysis of two MSF surgical trauma centers. *World J Surg* 34:415-419
 31. Ozgediz D, Riviello R (2008) The "other" neglected diseases in global public health: surgical conditions in sub-Saharan Africa. *Plos Med* 5:e121
 32. Handicap International (2009). Supporting disabled people in the Dadaab refugee camp. Retrieved from http://www.handicap-international.org.uk/where_we_work/africa/kenya/news/dadaab.htm
 33. Mathers CD, Salomon JA, Ezzati M, et al (2006) Sensitivity and Uncertainty Analyses for Burden of Disease and Risk Factor Estimates. In: *Global Burden of Disease and Risk Factors*, Oxford University Press, p. 399-426

Chapter 7

Quantifying the Disability from Congenital Anomalies Averted Through Pediatric Surgery: A Cross-Sectional Comparison of a Pediatric Surgical Unit in Kenya and Canada

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Abstract

Background

Pediatric surgical practice is different in low- and middle-income countries as compared to North America. While resources are limited, the impact of pediatric surgical procedures is significant. The objective of this study was to calculate and compare Disability-Adjusted Life Years (DALYs) averted in a Kenyan and Canadian surgical unit for a subset of pediatric congenital anomalies.

Methods

Medical records of children having undergone surgical procedures for 13 congenital conditions in both surgical units were collected over 12 months. DALYs for each condition were calculated using previously obtained disability weights derived in each country. Age-adjusted life expectancy rates from the WHO were used to determine years of life lost (YLL). Risk of permanent disability without surgery (RPD) and probability of successful treatment (PST) values were obtained from the literature and included in the DALY calculation.

Results

The conditions accounting for the largest total number of averted DALYs in Kenya were hydrocephalus (60.8%) and spina bifida (18.1%), whereas in Canada they were hydrocephalus (24.2%) and undescended testes (19.2%). A total of 23,169 DALYs were averted through 1,042 surgical procedures (22.2 DALYs per procedure) during the study period in Kenya, compared to 5,497 DALYs through 373 procedures (14.7 DALYs per procedure) in Canada.

Conclusions

Using recent developments in burden of disease measurement, the results point to the significant impact of pediatric surgical centers in addressing the global burden of congenital surgical disease. The study carries significant implications for resource allocation and training.

Introduction

An estimated 93 million children live with some form of moderate or severe disability worldwide [1]. Among the top ten leading causes of pediatric morbidity and mortality worldwide are congenital conditions, which affect 7% of all births around the world [2]. The burden of congenital anomalies on children, families, and health systems is particularly acute in low- and middle-income countries (LMICs), which experience a disproportionate number of these serious birth defects (94%) [3,4]. Children affected with these congenital anomalies, who survive infancy, often continue to live with disability and experience significant physiological and psychological effects, including community stigma and discrimination [5].

Timely surgical intervention prevents both death and disability associated with many congenital anomalies. Lack of access to surgically trained healthcare providers, operating room facilities, and surgical supplies are some of the factors that impair the delivery of adequate surgical care in LMICs for these conditions [6]. This lack of access continues to exist despite recent economic analyses that demonstrate that surgery is a more cost-effective means of reducing disease burden than many other medical interventions [7-9], and the recognition that access to surgical care is one of the top 8 interventions integral to the progression of welfare among the world's poorest populations [10].

Traditional research methods of quantifying and evaluating the effect of surgical interventions have underestimated the larger impact of surgery in preventing a lifetime of disability in a child. The Global Burden of Disease (BoD) Study uses the Disability-Adjusted Life Year (DALY), a health metric combining morbidity and mortality, which has become the tool of choice for policy-makers involved in priority-setting and resource allocation [11]. The DALY measure combines Years of Life Lost (YLL) with Years Lost due to Disability (YLD), recognizing the disease burden associated with non-fatal yet disabling conditions [12, 13]. Using the DALY methodology, previous studies have determined that surgical conditions account for 11% of the global BoD, and in Africa the surgical BoD is nearly double that estimated in North America [14]. Furthermore the Disease Control Priorities (DCP) study has established that congenital anomalies contribute 9% of the surgical BoD [15].

In terms of the need for surgical care, the surgical BoD can be subdivided into three categories: *met need* (averted DALYs), *unmet need* (avertable DALYs) and *unmeetable need* (unavoidable morbidity and mortality even with intervention) [16]. Estimates of *averted* DALYs can be used to compare the cost-effectiveness of surgical interventions with other medical treatments, as mentioned above, while estimates of *avertable* DALYs primarily inform priority-setting and the allocation of limited health resources.

Although some researchers have compared the incidence of neonatal surgical conditions in LMICs with high-income countries (HICs) [17], no studies have compared the impact of pediatric surgery in HICs with LMICs using the DALY metric. The objective of this study was to use the DALY methodology to compare the averted burden of surgical disease from 13 congenital health states addressed by pediatric surgeons working in a high-income country (Canada) with a low-income country (Kenya).

Materials and Methods

Settings

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

McMaster Children's Hospital (MCH) is located in Hamilton, a major metropolitan city in southern Ontario, Canada; it is a regional pediatric academic tertiary care center. Comparative data on the two sites are presented in **Table 1** [18-20].

Table 1 Comparison of the Two Study Settings

	<i>BethanyKids</i> at Kijabe Hospital (Kenya)	McMaster Children’s Hospital (Canada)
Number of pediatric beds	67	159
Number of operating rooms	2	4
Number of pediatric surgeons covering chosen subspecialties	2	11
Number of pediatric surgical procedures per year	2,200	4,935
Referral population	43 million (not exclusive provider)	2.3 million (exclusive provider)
Fertility rate (births/1000 women) [18]	4.5	1.6
Country rank in UN Human Development Index (out of 187) [19]	145	11
Country physician: population ratio (per 1,000) [20]	0.2	2.1

Data Collection

We chose to compare 13 congenital health states addressed through pediatric surgical procedures in both centers. We used the procedures as proxies for the health states and included pediatric surgical subspecialties available at both sites: pediatric general surgery, urology, plastic surgery, and neurosurgery. The 13 health states chosen included mild and severe forms of major congenital anomalies which could be easily compared. For example, “mild imperforate anus” (perineal fistula or low vestibular fistula) did not require a colostomy, while “severe imperforate anus” (rectourethral fistula or cloaca) meant that a colostomy was required. Similarly, “severe spina bifida” was defined as paraplegia, and “severe hypospadias” was defined as having a penile shaft or more proximal opening requiring a complex repair. All cases of gastroschisis or omphalocele were considered “severe abdominal wall defects”. In the case of health states requiring staged repairs, all necessary procedures were included.

After research ethics approval at both sites (Hamilton Health Sciences Integrated Research Board (#11-328) and 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 intervention, and age at time of procedure were abstracted from patient records.

BoD Analysis

The disability prevented through surgery at each site was estimated using published DALY-based methods [8]. We examined only the *met* need for surgical care, expressed in averted DALYs, using the formula:

$$\text{Averted DALYs} = \text{YLL} \times \text{DW} \times \text{RPD} \times \text{PST},$$

where Years of Life Lost (YLL) is the sum of years lost through premature mortality and the Disability Weight (DW) is a factor reflecting the severity of a disease on a scale from 0 (perfect health) to 1 (equivalent to death). Risk of Permanent Disability (RPD) and Probability of Successful Treatment (PST) are variables constrained between 0 and 1 that account for the long-term impact of an intervention [8].

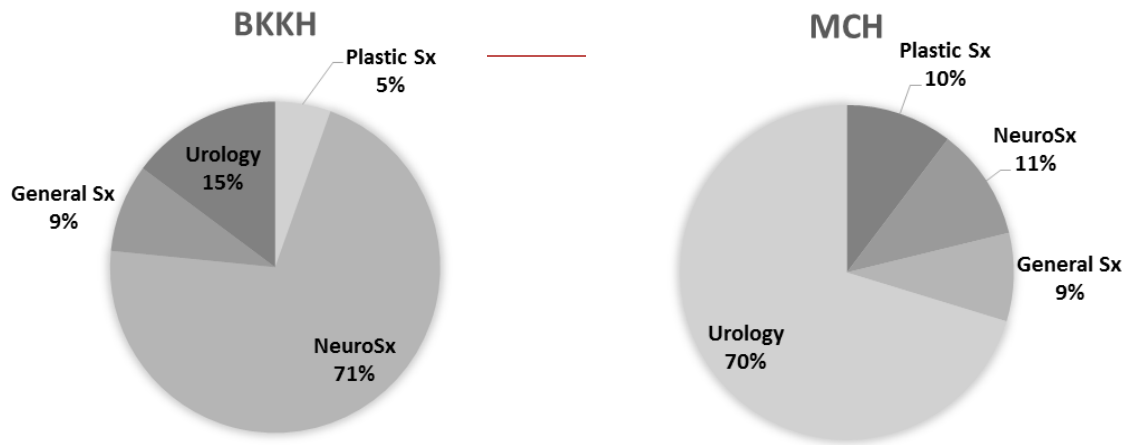
YLL values were determined by subtracting the age of the patient undergoing surgery from his or her life expectancy at the time of treatment, using country- and gender-specific discounted and age-adjusted life expectancy tables from the World Health Organization (WHO) [21]. Country-specific DW values for each health state were derived from our previous study [22]. RPD and PST values were determined using an established scoring system from the literature [8,23] and were agreed upon by an expert panel of surgeons. Given the nature of congenital anomalies, it was assumed that all health states would result in permanent disability or death if left untreated (RPD=1). The value chosen for PST was 0.7 for all conditions except for severe spina bifida (0.3), undescended testis (0.1) and mild hypospadias (0.1). In the case of health states requiring multiple (staged) procedures, the incremental BoD averted through each procedure was calculated separately, commensurate to its relative impact on the health state.

Once averted DALYs per patient were estimated, the total averted DALYs at each site over a period of one year were calculated and compared. All descriptive statistics, including counts, percentages, means and standard deviations, were performed using SPSS v20.0 (Chicago, Illinois).

Results

The mean age of the patients included was 2.2 years (SD=3.8) at BKKH and 3.0 years (SD=3.8) at MCH; 38% were female at BKKH and 16% at MCH. The distribution of surgical procedures by specialty in each site is presented in **Figure 1**. Over 2/3 of procedures at BKKH were neurosurgical, while at MCH were urological. **Table 2** lists the disability weights and numbers of procedures performed at each site for each health state, from which total and mean averted DALYs were estimated; the procedures performed for each health state are included, as well as a sample calculation of averted DALYs for clarification. The mean age at operation for each health state is provided in **Figure 2**, comparing values for each at BKKH with MCH. The delays in surgery at the Kenyan site are evident, particularly for children with cleft, gastrointestinal and genitourinary conditions. **Figure 3** compares mean DALYs averted through surgery per patient for each health state. Surgery for severe abdominal wall defects and intestinal atresia led to the highest mean averted DALYs per patient in both Kenya (38.06 and 36.29 DALYs respectively) and in Canada (50.16 and 53.93 DALYs respectively).

Figure 1 Proportion of Procedures at each Site by Pediatric Surgical Subspecialty



Distribution of the surgeries (Sx) performed per subspecialty at *BethanyKids* at Kijabe Hospital (BKKH) and McMaster Children's Hospital (MCH) between November 2011 and October 2012

Table 2 Health States and Averted DALYs Distribution at each Site

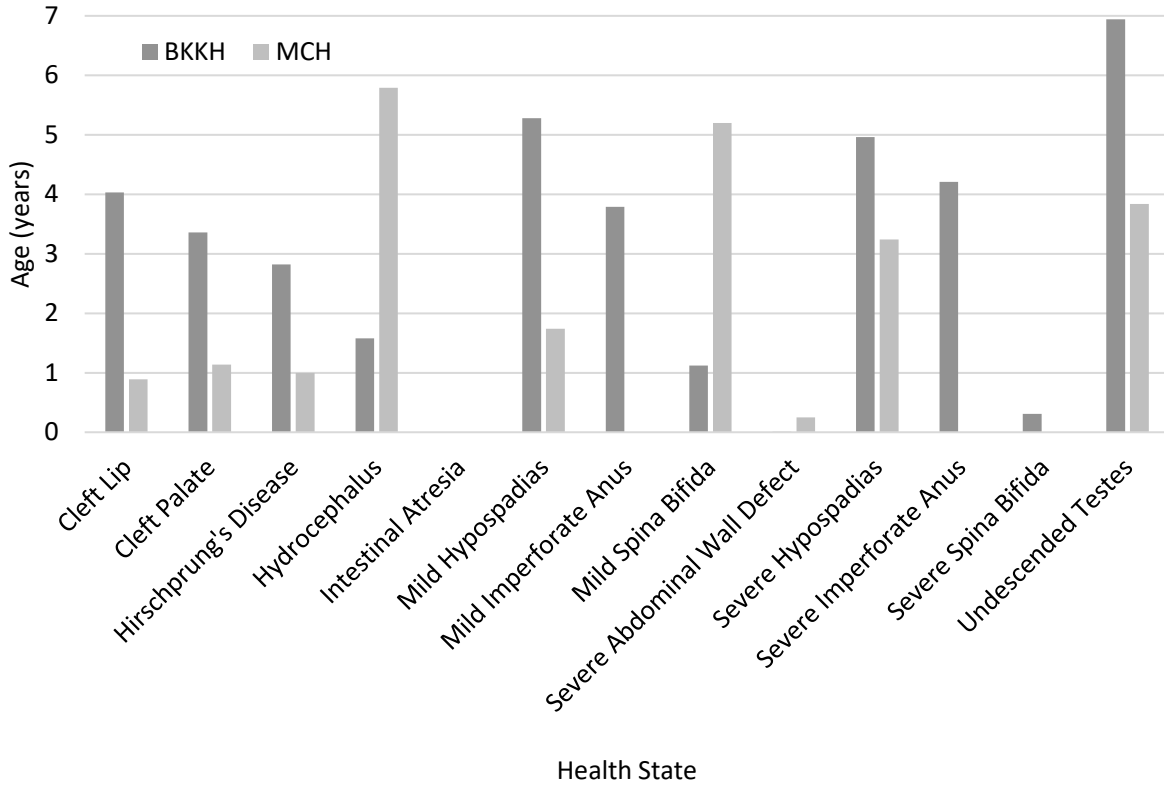
Health State	Surgical Procedure	Disability Weights (DW) ²		Number of Cases (%)		Mean Averted DALYs per patient		Total Averted DALYs (%)	
		BKKH	MCH	BKKH	MCH	BKKH	MCH	BKKH	MCH
Site									
<i>Plastic Surgery</i>									
Cleft Lip	cleft lip repair	0.18	0.22	46 (4.8)	19 (5.1)	7.63	12.39	351 (1.5)	235 (4.3)
Cleft Palate	cleft palate repair	0.38	0.35	11 (1.1)	21 (5.6)	15.93	12.39	175 (0.8)	417 (7.6)
<i>General Surgery</i>									
Hirschsprung's Disease	colostomy, pull-through, colostomy closure	0.72	0.69	5 (0.5)	5 (1.3)	30.08	39.28	150 (0.6)	196 (3.6)
Mild Imperforate Anus	anoplasty	0.28	0.32	31 (3.2)	4 (1.1)	11.62	18.66	360 (1.6)	75 (1.4)
Severe Imperforate Anus	colostomy, PSARP ³ , colostomy closure	0.81	0.73	37 (3.8)	2 (0.5)	33.82	40.32	1251 (5.4)	81 (1.5)
Severe Abdominal Wall Defect	abdominal wall closure	0.92	0.88	3 (0.3)	10 (2.7)	38.06	50.16	114 (0.5)	502 (9.1)
Intestinal Atresia	atresia repair	0.90	0.95	1 (0.1)	10 (2.7)	36.29	53.93	36 (0.2)	539 (9.8)
<i>Neurosurgery</i>									
Hydrocephalus	ventriculoperitoneal shunting or ETV ⁴	0.74	0.75	456 (47.4)	33 (8.8)	30.89	40.39	14085 (60.8)	1333 (24.2)
Mild Spina Bifida	closure spina bifida	0.46	0.33	20 (2.1)	4 (1.1)	19.35	17.94	387 (1.7)	72 (1.3)
Severe Spina Bifida	closure spina bifida	0.87	0.81	268 (27.8)	4 (1.1)	15.64	24.66	4193 (18.1)	99 (1.8)
<i>Urology</i>									
Undescended Testes	orchidopexy	0.22	0.08	87 (9.0)	164 (44.0)	12.59	6.45	1095 (4.7)	1058 (19.2)
Mild Hypospadias	hypospadias repair	0.12	0.10	40 (4.2)	65 (17.4)	6.98	7.83	279 (1.2)	509 (9.3)
Severe Hypospadias	hypospadias repair	0.50	0.22	25 (2.6)	32 (8.6)	20.54	11.93	513 (2.2)	381 (6.9)
Total				1,042	373			23,169	5,497

¹ An example of calculation of averted DALYs for one patient with hydrocephalus follows:

$$\text{Averted DALYs} = \text{YLL} \times \text{DW} \times \text{RPD} \times \text{PST} = 62.1 \times 0.74 \times 1 \times 0.7 = 32.17$$

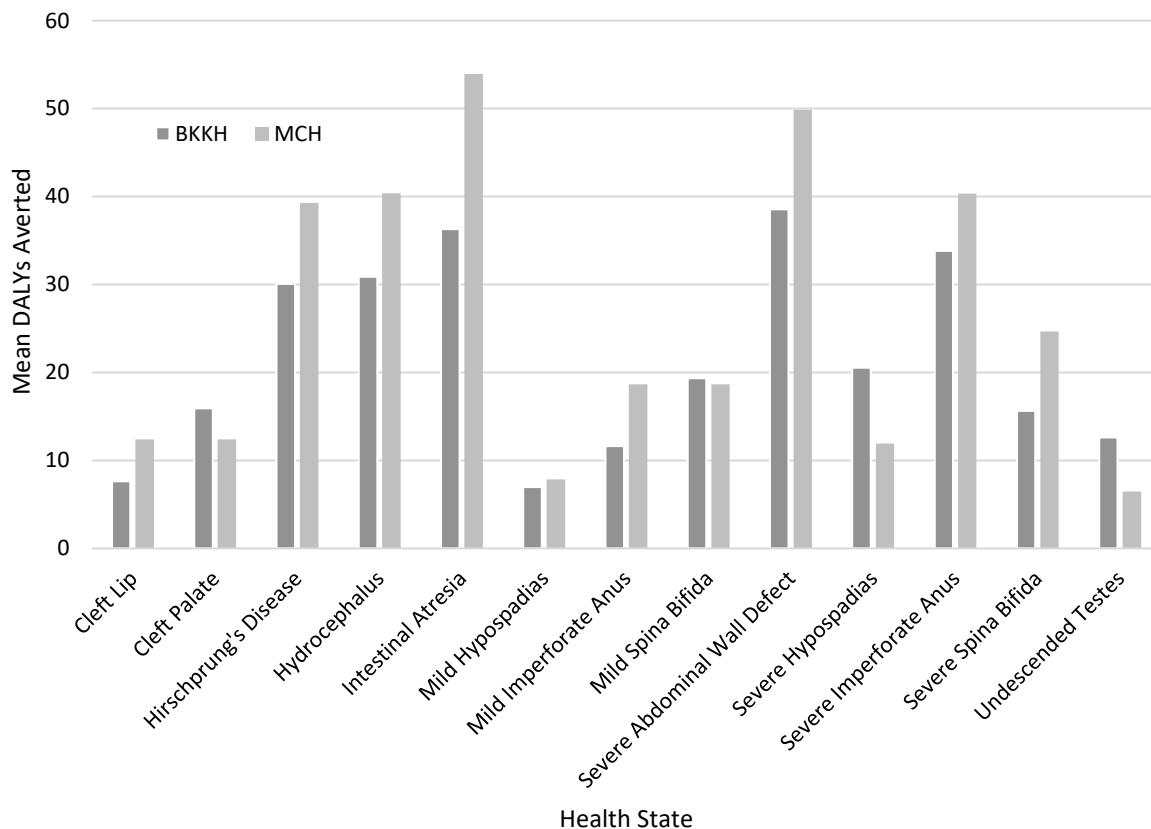
²DWs derivation detailed in concurrent publication [22]; ³ posterior sagittal anorectoplasty; ⁴ endoscopic third ventriculostomy

Figure 2 Mean Age at Operation at each Site by Health State



Distribution of ages of patients undergoing pediatric surgical procedures at *BethanyKids* at Kijabe Hospital (BKKH) and McMaster Children's Hospital (MCH) between November 2011 and October 2012

Figure 3 Mean DALYs Averted per Patient by Health State



Distribution of the DALYs Averted per health state based on the patient population at *BethanyKids* at Kijabe Hospital (BKKH) and McMaster Children's Hospital (MCH) between November 2011 and October 2012

The individual health states accounting for the largest total number of averted DALYs in Kenya were hydrocephalus (60.8%) and spina bifida (18.1%), whereas in Canada they were hydrocephalus (24.2%) and undescended testes (19.2%). A total of 23,169 DALYs were averted through 1,042 surgical procedures (22.2 DALYs per procedure) during the study period in Kenya, compared to 5,497 DALYs through 373 procedures (14.7 DALYs per procedure) in Canada.

Discussion

The current study compares the outputs of two pediatric surgical units in two very different populations, with very dissimilar resources. The countries were chosen non-randomly as sites of clinical activity of the authors, but are almost literally at the opposite ends of the spectrum of the Human Development Index [19]. What can we learn from such a comparison? Drilling down into

the detailed impact each unit has on the burden of surgical disease can help to better understand the training and resource needs in a lower-income country. The goal of improving pediatric surgical care in LMICs will not be met by copying the practices of a children's hospital in a developed country. Western surgeons who want to work with partners in developing countries must know what the needs there are, and whether they have the skills and experience to meaningfully contribute.

Settings and Practice patterns

The two settings compared in this study are both semi-autonomous, dedicated, academic pediatric surgical units, bringing together specialized physicians and other personnel, and involved in training multiple levels of health care providers. The similarities however stop there, as the infrastructure, resources, and practice of the units are very different. In general the Canadian site is much larger institution with more facilities and more dedicated surgeons (**Table 1**). As an example, the Canadian site includes a 60-bed fully-serviced neonatal Intensive Care Unit (ICU), while the Kenyan site only has access to a 20-bed nursery with no neonatal ventilators and no total parenteral nutrition.

In comparing the mortality and morbidity prevented through surgical procedures at the two sites, the first observation is that the two hospitals have very different patient populations. As ours is not a population-based study, the differences reflect to a large extent practice and referral patterns, rather than prevalence of disease. There are multiple factors likely involved in these patterns, including finances, transport, education, center reputation, and existing referral networks. BKKH is also a rather unique institution within a LMIC setting, providing high-quality specialty care at affordable prices through a significant reliance on expatriate physicians and donated equipment. While this model is different from that of the prevailing governmental institutions, it must be noted that there are in fact multiple non-governmental health care provision models in LMICs (private hospitals, church / mission hospitals, "niche" hospitals addressing selected specialized conditions, and temporary platforms) [24], and their combined impact towards the health care provision in many African countries is significant.

BKKH is also a national (and even African regional) referral centre for pediatric neurosurgery [25]: 70% of operative procedures at BKKH were for hydrocephalus or severe spina bifida. This might reflect the high prevalence of congenital neural tube defects in East Africa, largely preventable through better prenatal nutrition [26]. The prenatal incidence appears lower in North America, with the majority of such severe conditions identified prenatally leading to termination of pregnancy [26]. As an externally funded charity organization with North American-trained pediatric neurosurgeons and rehabilitation experts, BethanyKids fills a niche in treating these children who would otherwise often die untreated.

Conversely, the sum of annual pediatric surgical cases for 11 of the 13 selected conditions (excluding neurosurgery) is quite similar between MCH and BKKH (345 and 302, respectively). There are however several other differences in practice which stand out. For instance, BKKH recorded only one surgical procedure for intestinal atresia, whereas MCH had 10. This likely reflects delays in neonatal diagnosis of bowel obstruction in LMICs, as well as limited access to tertiary neonatal care required for such conditions, including ventilators and intravenous nutrition found in ICUs. The other difference observed is the large number of anorectal anomalies operated on at BKKH, which again likely reflects a referral bias, since BethanyKids is known to have this expertise, even when the initial life-saving colostomy surgery may have been done at other centers.

In the absence of the significant neurosurgical and general pediatric chronic surgical disabilities found in Kenya, the most common procedures captured at the Canadian site, accounting for 62% of the cases, were for mild hypospadias and undescended testis. These are generally less severe surgical conditions, accounting for only 29% of the total DALYs. This underscores the usefulness of the DALY metric, which offers a more complete picture of the impact of interventions on the BoD than is provided by the simple number of procedures.

Age at surgery

The comparison of age at time of surgery between the two sites reveals the overall older age of children operated on in Kenya. This is consistent with earlier work by the authors [9, 23], and thus believed to be a true reflection of delays in access to surgical care in LMICs. Only for three

health states were children operated in Canada at an older age: hydrocephalus, mild spina bifida, and abdominal wall defects. These three states include patients being operated on at an advanced age in Canada for mild defects, as reflected in the bimodal distributions of ages, with mode values of 0 for all.

Burden of disease

The next comparison involved calculating the DALYs averted per child for each health state. As shown in **Figure 3**, the mean averted DALYs per case were generally lower at BKKH, reflecting the shorter expected lifespan in Kenya, and therefore lower averted future disability. Only for two health states did procedures in Kenya result in a higher value of averted DALYs per case: severe hypospadias and undescended testis. This is accounted for by the higher DW assigned to these conditions in Kenya, possibly explained by a greater concern for male infertility [27].

The analysis of total averted DALYs for the 13 health states revealed the annual surgically averted BoD of BKKH to be 23,169 DALYs, four times that of MCH. This figure compares favorably with similar studies from adult limited-resource surgical settings: for instance Gosselin and Heitto estimated that in a district trauma hospital in Cambodia 3,786 DALYs were averted over a 3-month period through 895 surgical interventions, though many of these interventions were more minor [7]. Since the Kenyan site is operating with fewer staff and resources than the Canadian one, this comparison is striking and underscores the impact that surgical care can have in LMICs. Our study did not include a cost analysis, but it is known that the greater BoD averted through surgery in resource-poor settings is also very cost-effective [7-9, 23], thus further strengthening the key role of surgery in global health care. The addition of a cost-effectiveness analysis would increase the usefulness of the study for health policy and resource allocation, however obtaining accurate costing estimates for a hospital setting which receives substantial donations of equipment and other resources could be challenging.

The average impact of each procedure (22.2 DALYs in Kenya vs. 14.7 DALYs in Canada) is an indicator of both disease severity and local priorities. The main contributor to this difference is the higher DW attributed to undescended testis in Kenya. This is an example of how the DALY methodology can identify local priorities – male fertility appears to be more highly valued in

Kenya, thus resulting in a higher DW for undescended testes than in Canada [22]. Although the selected conditions are not a reflection of the entire spectrum of pediatric surgical practice in either institution (such common conditions as hernias, appendicitis and trauma were not included), the averted burden in Kenya is impressive in light of the resources available.

The BoD in DALYs averted through any given surgical intervention reflects the DW of the underlying health state, the age of the patient, and the effectiveness of the intervention. Thus, for instance, neurosurgical procedures are generally associated with higher mean averted DALYs than plastic surgical procedures. This however must not be interpreted as favoring one type of specialty care over another, as factors like costs of infrastructure, equipment and supplies, and expertise also vary widely among surgical specialties, and health care needs in LMICs are tremendous in all specialties.

The focus of this study was on the averted DALYs, or the *met* need. The avertable DALYs, or *unmet* need, is beyond the scope of this study as its estimation requires population-based data. The limited literature on the subject however suggests that this unmet need is very significant in LMICs: previous studies have estimated, for instance, that there is a backlog of over 300,000 uncorrected cleft lip cases in Africa alone [23], and that on average less than 15% of required surgical procedures for children are actually performed in a large African refugee camp [9]. Another relatively ignored component of the unmet need in surgery is that incurred through delay in care. This component, dubbed “delayed avertable DALYs”, has been found to be quite significant in LMICs [9, 23].

The DALY metric is most commonly used in LMIC settings, however DALYs have also been calculated in high-income country (HIC) settings. A WHO report used the DALY metric in a HIC as well as a LMIC setting to highlight the disparities and create a common comparison [28].

Research in HICs often uses Quality-Adjusted Life Years (QALYs) as a measure of the impact of interventions on surgical conditions. Robberstad investigated the utility of QALYs, DALYs and life years gained (LYs) on priority-setting [29]. The paper concluded that trade-offs in terms of quality and quantity are being made in health, especially where resources are scarce, and that both the DALY and the QALY, while representing different health states, are useful in priority-setting

wherever the resources are not sufficient to meet the needs of a population. This type of setting can be seen even within Canada and the United States, and DALYs could be an additional useful metric for assessing the impact of poor or delayed access to care in developed nations.

Training implications

The practice of sending someone from a LMIC to a HIC institution to train, and then expecting them to return home and replicate the HIC practice observed is a faulty training model. In light of the observed widely divergent practice patterns in LMICs, surgical training is best done locally, within the setting of the local disease patterns, and using local resources. Outside experts can provide specific subspecialty training and academic support, and short-term courses or rotations in HICs can complement and enrich this training. The local pediatric surgeon in LMICs requires a much broader range of experience and skills than does his or her HIC counterpart, since their practice may well include pediatric plastic surgery, urology and even neurosurgery in addition to the traditional scope of pediatric general surgery. BethanyKids models this philosophy of training, being part of the Pan-African Academy of Christian Surgeons (PAACS), an Africa-wide pediatric surgical training program with this stated goal – to train pediatric surgeons in Africa, for Africa [30].

Limitations

There are several significant limitations to the current study. In the first place, the comparison between the two sites is somewhat arbitrary in light of the widely divergent practice and referral patterns at each site. Pediatric surgeons in HICs mostly do pediatric general surgery, working alongside other pediatric surgical specialists – while in LMICs pediatric surgeons usually include urology and plastic surgery in their practice. In the absence of clearly set practice and referral guidelines, the nature of the work performed in any institution 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 site (MCH) may be fairly representative of other similar-sized pediatric surgical units in the developed world, the Kenyan site (BKKH) is quite unique in its focus and outputs in the developing world, and a pediatric surgical unit in a government hospital might provide a more useful comparison. The reality,

however, is that there is not one standard institutional type in LMICs, but a conglomerate of various approaches and health care venues. Thus caution must be exercised when attempting to generalize findings at any one site to the entire country or region.

The actual choice of the 13 health states was well-informed, but not random - representing the pediatric surgical conditions more prevalent at both sites and for which DW estimates existed. While most of these health states corresponded to a specific single surgical procedure, this was not always the case – as apparent in the use of colostomy in the management of children with Hirschsprung’s Disease in Kenya.

Finally, the calculation of DALYs also introduced a significant level of uncertainty. This is due to multiple assumptions and controversies in the disability weight estimates, universal vs. country-specific life expectancies, and the use of age-weighting and discounting [31]. Moreover, the *actual* impact of each intervention was not known as follow-up was not available or practical to include. While the PST component in theory accounts for this and is primarily predicated by the surgical condition, the diverse care environments of the two sites may lead to slightly different PST values, and this was not accounted for in the study.

The DALY calculation for health states requiring planned staged procedures is challenging and imperfect. While in principle the combined averted BoD through a one-stage protocol must be the same as for a 3-stage protocol for the same health state, the individual impact of each stage (such as a colostomy) is very hard to estimate, and no DW values are available in the literature for this. In our study we have accounted for the full impact of each staged intervention in the case of the 2 health states requiring staging (severe imperforate anus and Hirschsprung’s disease) – thus the total averted DALYs for these 2 health states may have been overestimated.

Conclusions

Despite the above limitations, the current study highlights both the widely divergent scope of practice of pediatric surgeons across the global South – North divide, and the significant impact of pediatric surgery in averting the global BoD of congenital anomalies in both settings. The harsh reality of pediatric surgical practice in resource-poor settings is that children with life-threatening neonatal conditions frequently die before reaching appropriate care facilities, while those with

non-threatening congenital conditions live with their disease often to an advanced age due to significant delays in access to surgical care. Thus the practice of the HIC pediatric surgeon focuses on the urgent treatment of neonatal conditions, while the LMIC surgeon often needs to treat children with elective disabling, unoperated chronic surgical conditions - in addition to a variable volume of neonatal emergencies depending on availability of urgent access to care. Furthermore, HIC pediatric surgeons wishing to operate in LMIC settings must understand the local scope of pediatric disease.

This study provides a unique comparative snapshot into global pediatric surgical practice, and utilizes the recent developments in BoD measurement to estimate the true impact of this practice using the DALY framework. A question that still remains is the appropriateness of the DALY metric in health policy setting. Future studies could look at the needs of health planners, decision makers, and health administrators to see if another health metric may be more applicable. Overall, this study opens multiple future research opportunities in other subspecialties for a global understanding of health measurement and impact of this measurement on health care practice and resource allocation.

References

- [1] UNICEF (2009) State of the World's Children 2009: Maternal and Newborn Health, UNICEF, New York, p. 18-23
- [2] Mathers C, Fat DM, Boerma JT (2008) The global burden of disease: 2004 update, World Health Organization, Geneva, p. 40-51
- [3] Howson C, Christianson A, Modell B (2008) Controlling Birth Defects: Reducing the hidden toll of dying and disabled children in low-income countries disease control priorities project. http://www.dcp2.org/file/230/dcpp-twpcongenitaldefects_web.pdf. Accessed 20 Jan 14
- [4] World Health Organization (2014) Congenital Birth Defects-Fact sheet N°370. www.who.int/mediacentre/factsheets/fs370/en/. Accessed 12 Feb 2014
- [5] Maulik PK, Darmstadt GL (2007) Childhood disability in low-and middle-income countries: overview of screening, prevention, services, legislation, and epidemiology. *Pediatrics* 120:S1-S55
- [6] Azzie G, Bickler S, Farmer D, Beasley S (2008). Partnerships for developing pediatric surgical care in low-income countries. *J Pediatr Surg* 43:2273-2274
- [7] Gosselin R, Heitto M (2008) Cost-effectiveness of a district trauma hospital in Battambang, Cambodia. *World J Surg* 32:2450–2453
- [8] McCord C, Chowdhury Q (2003) A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet* 81:83–92
- [9] Wu VK, Poenaru D (2013) Burden of surgically correctable disabilities among children in the Dadaab Refugee Camp. *World J Surg* 37:1536–1543
- [10] Kohler HP (2012) Copenhagen Consensus 2012: Challenge paper on population growth, PSC Working Paper Series, Philadelphia, p. 1-20
- [11] Ozgediz D, Poenaru D (2012) The burden of pediatric surgical conditions in low and middle income countries: A call to action. *J Ped Surg* 47:2305–2311

- [12] Lopez AD (2005) The evolution of the Global Burden of Disease framework for disease, injury and risk factor quantification: developing the evidence base for national, regional and global public health action. *Global Health* 1:1-8
- [13] Murray CJL (1994) Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bull World Health Organ* 72:429-445
- [14] Jamieson DT, Breman JG, Measham AR et al (2006) Disease control priorities in developing countries, Oxford University Press, New York, p. 2-25
- [15] Vos T (2009) Improving the quantitative basis of the surgical burden in low-income countries. *PLoS Med* 6:e1000149
- [16] Bickler S, Ozgediz D, Gosselin R et al (2010) Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg* 34:374–380
- [17] Nandi B, Mungongo C, Lakhoo K. A comparison of neonatal surgical admissions between two linked surgical departments in Africa and Europe. *Pediatr Surg Int* (2008) 24:939–942
- [18] The World Bank (2014) Fertility rate, total (births per woman). <http://data.worldbank.org/indicator/sp.dyn.tfrt.in>. Accessed 11 Feb 14
- [19] United Nations Development Programme. Human Development Index. <http://hdr.undp.org/en/statistics/hdi>. Accessed 12 Feb 14
- [20] The World Bank (2014). Physicians (per 1000 people). <http://data.worldbank.org/indicator/sh.med.phys.zs>. Accessed 12 Feb 14
- [21] World Health Organization (2012). WHO mortality database. http://www.who.int/healthinfo/mortality_data/en/index.html. Accessed 12 Feb 14
- [22] Poenaru D, Pemberton J, Frankfurter C et al (2013) Establishing disability weights for congenital paediatric surgical disease: a cross-sectional, multi-modal study. *Lancet* 381:S115
- [23] Poenaru D (2013) Getting the job done: Analysis of the impact and effectiveness of the SmileTrain program in alleviating the global burden of cleft disease. *World J Surg* 37:1562–1570

- [24] Shrime MG, Sleemi A, Ravilla TD. Charitable platforms in global surgery: A systematic review of their effectiveness, cost-effectiveness, sustainability, and role training. *World J Surg*. 2014 Mar 29 (epub ahead of print)
- [25] Gathura E, Poenaru D, Bransford R et al (2010) Outcomes of ventriculoperitoneal shunt insertion in Sub-Saharan Africa. *J Neurosci Pediatrics* 4:329–335
- [26] Warf B, *Pediatric Neurosurgery in the Third World* (2008) Principles and Practice of Pediatric Neurosurgery, Thieme Publishers, New York, p. 43-49
- [27] Inhorn MC (2004) Middle Eastern masculinities in the age of new reproductive technologies: male infertility and stigma in Egypt and Lebanon. *Med Anthropol Q* 8:162-182
- [28] World Health Organization (2009). Global health risks: mortality and burden of disease attributable to selected major risks report. http://www.who.int/healthinfo/global_burden_disease/global_health_risks/en/. Part 2, Figure 7 pg 10 Accessed June 18 14
- [29] Robberstad B. QALYs vs DALYs vs LYs gained: What are the differences, and what difference do they make for health care priority setting? *Norsk Epidemiologi* 2005; 15 (2): 183-191
- [30] Pollock JD, Love TP, Steffes BC et al (2011) Is it possible to train surgeons for rural Africa? A report of a successful international program. *World J Surg* 35:493–499
- [31] Lopez AD, Mathers CD, Ezzati M (2006) *Global burden of disease and risk factors*, Oxford University Press, New York, p. 399-405

Chapter 8

The Burden of Waiting: DALYs accrued from delayed access to pediatric surgery in Kenya and Canada

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Abstract

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 thus:

$$\text{Delayed DALYs} = DW * (\text{actual} - \text{ideal age at surgery}),$$

where DW=disability weight for each procedure, and ideal age at surgery derived by expert consensus.

Results: 1,208 first-time procedures in general surgery (338), neurosurgery (621), plastic surgery (93), and urology (156) were included (870 in Kenya, 338 in Canada). Delays were longest in general surgery (46 months), and longer in Kenya than in Canada in all specialties. The longest delays in Kenya were for orchidopexy (72 months) and posterior sagittal anorectoplasty (PSARP) (74 months), and in Canada for orchidopexy (40 months). Corresponding total delayed BoD was highest in general surgery (273 DALYs) and neurosurgery (159 DALYs), and higher again in Kenya than in Canada (484 cf. 84 DALYs). Mean delayed BoD was highest for PSARP (1.9 DALYs/procedure) and colostomy (1.7 DALYs/procedure) being the highest.

Conclusions: Estimating the 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.

Introduction

Congenital conditions are among the top ten leading causes of pediatric morbidity and mortality and are estimated to affect 7% of all births around the world [1]. This estimate translates into approximately 93 million children living with some form of moderate or severe disability [2]. Timely surgical intervention is known to prevent both the death and disability associated with many of these congenital anomalies, however lack of timely access to surgically trained healthcare providers, operating room facilities, and surgical supplies are some of the factors that impair the delivery of adequate surgical care, especially in resource limited settings [3]. This lack of access continues to exist despite recent economic analyses that demonstrate that surgery can be a more cost-effective use of scarce resources in reducing disease burden when compared to many other medical interventions and should be considered an essential part of public healthcare [4-6]. This delay in access to surgical care, especially in children with congenital anomalies, remains however an under-studied priority, even within the global burden of disease literature.

The Global Burden of Disease (BoD) Study, the gold standard in the field, employs the Disability-Adjusted Life Year (DALY) as the health metric of choice. This metric integrates measurement of both the morbidity and mortality of a specific health state and is the new standard framework for quantifying and evaluating the effect of different interventions based on their ability to avert disability [7]. The DALY measure combines Years of Life Lost (YLL) with Years Lost due to Disability (YLD), thus recognizing the disease burden associated with non-fatal yet disabling conditions [8, 9]. Included in the DALY calculation is a disease-specific assessment called a Disability Weight (DW). DWs are an empirically determined metric reflecting the decline of health associated with a certain health state, ranging between 0 (perfect health) and 1 (death) [10, 11]. Through the successful application of the DALY methodology previous studies have determined that surgical conditions account for 11% of the global BoD, and in Africa the surgical BoD is nearly double that estimated in North America [12]. Furthermore the Disease Control Priorities (DCP) study has established that congenital anomalies contribute 9% of the surgical BoD [13].

In terms of need for surgical care, BoD can be subdivided into 3 categories: *met need* (averted DALYs), *unmet need* (avertable DALYs) and *unmeetable need* (unavoidable morbidity and mortality even with intervention) [14]. Estimates of *averted* DALYs can be used to compare the cost-effectiveness of surgical procedures with other medical treatments, as mentioned above, while estimates of *avertable* DALYs primarily inform priority-setting and the allocation of limited health resources.

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).

Methods

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 has been presented in a recent publication [20].

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 co-morbidities.

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:

$$\text{Delayed DALYs} = DW * (\text{age at surgery} - \text{ideal age at surgery}),$$

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.

Table 1. Literature used to Determine Recommended Age at Surgery

Condition*	Recommended Age at Surgery	Publication Year	Source
Hypospadias	6-18 months	2009	European Association of Urology (EAU) and the National Guideline Clearinghouse (NGC) http://www.guideline.gov/content.aspx?id=12594
Cleft Lip with no Cleft Palate	3 months	2012	NHS http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx
Cleft Palate with or without cleft lip	6-12 months	2012	NHS http://www.nhs.uk/Conditions/Cleft-lip-and-palate/Pages/Treatment.aspx
Undescended Testes	12-18 months	2009	European Association of Urology (EAU) and the National Guideline Clearinghouse (NGC) http://www.guideline.gov/content.aspx?id=14430
Hirschsprung's Disease	at diagnosis		IPEG guideline reviewed with no mention of age http://www.ipeg.org/hirschsprungs/
Hydrocephalus	at diagnosis		American Association of Neurological Surgeons reviewed with no mention of age
Imperforate Anus	3-4 weeks of age		APSA http://www.pediatricsurgerymd.org/AM/Template.cfm?Section=List_Of_Conditions1&template=/CM/ContentDisplay.cfm&ContentID=4289
Spina Bifida	48 hours of life	2005	American Association of Neurological Surgeons
Intestinal Atresia	at birth		Expert consultation
Abdominal Wall Defect	at birth		Expert consultation

* "mild" and "severe" forms of hypospadias, spina bifida and imperforate anus are grouped together in the absence of distinct management protocols.

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®.

Results

The study included 1,423 surgical procedures in patients under 18 years of age within the identified 13 health states. After removing secondary procedures and those intentionally delayed for medical reasons, there were 1,208 surgical procedures left. The distribution of the 12 procedures across sites, genders and specialties is detailed in **Table 2**.

Table 2: Number of Consecutive Procedures by Site, Gender, and Specialty

	MCH			BKKH			Total
	F	M	Total	F	M	Total	
Abdominal wall closure	4	3	7	1	2	3	10
Anoplasty	0	2	2	0	1	1	3
Coloanal pull-through	3	2	5	1	9	10	15
Colostomy	0	0	0	7	15	22	22
Intestinal atresia repair	1	4	5	0	1	1	6
Orchidopexy	N/A	163	163	N/A	88	88	251
PSARP ¹	7	2	9	8	14	22	31
General Surgery Total	15	176	191	17	130	147	338
Spina bifida closure	3	3	6	139	144	283	289
VPS insertion/ETV ²	5	4	9	143	180	323	332
Neurosurgery Surgery Total	8	7	15	282	324	606	621
Cleft lip repair	7	11	18	18	30	48	66
Cleft palate repair	11	8	19	2	6	8	27
Plastic Surgery Total	18	19	37	20	36	56	93
Hypospadias repair	N/A	95	95	N/A	61	61	156
Urology Total	N/A	95	95	N/A	61	61	156
Grand Total	41	297	338	319	551	870	1208

MCH = McMaster Children's Hospital; BKKH = BethanyKids Kijabe Hospital; 1 PSARP = Posterior sagittal anorectoplasty; 2 VPS = Ventriculoperitoneal shunt; ETV = Endoscopic third ventriculostomy

The delay in surgical intervention was first estimated in months, and is detailed by procedure in **Table 3** and by specialty in **Figure 1**. As noted, mean delays in surgery were statistically

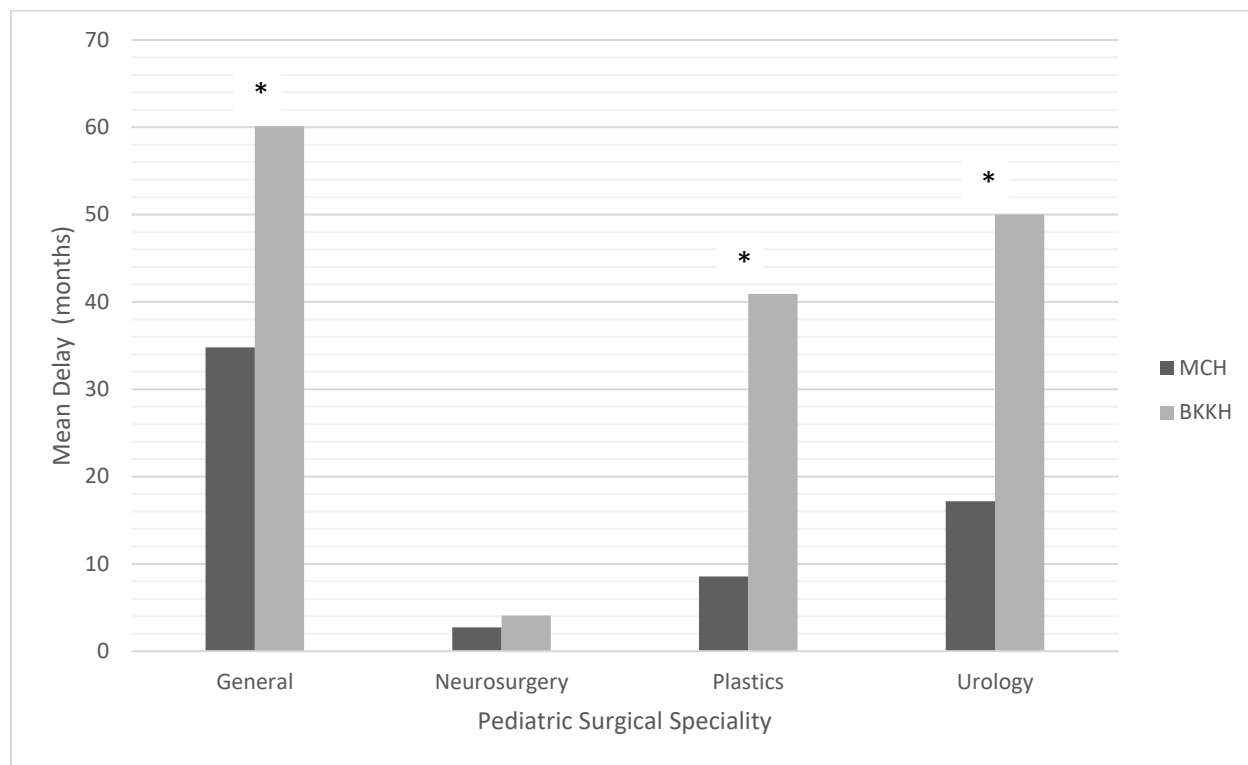
significantly higher in Kenya for most procedures which included sufficient data points, with the exception of hydrocephalus procedures which had similar mean ages across sites.

Table 3: Mean Delay to Surgery for each Procedure by Site (in months)

Procedure	MCH¹	BKKH	p value
Abdominal wall closure	0	0.1	0.05
Anoplasty	0	5.6	NS
Cleft lip repair	5.6	39.8	<0.001
Cleft palate repair	11.4	47.6	NS
Coloanal pull-through	13.0	30.3	NS
Colostomy	N/A	26.4	NS
Hypospadias repair	17.2	50.0	<0.0001
Intestinal atresia repair	0	0.1	NS
Orchidopexy	40.4	71.7	<0.0001
PSARP	0	74.4	<0.001
Spina bifida closure	0	4.5	<0.0001
VPS insertion/ETV	4.5	3.7	NS

MCH = McMaster Children's Hospital; BKKH = BethanyKids Kijabe Hospital; NS = Not significant; PSARP = Posterior sagittal anorectoplasty; VPS = Ventriculo-peritoneal shunt; ETV = Endoscopic third ventriculostomy; N/A = not applicable. ¹Age data from MCH was converted from years.

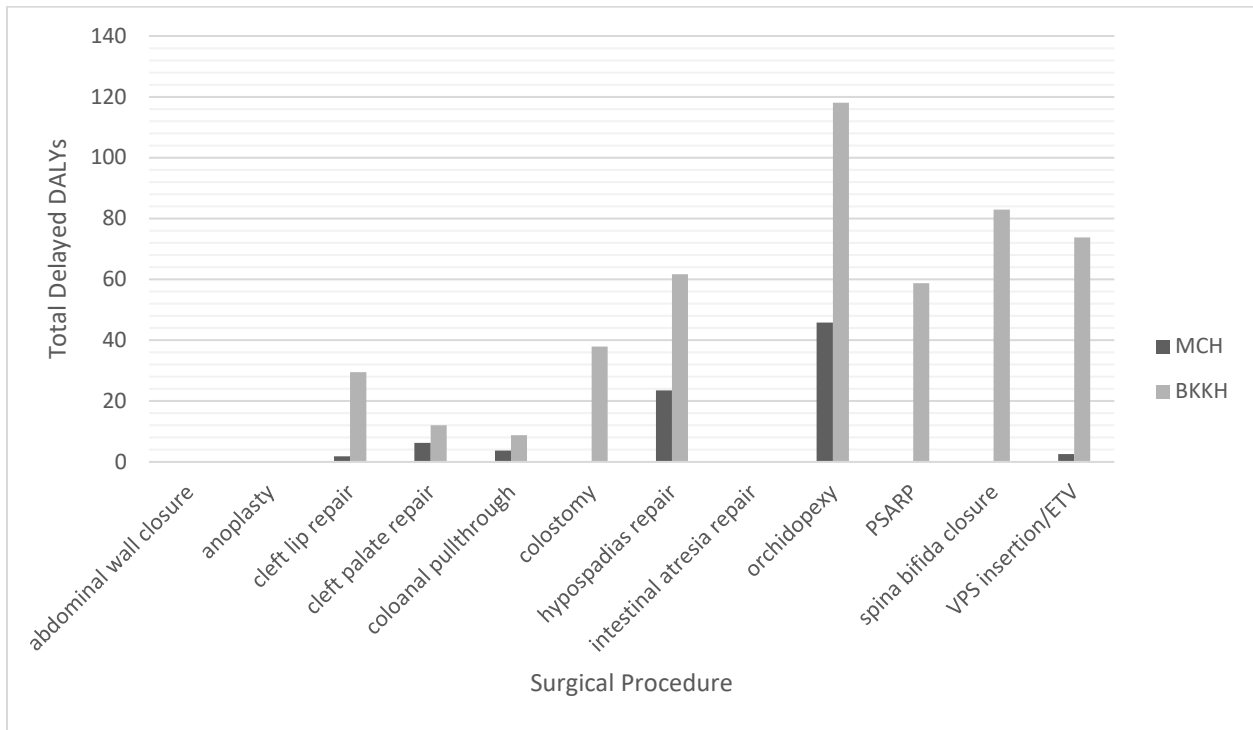
Figure 1: Mean Delay to Surgical Procedure for each Speciality by Site



MCH = McMaster Children’s Hospital; BKKH = BethanyKids Kijabe Hospital; * $p < 0.0001$

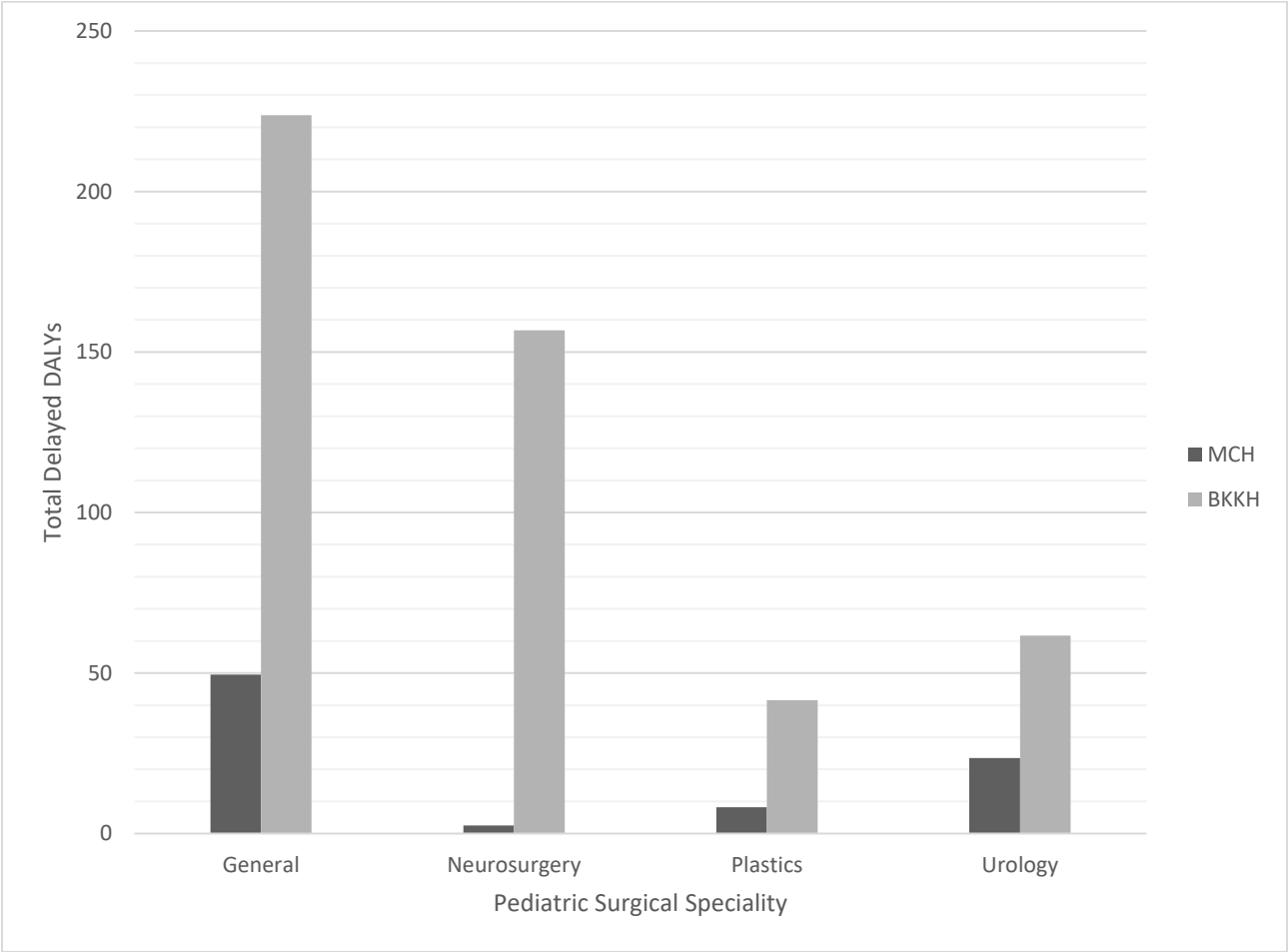
The cumulative DALYs resulted from this temporal delay were then estimated and depicted in **Figure 2** (by procedure) and **Figure 3** (by speciality). These cumulative values reflect both the predominance of neurosurgical procedures at BKKH, and the major delays at that site in the care of children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

Figure 2: Total Delayed DALYs Accrued by Procedure and Site



MCH = McMaster Children's Hospital; BKKH = BethanyKids Kijabe Hospital; PSARP = posterior sagittal anorectoplasty; VPS = ventriculo-peritoneal shunt; ETV = endoscopic third ventriculostomy.

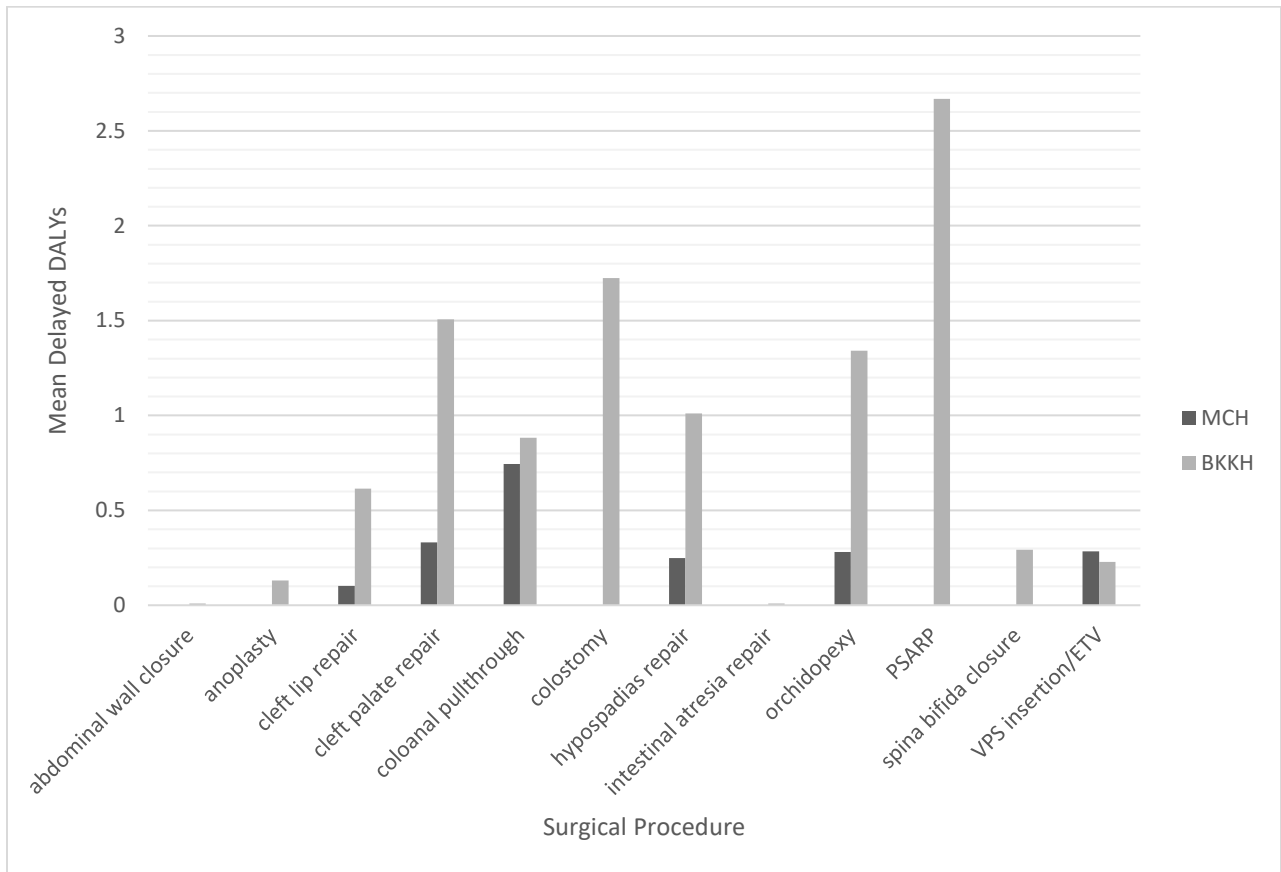
Figure 3: Total Delayed DALYs Accrued by Specialty and Site



MCH = McMaster Children’s Hospital; BKKH = BethanyKids Kijabe Hospital.

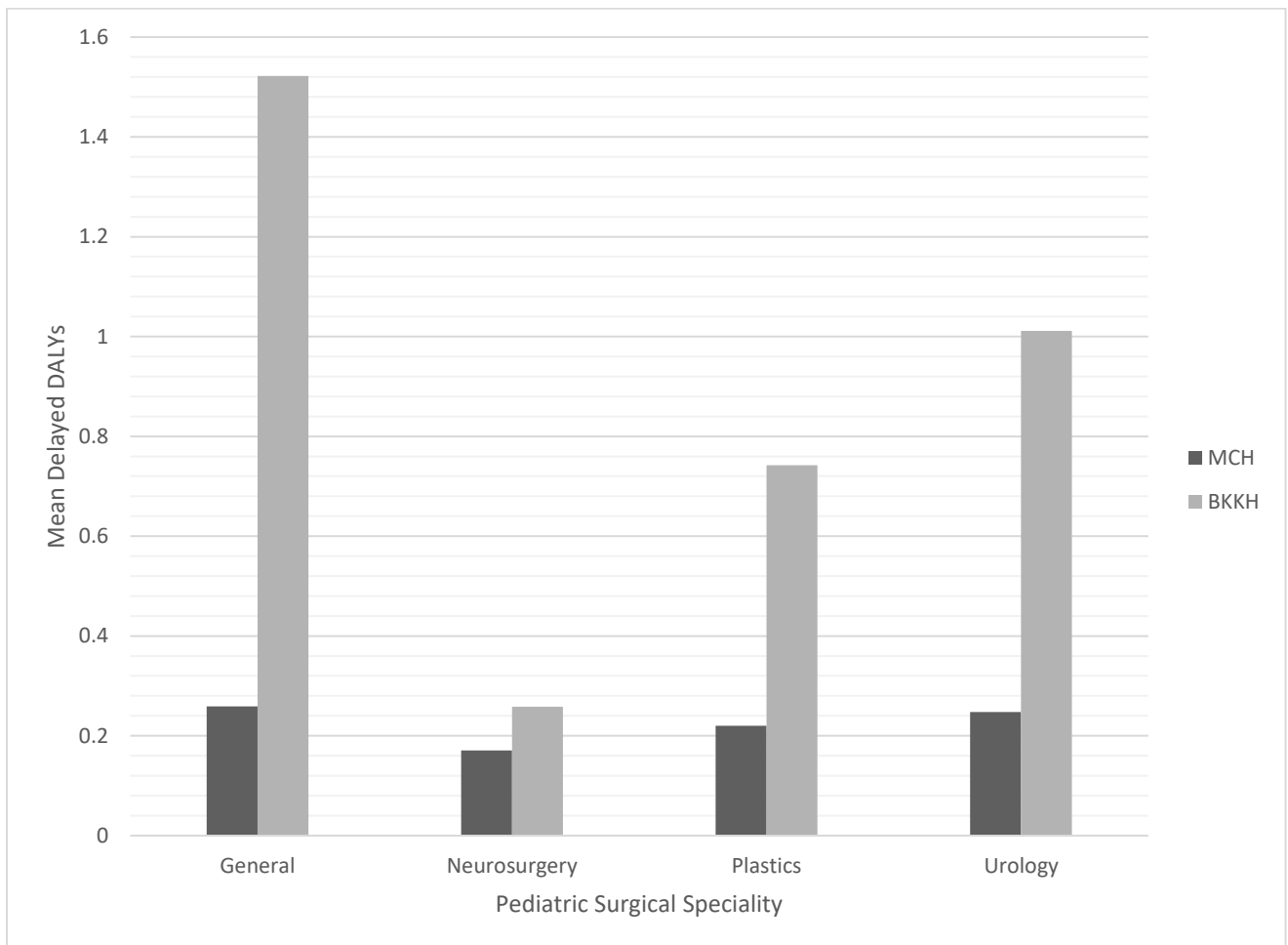
Finally, mean delayed DALYs incurred for each surgical procedure were also estimated by procedure type (**Figure 4**) and by specialty (**Figure 5**). These figures highlight the impact of delays in the care of children particularly with intestinal, urological, and plastic surgery conditions.

Figure 4: Mean Delayed DALYs per Surgery Procedure and Site



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

Figure 5: Mean Delayed DALYs per Surgical Specialty by Site



MCH = McMaster Children's Hospital; BKKH = BethanyKids Kijabe Hospital.

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 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 non-randomly as sites of clinical activity of the authors, and are nearly at the opposite ends of the spectrum of the Human Development Index [23]. This comparison aims to

highlight predictable inequities in access and delivery of surgical care between resource-rich and resource-limited settings.

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 neurosurgical procedures in BKKH, a reflection of a unique referral pattern established over the past decade in Kenya.

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 three years, for hypospadias four 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.

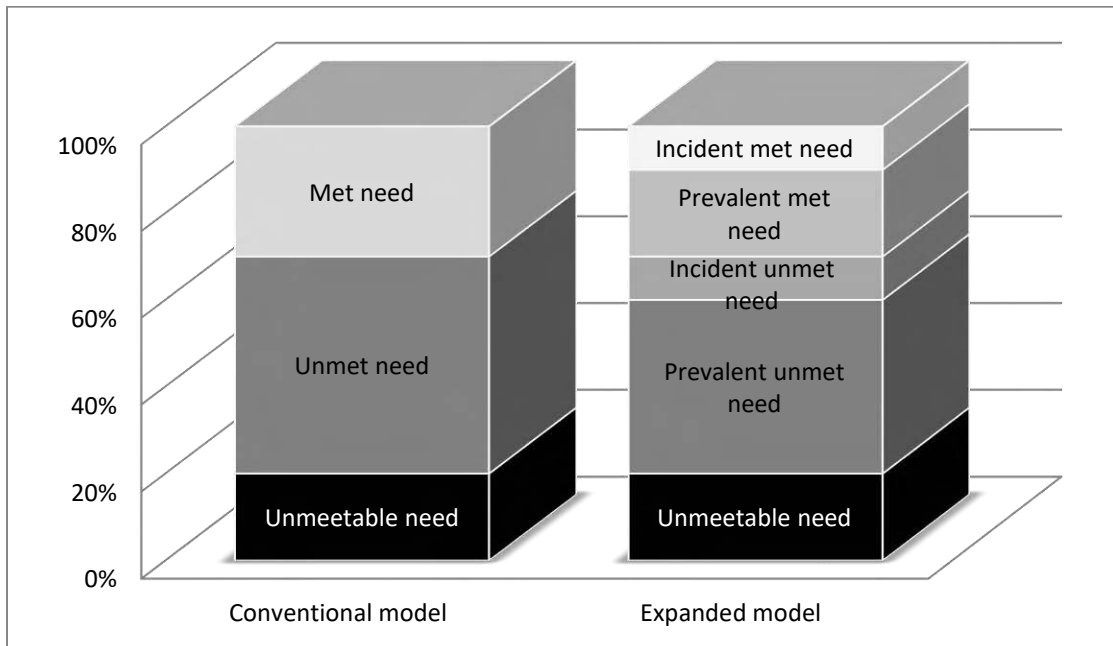
It is important to note that the delay observed at both sites in our study includes several components: delay of presentation to medical care, delay of referral for specialized care, and finally delay to actual surgery. Especially in HICs there are currently some major efforts underway to quantify and monitor the latter 2 delays (labelled 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.

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 Figures 2 and 3 are also modulated by institutional surgical volumes, thus reflecting of the actual delayed burden encountered in a high- and a low-income setting. The mean delayed DALYs per procedure, as shown in Figures 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 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 (**Figure 6**), the delayed burden fits within the prevalent need, as it results from children who have not been treated in a timely fashion.

Figure 6: The Conventional and Expanded Models for Components of Burden of Surgical Disease



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 multi-factorial, 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.

Implications for LMIC settings

The concepts of met and unmet BoD, access to care, effective coverage and backlog have only recently being 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 over 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.

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 waiting 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 waiting 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 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.

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 pre-selected 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 uncertainty. This is due to multiple assumptions and controversies in the DW estimates and age-weighting [10].

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

1. Mathers C, Fat DM, Boerma JT. The global burden of disease: 2004 update. World Health Organization 2008.
2. Unicef. The state of the world's children 2009. eSocialSciences.
3. Azzie G, Bickler S, Farmer D, Beasley S. Partnerships for developing pediatric surgical care in low-income countries. *J Pediatr Surg* 2008;43(12):2273-4.
4. Gosselin R, Heitto M. Cost-effectiveness of a district trauma hospital in Battambang, Cambodia. *World J Surg* 2008;32:2450–53.
5. McCord C, Chowdhury Q. A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet* 2003;81:83–92.
6. Wu VK and Poenaru D. Burden of surgically correctable disabilities among children in the Dadaab Refugee Camp. *World J Surg* 2013;37(7):1536–43.
7. Ozgediz D, Poenaru D. The burden of pediatric surgical conditions in low and middle income countries: A call to action. *J Ped Surg* 2012;47(12):2305–11.
8. Lopez AD. The evolution of the Global Burden of Disease framework for disease, injury and risk factor quantification: developing the evidence base for national, regional and global public health action. *Global Health* 2005;1(5):1-8.
9. Murray CJL. Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bull World Health Organ* 1994;72:429-45.
10. Lopez AD, Mathers CD, Ezzati M (2006) *Global Burden of Disease and Risk Factors*. Oxford University Press, New York.
11. Rehm J and Frick U. Valuation of health states in the US study to establish disability weights: lessons from the literature. *Int J Methods Psychiatr Res.* 2010;19(1):18-33. doi: 10.1002/mpr.300.
12. Jamison D. *Disease control priorities in developing countries*. World Bank Publications 2006.
13. Vos T. Improving the quantitative basis of the surgical burden in low-income countries. *PLoS Med* 2009;6:e1000149
14. Bickler S. Key Concepts for Estimating the Burden of Surgical Conditions and the Unmet Need for Surgical Care. *World J Surg* 2010;34:374–80.

15. Poenaru D, Ozgediz D, Gosselin R. Burden, need, or backlog: A call for improved metrics for the global burden of surgical disease. *International Journal of Surgery* 2014;2–5.
16. Poenaru D. Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease. *World J Surg* 2013;37:1562–70.
17. Alkire BC, Vincent JR, Burns CT, Metzler IS, Farmer PE, Meara JG. Obstructed labor and caesarean delivery: the cost and benefit of surgical intervention. *PloS One* 2012;7(4):e34595.
18. Warf, BC, Alkire BC, Bhai S, Hughes C, Schiff SJ, Vincent JR et al. Costs and benefits of neurosurgical intervention for infant hydrocephalus in sub-Saharan Africa. *Journal of Neurosurgery* 2011;8(5):509–21.
19. Corlew DS. Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. *World Journal of Surgery* 2010;34(3):391–6.
20. Poenaru D, Pemberton J, Frankfurter C, Cameron BH. Quantifying the Disability Averted through Pediatric Surgery: A cross-sectional comparison of a pediatric surgical unit in Kenya and Canada. *WJS* Submitted June 2014.
21. Bickler SW, Spiegel D. Improving surgical care in low- and middle-income countries: a pivotal role for the World Health Organization. *World Journal of Surgery* 2010;34(3):386–90.
22. Chirdan LB, Ameh EA. Untreated surgical conditions : time for global action. *The Lancet* 2012;380(9847):1040–41.
23. United National Development Programme. UNDP Human Development Index. Available from: <https://data.undp.org/dataset/Table-1-Human-Development-Index-and-its-components/wxub-qc5k>. Accessed June 12 2014.
24. Ugwu RO, Okoro PE. Pattern, outcome and challenges of neonatal surgical cases in a tertiary teaching hospital. *African Journal of Paediatric Surgery* 2013;10(3):226–30.
25. Chirdan LB, Ngiloi PJ, Elhalaby E. Neonatal surgery in Africa. *Seminars in Pediatric Surgery* 2012;21(2):151–9.
26. Warf, B. C. (2010). Pediatric hydrocephalus in East Africa: prevalence, causes, treatments, and strategies for the future. *World Neurosurgery*, 73(4), 296–3 00.

27. Canadian Pediatric Surgery Wait Times Project. http://www.ccyhc.org/work_surgical_projects.html. Assessed June 20, 2014.
28. Poenaru D, Pemberton J, Frankfurter C, Cameron B. Establishing disability weights for congenital paediatric surgical disease: a cross-sectional, multi-modal study. *Lancet* 2013;[381](#):S115.
29. Zafar SN, Fatmi Z, Iqbal A, Channa R, Haider AH. Disparities in Access to Surgical Care within a Lower Income Country: An Alarming Inequity. *World Journal of Surgery* 2013;[37](#)(7):1470–7.
30. Bowman KG, Jovic G, Rangel S, Berry WR, Gawande A. Pediatric emergency and essential surgical care in Zambian hospitals: a nationwide study. *Journal of Pediatric Surgery* 2013;[48](#)(6):1363–70.
31. Kushner AL, Groen RS, Kamara TB, Dixon-Cole R, Daoh KS, Kingham TP et al. Assessment of Pediatric Surgery Capacity at Government Hospitals in Sierra Leone. *World Journal of Surgery* 2012;[36](#):2554–58.
32. Grimes CE, Law RSL, Borgstein ES, Mkandawire NC, Lavy CBD. Systematic Review of Met and Unmet Need of Surgical Disease in Rural Sub-Saharan Africa. *World Journal of Surgery* 2011;[36](#)(1):8–23.
33. Rutherford, M. E., Mulholland, K., & Hill, P. C. (2010). How access to health care relates to under-five mortality in sub-Saharan Africa: systematic review. *Tropical Medicine International Health TM IH*, [15](#)(5), 508–519.
34. Gosselin R, Ozgediz D, Poenaru D. A Square Peg in a Round Hole? Challenges with DALY-based “Burden of Disease” Calculations in Surgery and a Call for Alternative Metrics. *World Journal of Surgery* 2013;[37](#)(11):2507–11.
35. Shengelia B, Tandon A, Adams OB, Murray CJL. Access, utilization, quality, and effective coverage: an integrated conceptual framework and measurement strategy. *Social Science & Medicine* 2005;[61](#)(1):97–109.
36. Groen RS, Samai M, Stewart KA, Cassidy LD, Kamara TB, Yambasu SE et al. Untreated surgical conditions in Sierra Leone: a cluster randomised, cross-sectional, countrywide survey. *The Lancet* 2012;[6736](#)(12):10–5.

37. Pemberton J and Cameron B. Essential Surgical Services : an emerging primary health care priority. *McMaster University Medical Journal* 2010;7(1):5–10.
38. Chirdan LB, Ameh E, Abantanga F, Sidler D, Elhalaby E. Challenges of training and delivery of pediatric surgical services in Africa. *Journal of Pediatric Surgery* 2010;45(3):610–8.
39. McQueen KK, Ozgediz D, Riviello R, Hsia RY, Jayaraman S, Sullivan SR et al. Essential surgery: Integral to the right to health. *Health and Human Rights* 2010;12(1):137–5.
40. Derrett S, Paul C, Morris JM. Waiting for elective surgery : effects on health-related quality of life. *International Journal for Quality in Health Care* 1999;11(1):47–57.
41. WHA. Improving Safe Emergency and Essential Surgical Care and Anaesthesia. In *Improving Safe Emergency and Essential Surgical Care and Anaesthesia*. 2004. Geneva.
42. GIEESC: WHO Global Initiative for Emergency and Essential Surgical Care. <http://www.who.int/surgery/globalinitiative/en/> . Accessed June 10, 2014.
43. International Collaboration for Essential Surgery (ICES). <http://essentialsurgery.com>. Accessed June 10, 2014.
44. Bellagio Working Group. Increasing access to surgical services in resource-constrained settings in Sub-Saharan Africa. *Bellagio Conference Center*. 2007:1-72.
45. Bae JY, Groen RS, Kushner AL. Surgery as a public health intervention: common misconceptions versus the truth. *Bulletin of the World Health Organization* 2011;89(6):394.
46. Ozgediz D and Riviello R. The “other” neglected diseases in global public health: surgical conditions in sub-Saharan Africa. *PLoS Medicine* 2008;5(6):e121.

Chapter 9

Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease

Poenaru D

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Abstract

Background

The study measured the success of SmileTrain, the largest cleft charity globally, in alleviating the global burden of disease (GBD). It was done by estimating averted disability-adjusted life years (DALYs) and delayed averted DALYs because of the global backlog in cleft procedures.

Methods

Anonymized data for all procedures in the SmileTrain global database were analyzed by age, sex, country, region, and surgery type. DALYs averted were calculated using life expectancy tables and established and estimated disability weights. The cost-effectiveness analysis used mean SmileTrain procedural disbursement figures. Sensitivity analysis was performed using various cleft incidence rates, life expectancy tables, and disability weights.

Results

During 2003–2010 a total of 536,846 operations were performed on 364,467 patients—86 % in Southeast Asia and the western Pacific region. Procedure numbers increased yearly. Mean age at primary surgery—6.2 years (9.8 years in Africa)—remained fairly constant over time in each region. Globally, 2.1–4.7 million DALYs were averted through the operations at a total estimated cost of US\$196 M. Mean DALYs per patient were 3.8–9.0, and mean cost per DALY was \$72–\$134. Total delayed GBD due to advanced age at surgery was 191,000–457,000 DALYs.

Conclusions

Despite an unparalleled number of surgeries performed and yearly increase by one charity, the unmet and delayed averted cleft GBD remains significant in all regions. Large geographic disparities reflect varied challenges regarding access to surgery. Cleft surgeries are cost-effective interventions to reduce the global burden of disease (GBD). Future challenges include increased collaboration among cleft care providers and a focus on remote global areas by building infrastructure and local training.

Introduction

Cleft lip and palate is a relatively common congenital anomaly across the world, with a variable incidence worldwide of 1 in 300–1 in 500 new births [1]. Although cleft lip is a disfiguring condition and cleft palate carries significant morbidity in terms of feeding and speech development, their mortality rate is generally low. Surgical correction is relatively straightforward but requires surgical specialty expertise and, particularly in the case of cleft palate, good postoperative facilities to avoid potentially deadly early complications.

In low- and middle-income countries (LMICs) large numbers of children with clefts remain untreated for many years, even into adulthood [2]. This situation has created a significant, yet unquantified surgical backlog in many LMICs.

SmileTrain is the world's leading charity (by numbers treated) providing care for patients with cleft lip and palate. Its stated mission is “to provide a child born with a cleft the same opportunities in life as a child born without” [3]. It pursues this mission by providing free cleft surgery across the world (more than 700,000 surgical interventions over 12 years in 84 countries) and training medical professionals (more than 1,400 medical conferences). It also offers rehabilitative care such as speech therapy and orthodontics. The unique approach of SmileTrain is based on building local surgical capacity through training and funding: currently there are more than 2,300 national partner surgeons in more than 1,100 partner institutions.

The metrics of GBD, expressed in disability-adjusted life years (DALYs), have become essential for setting priorities, particularly in LMICs [4, 5]. Within global surgery, the burden of disease has been divided into met need (surgical care already provided, or DALYs averted), unmet need (potentially avoidable/treatable disability and death due to surgical conditions, or DALYs avertable), and unmeetable need (disability and premature death that is unavoidable, even with the best surgical care) [4, 6]. When combined with GBD measurements, cost-effectiveness analysis can generate \$/DALY values for specific interventions.

This study estimates the averted and delayed averted global burden of cleft disease and the cost-effectiveness of the work of SmileTrain using GBD methods.

Materials and methods

Data collection

The main data set was extracted and anonymized from the current SmileTrain database of all cleft surgical procedures, obtained directly from and with courtesy of Smile- Train. Data accessed in Excel format included age, sex, country, region, and type of surgery. Countries were assigned to world regions according to the World Health Organization [7]. Only first-time surgical procedures were included in age calculations.

Effective coverage rates for cleft surgery in each country were calculated for the year 2010 using available country-specific population and crude birth rates. A sensitivity analysis was performed using three incidences for cleft lip/ palate: 1:700 live births (average global value); 1:300 (maximum average incidence); and 1:1,500 (minimum average incidence) [1]. Naturally, in many countries other cleft charities contribute to the work, although generally their impact is small relative to that of SmileTrain. Unfortunately, there are no global data on combined cleft interventions, so our calculations are limited to the current data set.

The backlog in cleft surgeries in each country or region was estimated using the following formula:

$$\text{Backlog} = (\text{actual} - \text{ideal age at surgery}) * \text{population} * \text{crude birth rate} * \text{cleft incidence}$$

This formula was based on the observation that, on a national or regional level, each year of advanced age at surgery (over and above the ideal age) represents a full cohort equal to the number of new cases during that year. For instance, an average age of 3 years at surgery corresponds to a backlog of 2.5 yearly cohorts. The age cohorts would be reduced by any deaths—a factor that is presumed low in high-income countries but remains unknown in LMICs.

Again, a sensitivity analysis was performed using the three incidences. DALYs

Burden of disease estimates were calculated using previously established methods and country-specific life tables [8]. DALYs were calculated as the product of years lived with disability (YLD) and disability weight (DW). Although DW values exist for only a few congenital surgical

conditions, they are fortunately available for primary cleft lip and palate [8]. DWs for other cleft-related procedures were estimated in the present study using paired comparisons with the available weights.

Averted DALYs (met need) were calculated using both country-specific [9] and universal [10] life expectancy tables. A sensitivity analysis was also performed using global average DWs (0.049 for cleft lip and 0.101 for cleft palate [8]) and treated/untreated DW estimates from the original GBD tables [5]. DWs were adjusted by the estimated success of surgery and risk of permanent disability as suggested by McCord and Chowdhury [11] and modified by Gosselin et al. [12, 13]. The types of surgical procedures with their corresponding DALY formula factors are listed in **Table 1**.

Table 1: Factors for calculating DALYs averted for each surgical procedure

	Disability weight	Risk of permanent disability without treatment [11Error! Bookmark not defined.]	Probability of successful treatment [11]
Unilateral cleft lip repair	0.05	1	1
Bilateral cleft lip repair	0.05	1	1
Cleft palate closure	0.1	1	0.7
Cleft lip revision	0.05	1	1
Closure palatine fistula	0.05	1	0.7

Severity of disease / risk of permanent disability without treatment: 1 for >95%, 0.7 for 50-95%, 0.3 for 5-50%, 0 for <5%. Effectiveness of treatment: 1 for >95%, 0.7 for 50-95%, 0.3 for 5-50%, 0 for <5%.

Age weighting and discounting [8] are common practices in GBD studies, but they remain somewhat controversial, especially when applied to children [14–16]. The assumption, for instance, that a cleft disability exists only in children <5 years of age is clearly suspect [17]. In LMICs there are literally thousands of older patients (even into their 60s) who continue to suffer from this disability. The author therefore chose to do a sensitivity analysis on life expectancy tables using both plain values without any age weighing or discounting [the so-called YLL(0,0)] and standard YLL(3,1) tables, denoting 3 % discounting of future years and applied age weighing [8]. To avoid crowding charts with all the potential values from these three sensitivity analyses for each DALY, we chose three permutations that provided a minimum, maximum, and average DALY value.

Delay in surgical care leads to a hitherto unestimated burden of disability. This “delayed averted burden” is probably a component of the unmet need, although the actual time lived with disability before provision of care is unrecoverable. We have attempted to estimate it using the formula:

$$\text{Delayed averted burden} = (\text{actual} - \text{ideal age at surgery}) \times \text{DW}$$

The actual age naturally applies only to the first surgery performed for each child. The ideal age is based on conservative international surgical standards of practice (6 months of age for cleft lip repair, 12 months for cleft palate). Two separate sets of estimates for delayed averted burden were derived based on the DW sensitivity analysis.

Cost-effectiveness analysis

The cost-effectiveness analysis (CEA) was performed using mean averted DALYs per procedure and the average SmileTrain global reimbursement rates for cleft surgery. These reimbursement rates varied slightly by country but not by type of procedure. They were set to cover all surgery-related expenses with no cost to the patient. Only primary cleft lip and palate repairs were included in this calculation. Again, three sets of \$/DALY values were derived for each region, corresponding to the three sets of averted DALY values previously calculated.

Statistical analysis

Simple descriptive statistics (totals and averages) were applied to the demographic variables, DALYs, and CEA results using Microsoft Excel software.

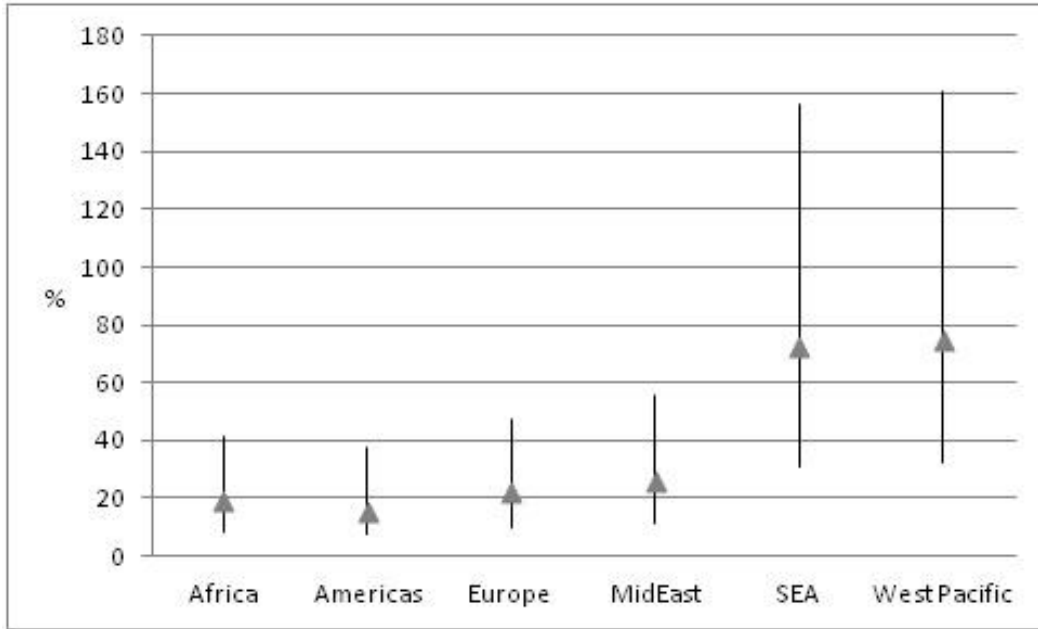
Results

During 2003–2010, a total of 536,846 operations were performed in 79 countries (**Fig. 1**) on 364,467 patients. The geographic locations of the procedures are detailed in **Table 2**. The ratio between numbers of cleft lip/palate patients operated on in 2010 and the expected number of new cases during that year is shown in **Fig. 2**. The high and low points of each bar correspond to the lower and upper extremes of incidence assumed. SmileTrain therefore performed that year approximately 36% of the yearly rate of new cleft surgeries needed in LMICs. In only five of the countries (four in Asia and Djibouti in Africa) was the number of surgeries performed equal to or larger than the mean yearly incidence of cleft lip/palate. The rare values over 100% simply reflect SmileTrain actually starting to address the unoperated cleft backlog.

Fig. 1: World map shows countries with SmileTrain presence during the study



Fig. 2: Range of percentage of expected new cases of cleft lip/palate operated on during 2010 by SmileTrain, by region.



Upper limit: 1:1,500 incidence; lower limit: 1:300 incidence; arrowhead marker: 1:700 incidence; SEA: Southeast Asia

Table 2: Numbers of cleft procedures by year and region

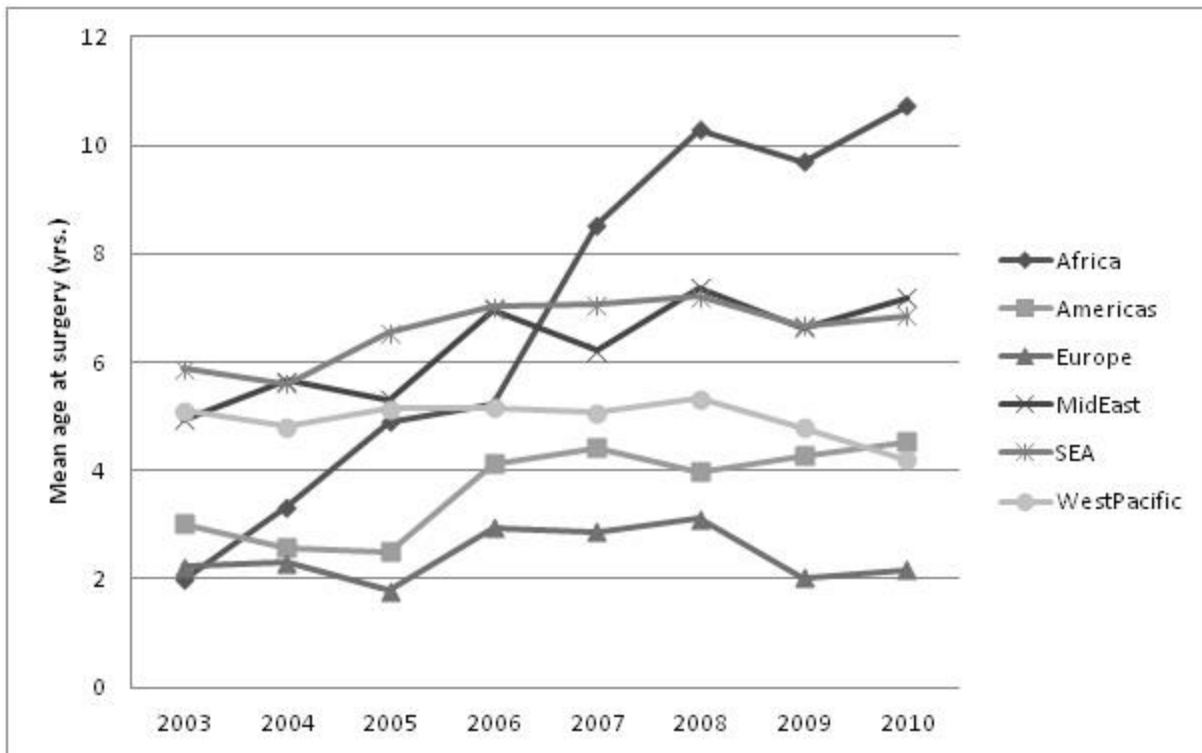
	Africa	Americas	Eastern Europe	Middle East	SEA*	West Pacific	Grand Total
2003	91	687	139	35	10630	13349	24931
2004	156	717	122	732	12256	13137	27120
2005	378	883	143	456	17732	16701	36293
2006	775	979	155	1497	27640	20388	51434
2007	3201	4536	245	1706	37680	27279	74647
2008	5272	6583	166	4629	48775	38445	103870
2009	6859	6431	904	5622	48317	35861	103994
2010	9973	7386	2163	8385	52237	34413	114557
Grand Total	26705	28202	4037	23062	255267	199573	536846

* SEA = south-east Asia

The sex of the patients was predominantly male, with 38.3% females (range 36.7 % in the western Pacific region–40.9 % in Africa). Patients with cleft lips with or without cleft palate were 36 % female, and those with isolated cleft palates were 48 % female.

The mean age at primary surgery was 6.2 years, with the highest age (9.8 years) in Africa, and the lowest (2.2 years) in eastern Europe ($p < 0.001$). The fluctuations in mean age in each region over time are depicted in **Fig. 3**. Similarly, most individual countries revealed no significant change in mean age over the 8-year period. The only notable exception was China, where the mean age decreased from 5.1 to 4.1 years.

Fig. 3: Mean age at primary surgery, by year and region



SEA: Southeast Asia

The size of the global backlog in cleft surgeries was estimated using various cleft incidences of 902,581, 2,106,021, and 421,204 patients (mean 1,143,269 patients). The distribution of this backlog by region for each incidence is illustrated in **Fig. 4**.

Fig. 4: Size of global backlog in cleft procedures, by region

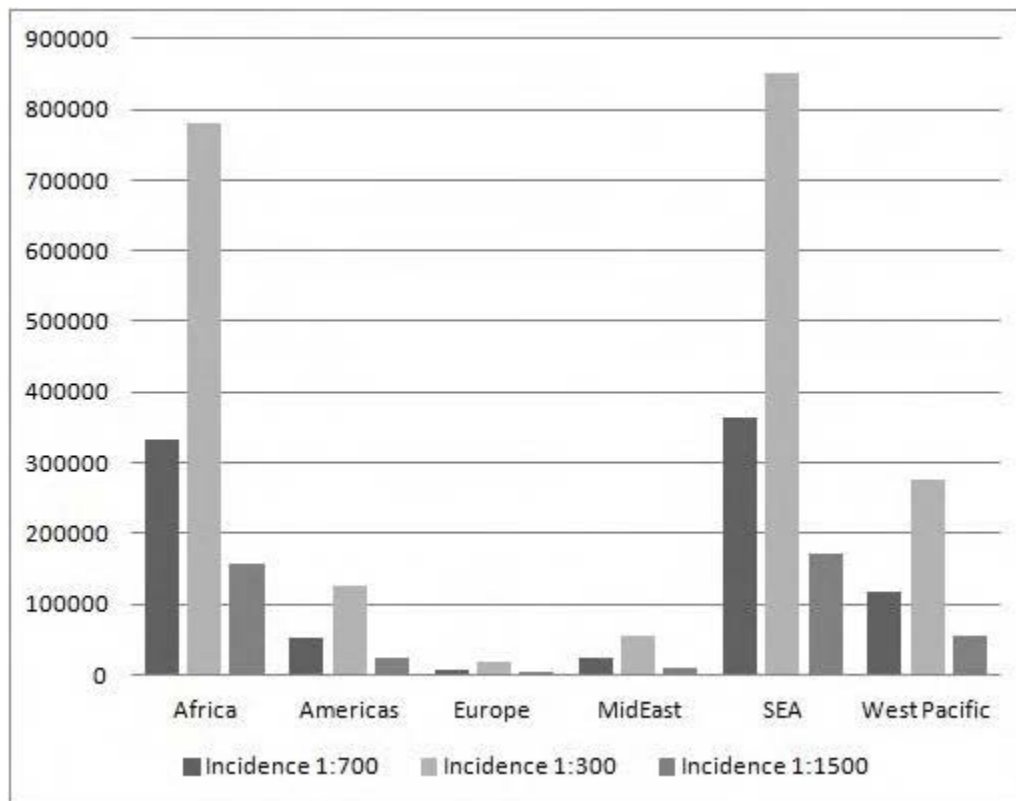


Table 3 shows the distribution of types of surgeries by region and year. Significantly, Africa has a great preponderance of cleft lip over cleft palate repairs compared to other regions ($p = 0.021$).

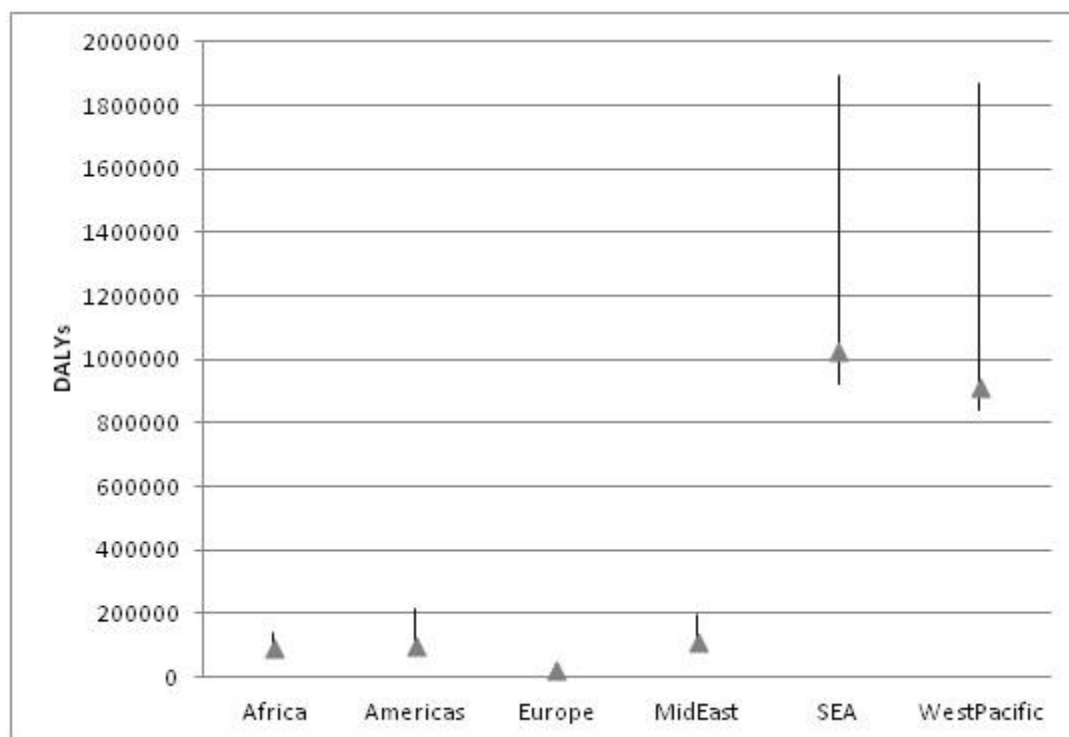
The sensitivity analysis permutations selected yielded 2.1, 4.7, and 2.3 million DALYs averted through the surgeries over the 8-year period, with an average value of 2.9 million. The range of burden of disease values generated by each method is depicted in **Fig. 5**. The largest component of the burden of disease (86 %) was averted in Southeast Asia and the western Pacific region. Similar methods used to derive averted DALYs per procedure yielded 3.8, 9.0, and 4.7 DALYs (mean 5.8 DALYs). This mean value was lowest in Africa (3.7 DALYs) and highest in eastern Europe (6.7 DALYs).

Table 3: Distribution of surgical procedures by region, in %

	Africa	Americas	Eastern Europe	Middle East	SEA	West Pacific
Unilateral CL	71.3	29.5	28.9	40.8	46.5	37.1
Bilateral CL	12.2	8.3	7.7	8.2	8.2	6.6
CP	10.9	34.8	47.8	38.8	31.6	43.2
Revision CL	3.9	21.0	12.5	10.5	10.0	11.0
Fistula closure	1.7	6.3	3.2	1.7	3.7	2.1

CL = cleft lip; CP = cleft palate; SEA = South-East Asia

Fig. 5: Averted cleft disability-adjusted life years (DALYs), by region.

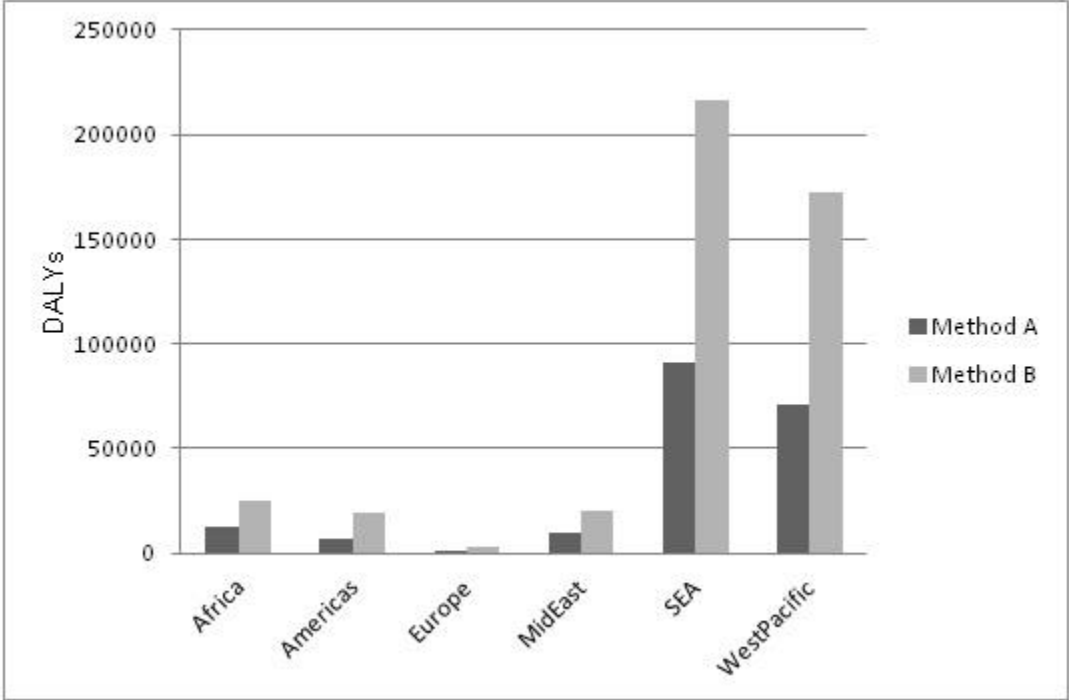


Estimate A: $YLL(0,0) * \text{global average disability weights (DWs)} * \text{risk of permanent disability} * \text{effectiveness of treatment}$. Estimate B: $YLL(0,0) * \text{DCP1 DWs without/with surgery}$. Estimate C: $YLL(3,1) * \text{DCP1 DWs without/with surgery}$

The total delayed averted burden of cleft disease due to advanced age at surgery by region is shown in **Fig. 6**. The total burden over 8 years was estimated by two methods at 190,910 and 456,625 DALYs, respectively (average 323,768 DALYs). The distributions of the delayed averted

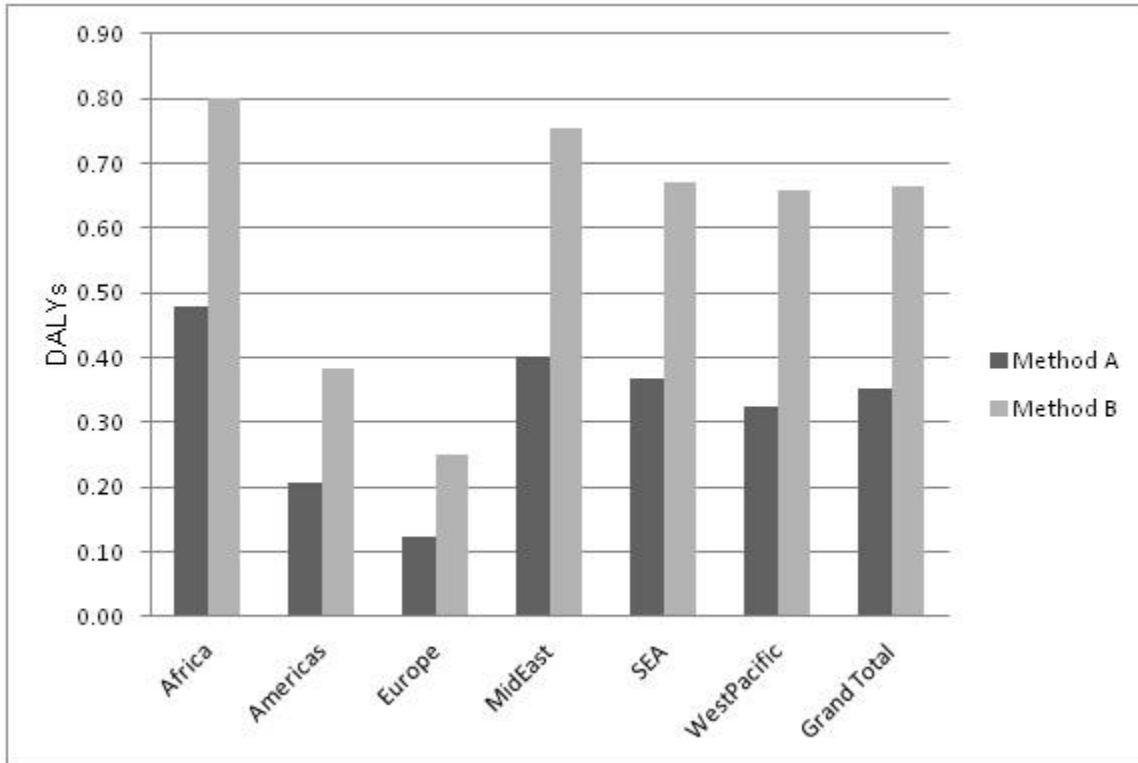
burden by year and sex were similar to those of the averted DALYs. **Figure 7** demonstrates that the mean delayed averted burden per patient, regardless of the method used, correlated directly with the age at surgery, being highest in Africa and lowest in eastern Europe.

Fig. 6: Delayed averted total DALYs derived using two methods, by region.



Method A: using average global DWs. Method B: using DWs with/without treatment

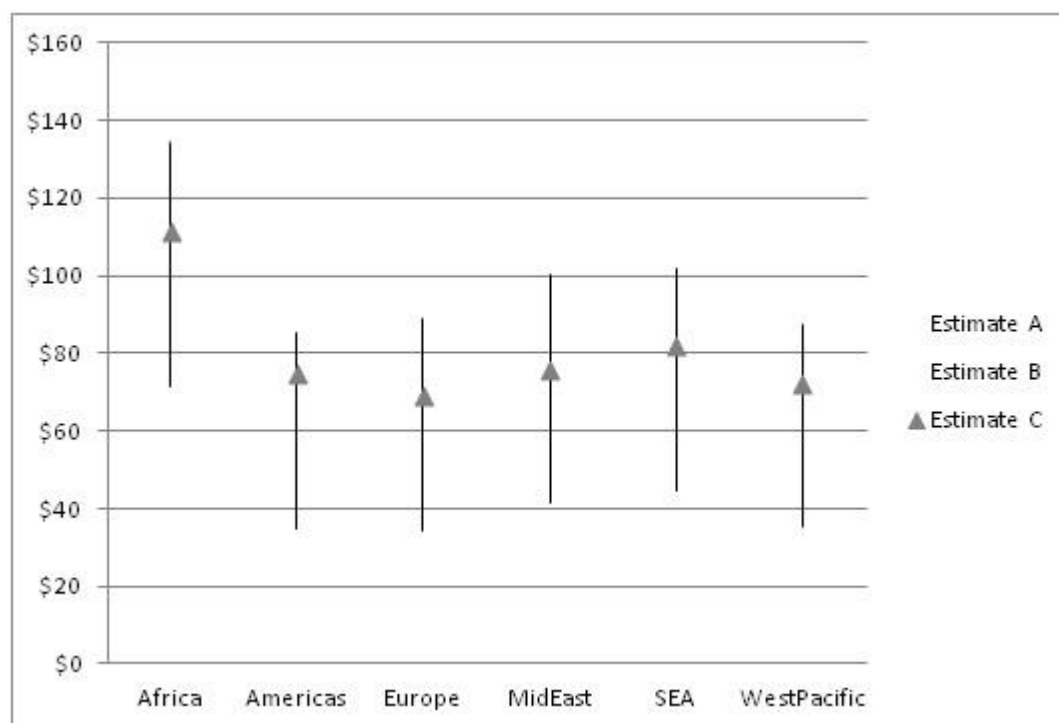
Fig. 7: Delayed averted burden of cleft disease per patient derived using two methods, by region.



Method A: using average global DWs. Method B: using DWs with/without treatment

The total cost estimated for the procedures performed was US\$195,948,790. The sensitivity analysis carried out for averted DALYs per procedure was then used to calculate the mean cost per DALY, resulting in values of \$100, \$44, and \$81, respectively. The cost per DALY by region is charted in **Fig. 8**, which shows the cost to be highest in Africa.

Fig. 8: Range of dollars per DALY derived using three methods, by region



The same methods used in Fig. 5 were used here.

Discussion

Using the GBD framework, initial estimates suggested that 14 million DALYs, or about 9 % of surgical DALYs and 1 % of all DALYs, are due to congenital conditions [6]. Subsequent work estimated averted DALYs through individual hospitals and surgical missions [11–13]. Although less useful for health systems planning than population-based data, this approach documented the impact of specific interventions and generated simple cost-effectiveness analyses. Though not population-based, the current study is truly global: In 2010, SmileTrain treated between 13 and 66 % of all new cleft cases in its 79 countries of activity.

Table 2 shows that SmileTrain-funded surgical procedures have steadily increased in all regions. The vast majority (84.7 %) were done in Southeast Asia and the western Pacific. These regions are densely populated, and the infrastructure and human resources allow scaling up of programs. As SmileTrain primarily funds procedures by local surgeons, countries with a critical mass of surgeons can increase significantly their surgical output with external funding. Consequently, Fig.

2 shows that Asian countries had the highest ratio of repaired versus estimated new cleft lip/palate cases (73.8 %), regardless of the calculation method. In contrast, fewer than 20 % of new cases in Africa were repaired in 2010, despite a huge backlog. This is likely multifactorial: limited availability of surgeons and adequate facilities, poor communication and access to information and care in remote areas, and administrative challenges.

The sex split in procedures favors males (62 %). This is consistent with the literature for cleft lip with or without palate (60–67 %) but is higher than expected for isolated cleft palate, where the expected male ratio is only 33 % [18]. In some areas, this suggests that sex bias may affect families' decision to seek care for children with less visible deformities (cleft palate).

The average age at surgery for any congenital condition ideally repaired soon after birth can be used as a surrogate marker of “surgical backlog” (the number of unoperated children/adults in a population who are older than the ideal age for that procedure). Predictably, African patients were the oldest at surgery (mean 9.8 years) and European patients the youngest. In fact, many cleft lip repairs in Africa and other LMICs were performed on senior citizens.

The size of the global backlog in cleft surgeries was estimated by three different methods to be between 421,000 and 2.1 million—a tremendous challenge for any cleft intervention. The backlog distribution mirrors not only population trends but also effective coverage for the condition. Figure 4 shows that Africa and SEA have the most severe problem. Addressing the backlog is complicated by the increasing surgical challenge and worsening functional results of cleft palate repairs in patients with increasing age [2].

The natural expectation is that at any given site the age distribution will steadily decrease over time as the backlog is addressed. Although this may indeed be happening in some individual institutions [19], it was not observed at any country or regional level except China. With more than 26,000 primary cleft surgeries performed in 2010, China is probably one of the few countries where SmileTrain and other organizations are at least beginning to address the global cleft backlog.

In contrast to other regions, age distribution in Africa actually increased from 2.0 to 10.7 years (Fig. 3). This is likely because early in each program procedures were performed at national

centers of expertise, where the mean age of patients was already quite low [20]. The program then expanded to remote areas, with much older patients. Another factor for the age plateau is the continuous opening of new programs in each country, generating a mixed picture nationally and regionally.

The balance between cleft lip and palate procedures in Asia, the Americas, and eastern Europe is similar, resembling incidence statistics for the condition [19]. For several likely reasons, Africa has a significantly lower rate of cleft palate repairs, as reported previously [2, 21–23]. First, cleft palate, unlike isolated cleft lip, can be fatal in infants, thus creating a “hidden mortality” [24], resulting in fewer cleft palate operations. In addition, patients are more likely to access care for a visible, disfiguring condition (cleft lip) than for a hidden, functional one (cleft palate). In fact, Wilson and Hodges [24], as well as others, have suggested performing the palate surgery with or even before the lip surgery, thereby increasing the chance of patients returning for the second surgery [2, 21]. Finally, surgery for cleft palate is more challenging than for cleft lip, requiring better facilities and postoperative care. As SmileTrain disbursement rates are identical regardless of procedure, the incentive to repair palates is diminished [19].

Bickler et al. [4] have suggested that burden of surgical disease can be divided into met, unmet, and unmeetable need. Our study focused on the met need (avertable DALYs), generally the only component estimable without population-based data. DALY calculations confirmed the large burden of disease averted through SmileTrain funding. Whereas the DALY values correlated closely with numbers of procedures, the mean DALY values per procedure were lowest in Africa. This was because of the older patients (fewer averted years left to live with disability) and lower life expectancy in the region. Overall, most DALYs were averted in Southeast Asia and the western Pacific, consistent with the literature [6]. In 2010, an estimated 308,476–638,030 DALYs were averted in those two regions, corresponding to 4.4–9.1 % of the 7 million DALYs estimated by the Bellagio Working Group for all congenital surgical conditions in those regions [25].

Our data also allowed estimation of the delayed averted burden caused by the YLD before surgery. This previously unidentified factor is part of the unmet need, although its irrecoverable nature makes it behave more like an unmeetable need. Not only is past suffering not recoverable, but in the case of many pediatric surgical conditions delay in care may be associated with more

difficult procedures and higher likelihood of complications. A 20-year-old patient with unoperated cleft palate will not only have needlessly suffered for 19 years from the sequelae of her condition, but her surgery will be more challenging than in a 1-year-old, and her speech deficits likely irreversible.

This study provides the first minimum estimate of the delayed averted burden of disease and highlights its magnitude. The general lack of decrease in mean delayed averted DALYs in all regions (and actual increase in Africa) is explained by the significantly less than 100 % of new cleft cases repaired each year by SmileTrain. Singh estimated the cleft backlog in India at approximately 1 million cases, which would take 100 years to clear if 15,000 surgeries were performed yearly above the cleft incidence [26]. Our estimates of backlog were more conservative (e.g., between 233,000 and 544,000 in India), but the magnitude remains significant. Other cleft charities also operate in most countries but with limited additional impact on the overall backlog.

Cost-effectiveness analysis has been a natural companion to GBD work. Several recent publications explored the cost-effectiveness of short-term surgical missions [11–13]. In cleft work, Magee et al. [17] assessed the cost-effectiveness of eight Operation Smile missions, costing primary cleft surgeries at \$796 (using standard life tables) or at \$34 after extending disability beyond the age of 5 years. They included only direct costs incurred by the mission, with no mention of institutional or patient costs. Corlew [16] compared two methods of estimating the economic impact of cleft repairs (gross national income per capita and value of a statistical life), documenting favorable cost-effectiveness.

Cost-effectiveness analysis in this study relied on a simple, preset reimbursement rate for all cleft procedures, rather than actual costs incurred. Figure 8 highlights the significantly higher cost per DALY in Africa compared to other world regions. This is likely due to the artificial “leveling of the playing field” through similar SmileTrain reimbursements globally combined with the lower number of averted DALYs per patient in Africa. Moreover, health care costs in Africa are often surprisingly high compared to those in other LMICs—evidenced by 50,000 Kenyans opting for surgical tourism to India, for instance [27].

Estimates of \$72–\$134 per DALY compare favorably with other interventions in LMICs [8, 11]. Even for the poorest countries, \$100 represents only a fraction of the gross domestic product (GDP) per capita when, according to the World Health Organization, “interventions costing less than 3.9 the GDP per capita for each DALY averted represent good value for money” [28]. Our data thus support the cost-effectiveness of specialized surgery in children and greater resource allocation toward pediatric surgical care in LMICs [4, 6].

Limitations

There are several limitations to the current study. Smile-Train’s involvement in any given country is nonrandom, being governed by a variety of logistical ad hoc reasons. Our sample, although large, is not fully representative of LMICs (only 79 of 171 LMICs are represented). Also, within each country additional cleft work is often performed by other providers.

Second, DWs for procedures not included in the GBD project [8] and values for risk of permanent damage and probability of successful surgery are, in the absence of available literature data, entirely the author’s educated estimates. The current paucity of DW data in the surgical GBD literature reflects the challenge of empirically deriving valid weights [29] and the limited interest in covering the spectrum of individual condition weights. As such, it behooves our surgical community to contribute to the literature and validate DW values for the conditions of interest to us.

Another limitation in our backlog calculations is the ideal age of surgery for each condition. Standards for timing of surgery exist in developed countries but continuously change and evolve: cleft lip surgery is usually performed at 3 months of age. In recent years, it is often accompanied by soft palate closure, with delayed hard palate closure by 12–15 months. These standards may need to be modified in LMICs. We used more traditional repair guidelines, setting the ideal repair time for cleft lip at 6 months and for cleft palate at 1 year.

Finally, in the CEA we opted for the reimbursement value for each surgery as an appropriate surrogate of cost. Although the actual costs may be (in the author’s experience) anywhere between 50 and 100 % of the reimbursement rate, their value is less important because the ultimate cost of the surgery among all providers (hospital, surgeon, anesthetist, cleft charity) is

the reimbursement value. It must be noted, however, that this value primarily covers variable (operating), rather than fixed (capital depreciation), costs [13].

Conclusions

Despite a remarkable number of surgeries performed and the yearly increase by one cleft charity, the unmet and delayed averted global burden of cleft disease remains significant in all regions. Large geographic disparities reflect varied challenges regarding access to surgery. Cleft surgeries are cost-effective interventions to reduce GBD. Collaboration between cleft surgery providers is necessary to advocate for resources and allocate them most appropriately. Scaling up care for cleft patients must focus on more difficult-to-reach areas globally by building infrastructure and training local expertise.

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References

1. Vanderas AP (1987) Incidence of cleft lip, cleft palate, and cleft lip and palate among races: a review. *Cleft Palate J* 24:216–225
2. Adeyemo WL, Ogunlewe MO, Desalu I et al (2009) Cleft deformities in adults and children aged over six years in Nigeria: reasons for late presentation and management challenges. *Clin Cosmetic Investig Dentistry* 1:63–69
3. Anonymous (2012) SmileTrain Mission. <http://www.smiletrain.org/about/>. Accessed 4 Jan 2012

4. Bickler S, Ozgediz D, Gosselin R et al (2010) Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg* 34:374–380. doi:10.1007/s00268-009-0261-6
5. Murray CJ, Lopez D (1997) Global mortality, disability, and the contribution of risk factors: global burden of disease study. *Lancet* 349:1436–1442
6. Vos T (2009) Improving the quantitative basis of the surgical burden in low-income countries. *PLoS Med* 6:e1000149
7. Anonymous (2012) Country profiles of environmental burden of disease by WHO regions. http://www.who.int/quantifying_ehimpacts/national/countryprofile/regions/en/index.html. Accessed 4 Jan 2012
8. Lopez AD, Mathers CD, Ezzati M et al (eds) (2006) *Global Burden of Disease and Risk Factors*. Oxford University Press, New York
9. World Health Organization (2012) Life tables for WHO Member States. http://www.who.int/healthinfo/statistics/mortality_life_tables/en/. Accessed 4 Jan 2012
10. Murray CJL (1994) Quantifying the burden of disease: the technical basis for disability-adjusted life years. *Bull World Health Organ* 72:429–445
11. McCord C, Chowdhury Q (2003) A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet* 81:83–92
12. Gosselin R, Thind A, Bellardinelli A (2006) Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World J Surg* 30:505–511. doi:10.1007/s00268-005-0609-5
13. Gosselin R, Maldonado A, Elder G (2010) Comparative costeffectiveness analysis of two MSF surgical trauma centers. *World J Surg* 34:415–419. doi:10.1007/s00268-009-0230-0
14. Keren R, Pati S, Feudtner C (2004) The generation gap: differences between children and adults pertinent to economic evaluations of health interventions. *Pharmacoeconomics* 22:71–81

15. Anand S, Hanson K (1997) Disability-adjusted life years: a critical review. *J Health Econ* 16:685–702
16. Corlew DS (2010) Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. *World J Surg* 34:391–396. doi:10.1007/s00268-009-0198-9
17. Magee WP, Vander Burg R, Hatcher KW (2010) Cleft lip and palate as a cost-effective health care treatment in the developing world. *World J Surg* 34:420–427. doi:10.1007/s00268-009-0333-7
18. Coleman JR Jr, Sykes JM (2001) The embryology, classification, epidemiology, and genetics of facial clefting. *Facial Plast Surg Clin North Am* 9:1–13
19. Patil SB, Kale SM, Khare N et al (2011) Changing patterns in demography of cleft lip-cleft palate deformities in a developing country: the Smile Train effect—what lies ahead? *Plastic Reconstr Surg* 127:327–332
20. Donkor P, Plange-Rhule G, Amponsah EK (2007) A prospective survey of patients with cleft lip and palate in Kumasi. *West Afr J Med* 26:14–16
21. Onah II, Opara KO, Olaitan PB et al (2008) Cleft lip and palate repair: the experience from two West African sub-regional centres. *J Plast Reconstr Aesthet Surg* 61:879–882
22. Manyama M, Rolian C, Gilyoma J et al (2011) An assessment of orofacial clefts in Tanzania. *BMC Oral Health* 11:1–6
23. Gupta K, Bansal P, Dev N et al (2010) Smile Train project: a blessing for population of lower socio-economic status. *J Indian Med Assoc* 108:723–725 *World J Surg* (2013) 37:1562–1570 1569

24. Wilson J, Hodges A (2011) Cleft lip and palate surgery carried out by one team in Uganda: where have all the palates gone? *Cleft Palate Craniofac J* 49:299–304
25. Bellagio Working Group (2007) Increasing access to surgical services in resource-constrained settings in Sub-Saharan Africa. In: Proceedings of a conference at the Rockefeller Foundation, Bellagio Conference Center, pp 1–72
26. Singh SK (2009) Smile Train: the ascendancy of cleft care in India. *Indian J Plast Surg* 42:S192–S198
27. Anonymous (2012) Indian medical tourism destination: India emerges hot destination for medical tourism—more than 100 per cent jump likely in Kenyan patients opting for treatment in India in 2011. <http://www.businesswireindia.com/PressRelease.asp? b2mid=26376/>. Accessed 10 Jan 2012
28. Edejer TT, Evans D, Lowe J (2002) World health report, p 108. http://www.who.int/whr/2002/en/whr02_en.pdf/. Accessed 4 Jan 2012
29. Salomon J (2010) New disability weights for the global burden of disease. *Bull World Health Organ* 88:879–1570 *World J Surg* (2013) 37:1562–1570 123

Chapter 10

Economic Valuation of the Global Burden of Cleft Disease Averted by a Large Cleft Charity

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Abstract

Background

This study attempts to quantify the burden of disease averted through the global surgical work of a large cleft charity, and estimate the economic impact of this effort over a ten-year period.

Methods

Anonymized data of all primary cleft lip and cleft palate (CLP) procedures in the Smile Train database were analyzed and disability-adjusted life years (DALYs) calculated using country-specific life expectancy tables, established disability weights (DWs), and estimated success of surgery and residual disability probabilities; multiple age weighting and discounting permutations were included. Averted DALYs were calculated and gross national income (GNI) per capita was then multiplied by averted DALYs to estimate economic gains.

Results

548,147 primary cleft procedures were performed in 83 countries between 2001-2011. 547,769 records contained complete data available for the study; 58% were cleft lip and 42% cleft palate. Averted DALYs ranged between 1.46M and 4.95M. The mean economic impact ranged between USD 5510 and 50,634 per person. This corresponded to a global economic impact of between USD 3.0B and 27.7B USD, depending on the DALY and GNI values used. The estimated cost of providing these procedures based on an average reimbursement rate was USD 197M (0.7-6.6% of the estimated impact).

Conclusions

The immense economic gain realized through procedures focused on a small proportion of the surgical burden of disease highlights the importance and cost-effectiveness of surgical

treatment globally. This methodology can be applied to evaluate interventions for other conditions, and for evidence-based health care resource allocation.

Introduction

Several metrics have been used in the measurement of health interventions, including life expectancy, mortality rates, Disability-Adjusted Life Years (DALYs), Quality-Adjusted Life Years (QALYs), and others [1,2,3]. However, in order to compare the societal impact of health care to other sectors, a common measurement tool is needed. Economic assessment is such a cross-sector “universal currency”, its main drawback being the difficulty of measuring economic benefit.

This study uses a framework derived from previous work [4,5,6,7,8,9,10,11] to examine the economic value of the programs of a non-governmental organization. Smile Train (ST) supports the surgical care of patients with cleft lip and palate (CLP) in low- and middle-income countries (LMICs). ST identifies and trains surgical teams in each country and contracts to provide operative care for a pre-determined fee per case [12]. This model is akin to the specialty surgical hospital platform described by Shrime et al [13], but uses LMIC surgeons exclusively. Given that CLP impacts the social, physical, and economic lives of affected individuals, this study estimates the economic impact of the ST work, providing a framework for examining the value of health interventions for cross-sector comparison.

Methods

The approach was to estimate the economic productivity of treated individuals, then derive the counterfactual of their economic productivity had they not been beneficiaries of the programs.

The database of all procedures performed in ST programs between 2001 and 2011 was utilized. Only primary (first-time) CLP repair procedures were included. Country-specific life expectancy (LE) values were used, rather than a universal value as used by the Global Burden of Disease (GBD) study [14].

The standard DALY formula was used for burden of disease (BoD): DALYs = YLL (Years of Life Lost) + YLD (Years Lived with Disability). The YLL factor was omitted in light of the small mortality of cleft lip and palate.

The counterfactual BoD in DALYs potentially incurred by each patient without the surgical intervention (which includes the non-avertable as well as the avertable DALYs [15]), was estimated thus:

$$\text{BoD}_{\text{without intervention}} = (\text{DW}_{\text{untreated}} * \text{age}_{\text{operation}}) + (\text{DW}_{\text{untreated}} * \text{LE}_{\text{age at treatment}})$$

where $\text{DW}_{\text{untreated}}$ = disability weight for CLP untreated (both $\text{DW}_{\text{untreated}}$ and $\text{DW}_{\text{treated}}$ from the GBD study [16] and $\text{LE}_{\text{age at treatment}}$ = life expectancy at the age the operation occurred, from the Standard West Level 26 Life Table [17]).

DALYs actually incurred by each patient, or the $\text{BoD}_{\text{with intervention}}$, were then estimated in two ways. The first used the GBD study method for calculating DALYs :

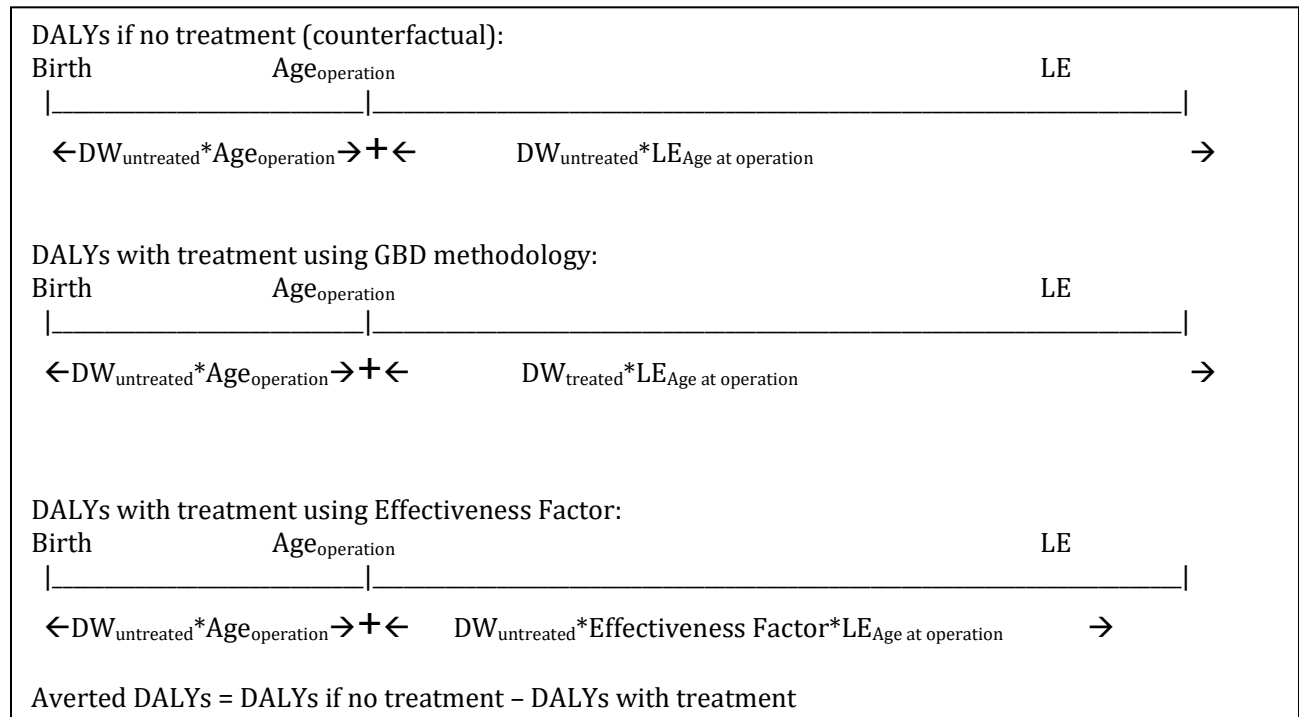
$$\text{BoD}_{\text{with intervention}} = (\text{DW}_{\text{untreated}} * \text{age}_{\text{operation}}) + (\text{DW}_{\text{treated}} * \text{LE}_{\text{age at operation}})$$

DALYs incurred with the intervention were also estimated by the method published by McCord et al [11] and widely followed in the surgical literature [18,19,20,21]. This method multiplies the ideal impact of surgical intervention by the Risk of Permanent Disability without surgery (RPD) and the Estimate of Residual Disability (ERD) after the operation. The latter factor aims to account for the residual effects of the deformity, the possibility of post-operative complications, and the possible need for further procedures. In CLP the RPD is 1.0, since disability is expected in the absence of surgery in 100% of cases, while the resolution of disability (1-ERD) was estimated to be between 75 – 94% for cleft lip and between 25-74% for cleft palate. For the purpose of this study the two factors were combined into a single “Effectiveness Factor” (EF) of 0.8 for cleft lip and 0.5 for cleft palate, similar to the previously used “Probability of Successful Treatment” (PST) for CLP [10]. This Effectiveness Factor postulates that the operation, on average, resulted in resolution of 80% of the residual disability for cleft lip and 50% for cleft palate patients. This EF was hence used in place of the $DW_{treated}$:

$$BoD_{with\ intervention} = (DW_{untreated} * Age_{operation}) + (DW_{untreated} * (1-EF) * LE_{Age\ at\ operation})$$

The difference between DALYs *without* and *with* surgical intervention represents the effect of the intervention on the health status of the patient and, globally, on alleviating BoD. **Figure 1** shows the calculation markers in a schematic fashion.

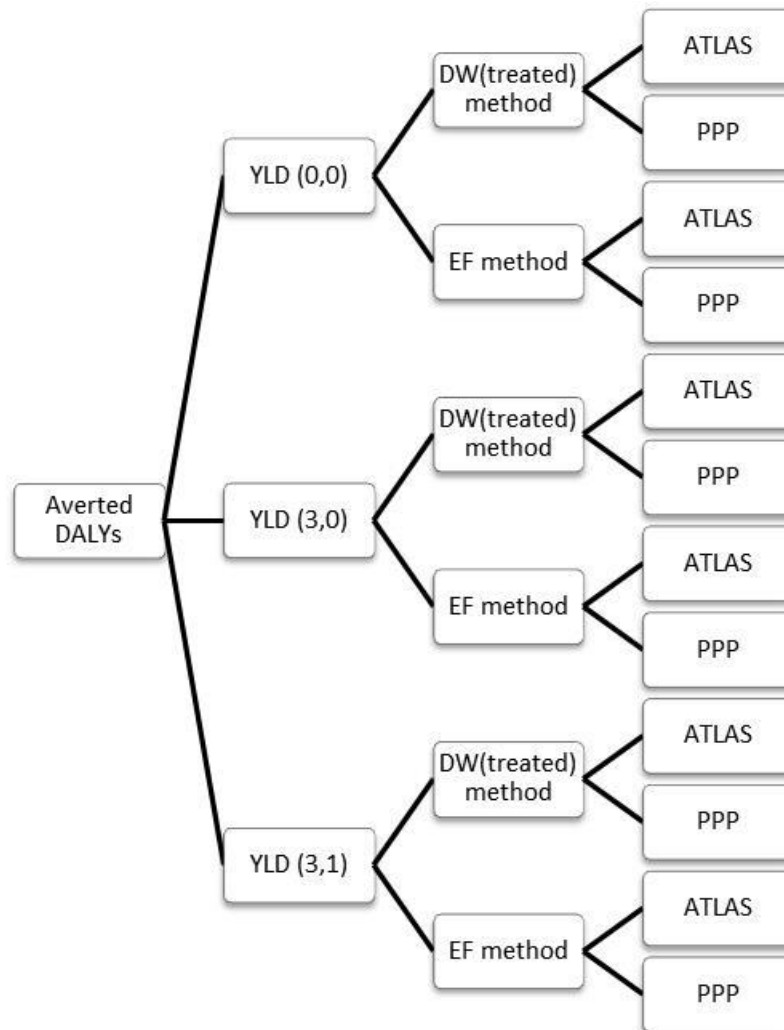
Figure 1: Diagram of age milestones and calculations used for estimating averted DALYs



DALYs were calculated with and without age weighting as was done in the original GBD study [22], and with and without 3% future discounting.

These averted DALY values were then multiplied by the GNI per capita for each country [23] to give the economic value added to the national economy over the lifetime of each patient. Using both the Atlas and Purchasing Power Parity (PPP) methods, two economic totals for each country were derived for each DALY calculation. The methodologies used in this study for calculating DALYs and the economic gain are shown diagrammatically in **Figure 2**.

Figure 2: Diagram of sensitivity analysis methodologies used in calculating the economic value of interventions



Individual totals were summed to give an estimate of economic value added for the entire program. This was reported by year and for the entire period of the study.

A cost-effectiveness analysis (CEA) was performed as previously reported for Smile Train [10].

As actual costs at the hundreds of individual sites vary widely and were not available, the set contributions per procedure paid by the organization were used as a proxy.

Data analysis and simple descriptive statistics were performed in Microsoft Excel®.

Results

Tables 1 and 2 show the results in very accessible format. Between 2001-2011, 548,147 primary operations to repair CLP were performed in 83 countries. **Figure 3** shows the cases available for study and the patient characteristics.

Figure 3: Cases available for study and Patient characteristics

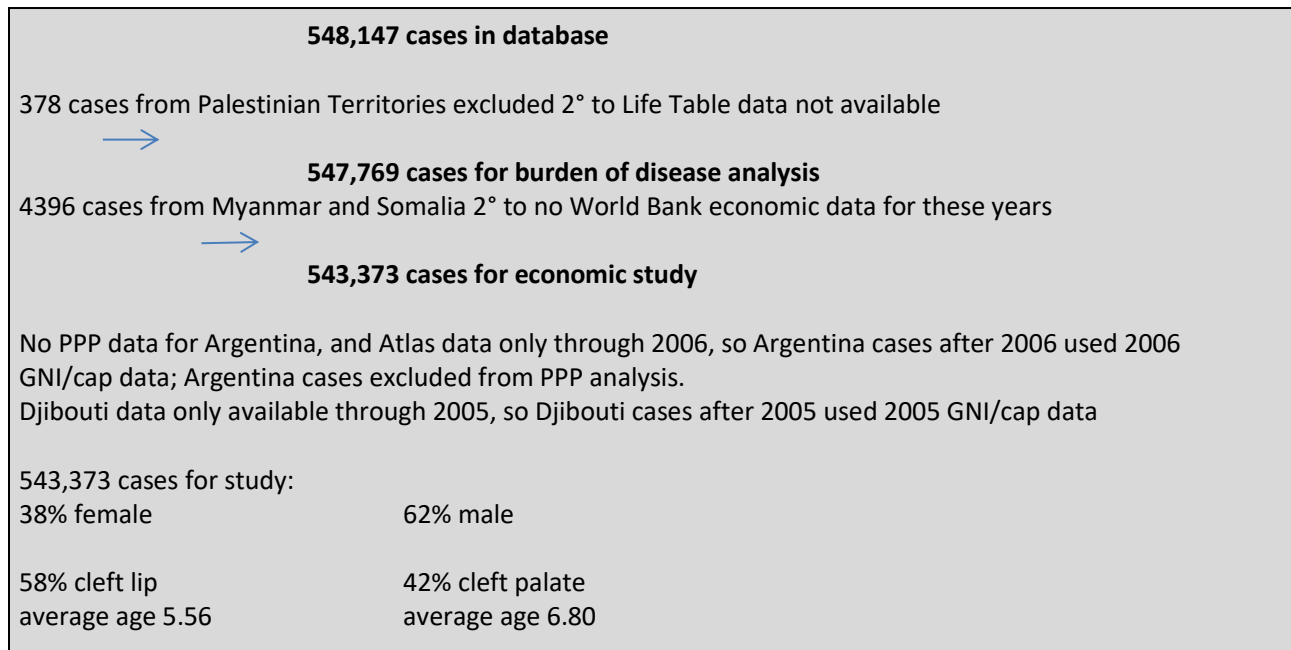


Table 1: Averted Burden of Disease in DALYs per patient and total

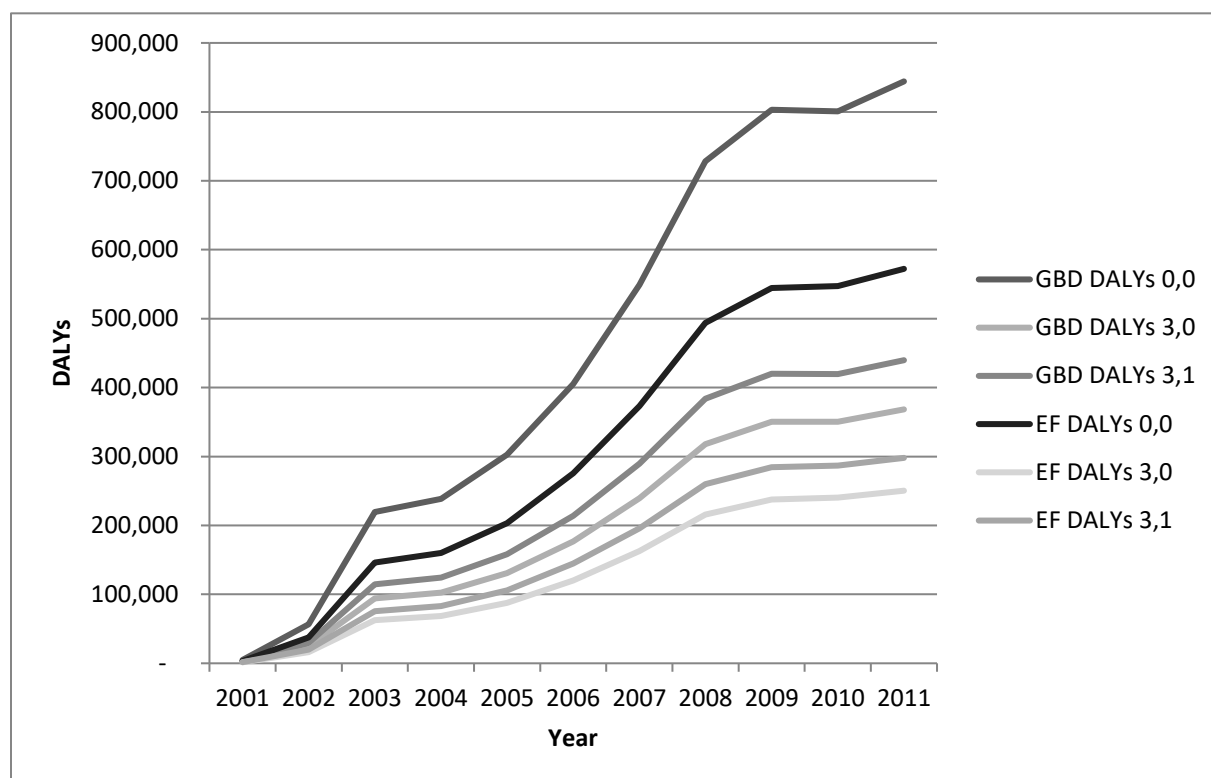
	Sum of averted DALYs using Eff factor		
	0,0	3,0	3,1
Cleft Palate	1,749,252	759,125	921,451
Cleft Lip	1,607,513	704,900	834,406
Total	3,356,765	1,464,025	1,755,857
	Sum of averted DALYs using GBD DW treated vs. untreated		
Cleft Palate	3,271,329	1,419,662	1,723,233
Cleft Lip	1,681,327	737,268	872,721
Total	4,952,656	2,156,930	2,595,954
	Average of averted DALYs per pt using Eff factor		
Cleft Palate	7.61	3.30	4.01
Cleft Lip	5.06	2.22	2.63
Total	6.13	2.67	3.21
	Average of averted DALYs per pt using GBD DW treated vs. untreated		
Cleft Palate	14.22	6.17	7.49
Cleft Lip	5.29	2.32	2.75
Total	9.04	3.94	4.74

Table 2: Economic Gain per patient and total by method of calculation

Average economic gain per pt using DW untreated vs. treated (USD)						
	Atlas 0,0	PPP 0,0	Atlas 3,0	PPP 3,0	Atlas 3,1	PPP 3,1
CP	32,216	82,405	13,693	35,303	16,478	42,629
CL	10,362	27,639	4,391	11,843	5,102	13,847
Total	19,538	50,634	8,297	21,693	9,878	25,932
Average economic gain using Effectiveness Factor (USD)						
	Atlas 0,0	PPP 0,0	Atlas 3,0	PPP 3,0	Atlas 3,1	PPP 3,1
CP	17,227	44,064	7,322	18,877	8,811	22,795
CL	9,907	26,426	4,198	11,323	4,878	13,239
Total	12,981	33,832	5,510	14,495	6,529	17,251
Sum of economic gain using DW untreated vs. treated (USD)						
	Sum of 0,0 Atlas economic gain	Sum of 0,0 PPP economic gain	Sum of 3,0 Atlas econ gain	Sum of 3,0 PPP econ gain	Sum of 3,1 Atlas econ gain	Sum of 3,1 PPP econ gain
CP	7,409,516,181	18,952,646,713	3,149,314,664	8,119,377,845	3,789,848,882	9,804,409,158
CL	3,292,831,886	8,782,991,330	1,395,432,488	3,763,409,139	1,621,160,945	4,400,167,793
Total	10,702,348,067	27,735,638,043	4,544,747,152	11,882,786,984	5,411,009,827	14,204,576,950
Sum of economic gain using Effectiveness Factor (USD)						
	Sum of 0,0 EF Atlas econ gain	Sum of 0,0 EF PPP econ gain	Sum of 3,0 EF Atlas econ gain	Sum of 3,0 EF PPP econ gain	Sum of 3,1 EF Atlas econ gain	Sum of 3,1 EF PPP econ gain
CP	3,962,032,958	10,134,401,368	1,684,008,536	4,341,611,764	2,026,516,416	5,242,635,452
CL	3,148,268,535	8,397,396,589	1,334,169,598	3,598,186,299	1,549,988,025	4,206,989,694
Total	7,110,301,493	18,531,797,956	3,018,178,134	7,939,798,063	3,576,504,442	9,449,625,147

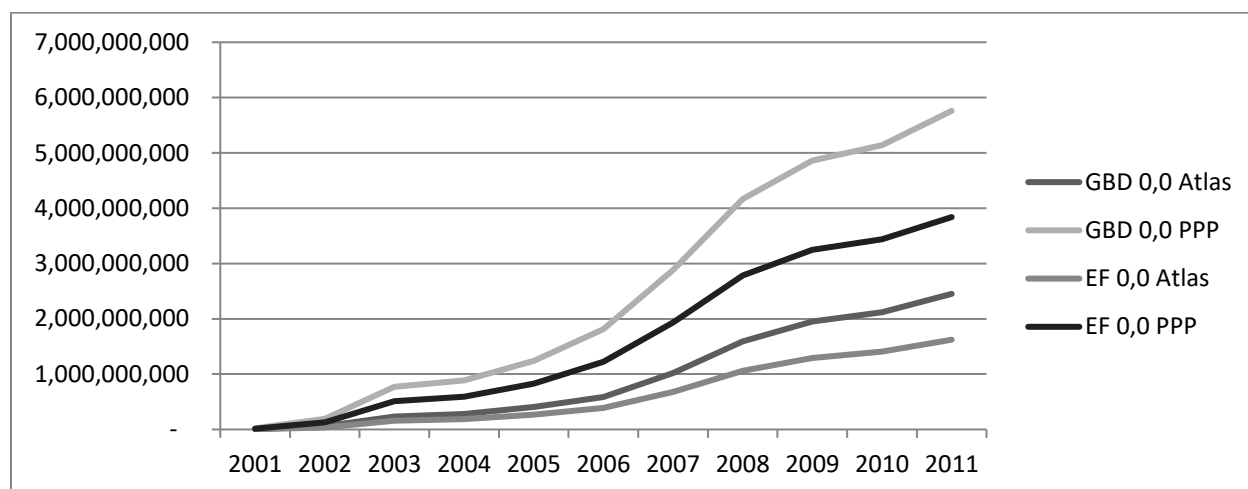
Total averted DALYs achieved by method of calculation and year are shown in **Figure 4. Table 1** details the DALYs averted by procedure and by the various calculation methods. Using the GBD DW method, about one-third of the averted BoD was due to cleft lip repairs and two-thirds to cleft palate repairs; using the effectiveness factor method this was slightly greater than half, both reflecting the greater disability associated with cleft palate.

Figure 4: Averted BoD through cleft lip and palate surgery by method and year



Values derived for the economic gain from repair of cleft lip and palate using the Atlas methodology for GNI ranged between 3.0 billion and 10.7 billion USD and between 7.9 billion and 27.7 billion USD using PPP. Using the effectiveness factor, for each person undergoing cleft lip repair, the average economic gain, calculated without age weighting or discounting, was 9,907 USD using the Atlas method and 26,426 USD using PPP. For cleft palate, these gains were 17,227 USD by Atlas and 44,064 using PPP. Using the GBD method of calculating DALYs, these figures were 10,362 USD Atlas and 27,639 USD PPP for cleft lip and 32,216 USD Atlas and 82,405 USD PPP for cleft palate. These data are detailed in **Table 2**. **Figure 5** depicts the ranges of economic impact by method per year.

Figure 5: Economic impact by year and calculation method without age weighting or discounting.



The overall cost for the interventions studied was estimated at 197 million USD, which represents between 0.7 and 6.6% of the estimated economic gain, depending on the valuation method used. In terms of cost-effectiveness, this represents between \$40 - 135 / DALY.

Discussion

This study is an effort to quantify the value of a global surgical program in economic terms. Besides providing economic data on the impact of surgical repair of CLP, the methodology lends itself to similar calculations in other specialties and programs.

The current study builds on several preceding reports of economic modeling of interventions [4,6,8,9,24]. Methodologically, measuring the economic benefit of a health intervention requires four data points: definition of the health problem, definition of the intervention and its probability of success, quantitative estimates of the change due to the intervention, and a method of converting the health effect into economic terms. [5] All of these require significant assumptions and are subject to multiple methodological approaches to address the

assumptions. These lead to wide confidence intervals, but barring extensive, expensive, and lengthy direct studies of individual economic productivity, such assumptions are necessary.

While the anatomic clefts are the core problems comprising the defects studied, there are secondary issues: hearing problems, increased rates of infection, difficulties eating and corresponding malnutrition, and orthodontic problems. These are addressed through the DWs from the GBD study and the “effectiveness factor” (EF) method.

In regard to the interventions, the mean age of the patients was quite advanced, reflecting a significant backlog [10] and delayed access to surgical care in LMICs. The preponderance of cleft lip repairs does not reflect the expected relative incidence of CLP, thus pointing towards a possible tendency to repair cleft lips but not palates. This has been observed in low-resource settings [10] and may even reflect a hidden mortality of cleft palate infants [25]. Finally, the gender distribution is skewed with a preponderance of males, a potential reflection on gender inequity issues in many low-resource settings.

The DALY data reflect the large BoD which is avertable through surgical intervention even in a narrow specialty. The 1-5 million DALYs averted over 10 years is viewed in the context of an estimated 25 million DALYs for overall surgical BoD in Africa [26], and the mean averted DALYs per patient is similar to those estimated for hydrocephalus and inguinal hernia [6,27].

The economic value of improved health resulting from the surgical interventions was determined using GNI per capita, based on the premise that each individual theoretically contributed an equal share. A first assumption was that CLP affects equally all sectors of society regardless of socioeconomic stratum, a reasonable assumption based on current epidemiologic

knowledge. A second assumption was that DALYs account for all the social, psychological, and secondary losses associated with CLP. Alternative econometric methodologies such as labor productivity, willingness to pay, value of a statistical life, or direct income studies may be the focus of future studies.

The GNI data indicate a very substantial economic impact, in keeping with other reports on surgery in limited resource settings. Using a similar methodology, Alkire et al estimated the economic impact of treating CLP in Sub-Saharan Africa (SSA) to USD 252 - 441 million, while Warf et al projected the economic impact of surgically treating hydrocephalus in SSA to around 1 billion USD [6].

Cost-effectiveness analysis (CEA) is significantly limited in this context by the assumptions necessary in the modeling, but is a natural extension of studies of economic impact. A frequent metric of CEA is the \$/DALY. The current estimate of \$40-135/DALY compares favorably with a previous report from Smile Train of \$70-134/DALY for CLP repairs [10] and with other CLP CEA studies ranging between USD 29 – 285/DALY [4,28,29].

Limitations

There are several limitations to the current study, some of which have been alluded to above. First, the sample is not random – the locale of the interventions, both nationally and regionally, reflects strategic and logistic choices within one large non-governmental organization. Absent data for some countries compounds this effect, though it affects a very small proportion of the entire dataset.

DALY calculations rely on DWs which are notoriously difficult to estimate accurately [30] and on subjective effectiveness factors. In the face of multiple competing strategies such as age weighting and future discounting, the authors' only recourse was to offer ranges rather than precise values. One specific additional limitation of our study was the assumption that mortality from CLP is negligible, which in some low-resource settings may not be true [25]. Higashi et al. found that in addition to a small mortality attributable to the cleft deformity, there also was a higher all-cause mortality in unrepaired than repaired clefts throughout life. [31] Any mortality associated with cleft deformities would serve to increase our estimates of economic benefit to repair. The uncertainty is also exacerbated by the econometric estimates, which include two alternative methodologies (Atlas and PPP) yielding disparate results.

This study does not purport to evaluate complications, quality of care, externalities, or any other measures of the specific procedures performed. It also does not specifically account for the costs of the procedures; our cost-effectiveness analysis relied on the average reimbursement rates paid by the organization for each intervention.

There are other reasons for believing that our economic estimates, while impressively high, are still underestimates. It is well-recognized that school attendance, cognitive development, and social development during childhood have significant effects on adult productivity [32], and this is not included in the DALY estimates. The Smile Train work also includes a number of secondary procedures which increase the economic impact of this surgical program.

Conclusions

The current study documents the tremendous economic value of a scaled surgical program. Whether the actual contribution to the global economy is closer to 3 billion or 30 billion USD, it

highlights the significant economic and public health value of vertical surgical interventions like CLP repair in low-resource settings. For a donor cost under 200 million USD, a 15- to 150-fold increase was added to the economies of the 83 beneficiary countries. Moreover, the clinical effect of these surgical interventions is a permanent one, not requiring ongoing therapy or expense to maintain or renew the gain. Congenital anomalies such as CLP are the ultimate “poverty trap” as described by Banerjee and Duflo [33], as without resolution of these issues there is little opportunity for the individual who otherwise has the same potential as any other person to break out of her/his economic state.

While the current study is limited to a small area of surgical care, similar studies can be undertaken in other specialties. Such studies would add to the growing body of evidence supporting the value and cost-effectiveness of surgical care as a primary health intervention globally. This should inform advocacy efforts for resource allocation in the funding of health care globally.

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References

- 1 Lopez AD, Mathers CD, Ezzati M, Jamison DT, Murray CJL, (2006) Global burden of disease and risk factors. Oxford University Press and The World Bank
- 2 Murray CJL, Salomon JA, Mathers C (2000) A critical examination of summary measures of population health. Bull WHO 78(8):981–994
- 3 Gold MR, Stevenson D, Fryback DG (2002) HALYS AND QALYS AND DALYS, OHMY: Similarities and Differences in Summary Measures of Population. HealthAnnu. Rev. Public Health 2002. 23:115–34
- 4 Corlew, D. S. (2010). Estimation of impact of surgical disease through economic modeling of cleft lip and palate care. World Journal of Surgery, 34(3), 391–6
- 5 Corlew, D. (2013). Economic Modeling of Surgical Disease: A Measure of Public Health Interventions. World Journal of Surgery, 37, 1478–1485
- 6 Warf, B. C., Alkire, B. C., Bhai, S., Hughes, C., Schiff, S. J., Vincent, J. R., & Meara, J. G. (2011). Costs and benefits of neurosurgical intervention for infant hydrocephalus in sub-Saharan Africa. Journal of Neurosurgery. Pediatrics, 8(5), 509–21
- 7 Tan-Torres Edejer T, Baltussen R, Adam T, Hutubessy R, Acharya A, Evans DB, Murray CJL, eds. (2003). WHO Guide to cost - effectiveness analysis (pp. 1–239). Geneva.
- 8 Alkire, B., Hughes, C. D., Nash, K., Vincent, J. R., & Meara, J. G. (2011). Potential economic benefit of cleft lip and palate repair in sub-Saharan Africa. World Journal of Surgery, 35(6), 1194–201
- 9 Alkire, B. C., Vincent, J. R., Burns, C. T., Metzler, I. S., Farmer, P. E., & Meara, J. G. (2012). Obstructed labor and caesarean delivery: the cost and benefit of surgical intervention. PloS One, 7(4), e34595
- 10 Poenaru, D. (2013). Getting the Job Done: Analysis of the Impact and Effectiveness of the SmileTrain Program in Alleviating the Global Burden of Cleft Disease. World Journal of Surgery, 37(7), 1562–1570

- 11 McCord, C., & Chowdhury, Q. (2003). A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *International Journal of Gynaecology and Obstetrics: The Official Organ of the International Federation of Gynaecology and Obstetrics*, 81(1), 83–92
- 12 Smile Train: leading children’s charity. www.smiletrain.org. Accessed March 31, 2014.
- 13 Shrima MG, Sleemi A, Ravilla TD, Charitable platforms in global surgery: a systematic review of their effectiveness, cost-effectiveness, sustainability, and role training (2015) *World J Surg* 39:10-20
- 14 Murray, C. J. L., & Lopez, A. (1996). A comprehensive assessment of mortality and disability from disease, injuries and risk factors in 1990 and projected to 2020. In *The Global Burden of Disease* (pp. 1–51). Cambridge, MA: Harvard University Press.
- 15 Bickler, S., Ozgediz, D., Gosselin, R., Weiser, T., Spiegel, D., Hsia, R., Dunbar P, McQueen K, Jamison, D. (2010). Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World Journal of Surgery*, 34(3), 374–80
- 16 Lopez, A. (2006). *Global Burden of Disease and Risk Factors*. (A. D. Lopez, C. D. Mathers, M. Ezzati, D. T. Jamison, & C. J. L. Murray, Eds.) Library. New York, NY: Oxford University Press & The World Bank.
- 17 World Health Organization. Standard Life Tables. Who.int/healthinfo/bodreferencesstandardlifetable.xls, accessed January 21, 2015
- 18 Shillcutt, S. D., Clarke, M. G., & Kingsnorth, A. N. (2010). Cost-effectiveness of groin hernia surgery in the Western Region of Ghana. *Archives of Surgery (Chicago, Ill. : 1960)*, 145(10), 954–61
- 19 Gosselin, R.A., Maldonado, A., & Elder, G. (2010). Comparative cost-effectiveness analysis of two MSF surgical trauma centers. *World Journal of Surgery*, 34(3), 415–9
- 20 Gosselin, R.A., Thind, A., & Bellardinelli, A. (2006). Cost/DALY averted in a small hospital in Sierra Leone: what is the relative contribution of different services? *World Journal of Surgery*, 30(4), 505–11

- 21 Wu, V. K., & Poenaru, D. (2013). Burden of surgically correctable disabilities among children in the Dadaab Refugee Camp. *World Journal of Surgery*, 37(7), 1536–1543
- 22 Murray CJL, Lopez AD, Jamison DT, The global burden of disease in 1990: summary results, sensitivity analysis and future directions *Bull WHO* (1994) 72(3) 495-509
- 23 World Bank website: <http://databank.worldbank.org/data/views/reports/tableview.aspx>; accessed January 21, 2015
- 24 Hughes et al., The Clinical and economic impact of a sustained program in global plastic surgery: valuing cleft care in resource-poor settings, *PRS* (2012) DOI: 10.1097/PRS.0b013e318254b2a2
- 25 Wilson, J., & Hodges, A. (2011). Cleft Lip And Palate Surgery Carried Out By One Team In Uganda: Where have all the palates gone? *The Cleft Palate-Craniofacial Journal : Official Publication of the American Cleft Palate-Craniofacial Association*, 49(3), 1–7
- 26 Ozgediz, D., & Riviello, R. (2008). The “other” neglected diseases in global public health: surgical conditions in sub-Saharan Africa. *PLoS Medicine*, 5(6), e121
- 27 Shillcutt, S. D., Sanders, D. L., Teresa Butrón-Vila, M., & Kingsnorth, A. N. (2013). Cost-effectiveness of inguinal hernia surgery in northwestern Ecuador. *World Journal of Surgery*, 37(1), 32–41
- 28 Alkire, B., Vincent, J., & Meara, J. (2014). Benefit-Cost Analysis of a Cleft Lip and Palate Surgical Subspecialty Hospital in India. In DCP3 draft (pp. 1–17)
- 29 Moon W, Perry H, Baek R-M, (2012). Is international volunteer surgery for cleft lip and cleft palate a cost-effective and justifiable intervention? A case study from east Asia. *World Journal of Surgery* 36:2819-2830
- 30 Gosselin, R., Ozgediz, D., & Poenaru, D. (2013). A Square Peg in a Round Hole? Challenges with DALY-based “Burden of Disease” Calculations in Surgery and a Call for Alternative Metrics. *World Journal of Surgery*, 37(11), 2507–2511

31 Higashi H, Barendregt JJ, Kassebaum NJ, Weiser TG, Bickler SW, Vos T (2015), The burden of selected congenital anomalies amenable to surgery in low and middle-income regions: cleft lip and palate, congenital heart anomalies and neural tube defects, *Arch Dis Child* 100:233-238

32 Bloom DE and Canning D (2008), Population health and economic growth, Working paper 24, Commission on Growth and Development, The World Bank

33 Banerjee AV and Duflo E, *Poor Economics: A radical rethinking of the way to fight global poverty*, Public Affairs, 2012

Chapter 11

General Discussion

This section will overview the entire work presented in chapters 1 through 9 and summarize it, highlighting the key findings and identifying the key limitations and gaps left. Finally, it will point to the areas of further research generated by the current work, and the most potentially rewarding directions for expanding this work.

Major findings

Part I

In a global health measurement context dominated by a wealth of DALY-based data painstakingly generated each year by the Institute for Health Measurement Evaluation (IHME)(6), the current work started, in chapters 1 and 2, by asking some uncomfortable questions. Yes, the DALY has proven itself a useful metric in such global-sized estimates of BoD, enabling country comparisons and, in particular, identification of detailed trends over time for the health ailments of our society. But when it comes to the surgeon who joins a surgical mission and wants, at the end of her stay, to know the real impact she has made, does this measurement system work?

The answer is generally guarded. The same DALYs which seemed to perform well in expressing the global BoD from malaria or hypertension seem much more difficult to apply to pediatric surgery. There are several obvious reasons for this: first, in an attempt to cover the entire spectrum of human disease, we are lacking disability weights (DWs) for many important surgical conditions. Secondly, even the published DW values available often appear to lack face and construct validity, and rarely include both pre-operative and post-operative (residual) values (31). Moreover, a multitude of uncertainties limit the value and credibility of the actual DALY values, with debates still raging around the validity of age weighing and future discounting, use of universal or local life expectancies, and the best way to estimate DW values cross-culturally (32). And ultimately, DALY values were designed to measure global burden of disease rather than impact of treatment, thus averted DALY values in isolation have little meaning.

If the current DALY framework does not serve the surgical community well, what alternatives do we have? While chapter 1 focused on the weaknesses and limitations in the traditional use of DALYs, chapter 2 offered several practice-based alternatives. These could be divided into

evolutionary (step-wise) and *revolutionary* (disruptive) approaches. The evolutionary strategies retain the DALY as choice metric for BoD, but tweak it for improved use in surgical work. Here the traditional division of BoD into avertable, averted, and non-avertable (11) stands out, together with the novel addition by this author of the fourth category: the *delayed (or prevalent) averted burden* suffered through delayed access to care. Alternative methods of DW estimation before and after surgical intervention are also included here.

More disruptive alternatives offered include the concepts of “need” rather than burden, again divided into unmet, met, unmeetable, and delayed (or prevalent) need. Other ways to quantify BoD include surgical backlog (for non-fatal disease) and effective coverage, as well as economic approaches to evaluating surgical care provision, such as cost-effectiveness analysis and long-term economic impact.

The conclusion of part I is that the framework for surgical BoD measurement is not currently adequate in providing reliable, defensible and convincing data needed in surgical subspecialties. The field is however young and opportunities for different or revised metrics exist.

Part II

The second part of this thesis explored some venues for addressing the concerns expressed in part I, and identifying better ways to quantify surgical need and surgical impact.

As this entire work is within the subspecialty of pediatric surgery, the first task was to generate a solid set of DW values within that specialty. This presented the challenge of selecting in a small-scale DW derivation experiment a solid methodology, comparable to the large-scale GBD studies produced by the IHME. The challenge however became a unique opportunity: to compare and select the most appropriate valuation method(s) for DW estimation, and to compare the resulting DWs across cultures.

These objectives were accomplished in chapter 3 by selecting, based on existing literature, two psychometric and two econometric valuation methods used in other similar DW estimation projects, and comparing the DW values thus obtained for a set of 15 congenital surgical health states. Reassuringly, the four methods yielded strikingly similar results for each health state, thus supporting the validity of using any one of these methods in future DW estimates. Moreover, the

values obtained in an LMIC setting (Kenya) were quite similar to those obtained in an HIC setting (Canada), supporting the cross-cultural usability of the results. In fact the only geographic discrepancies noted were in the health states related to fertility (ranked more important in low-resource settings) and cosmetic appearance (ranked less important). For the few health states for which previous DW values had been published by the GBD group, the newly derived values showed high levels of agreement with existing values. This initial study thus demonstrated that valid DW values can be readily generated in surgical specialties.

An alternative method of estimating DW values had been introduced two decades ago by Murray (4) and subsequently widely used in surgical BoD studies (16–18). It grades each surgical condition on a functional scale in terms of specific limitations in ability (of recreation, education, procreation, and occupation). While less precise than direct estimation, this method expands the DALY metric calculations to any disease condition or health state, and thus formed the basis for the studies presented in chapters 4 and 5. Moreover, the method lends itself to quantifying overall BoD, as would be encountered in a population. Thus in a field dominated by institutional studies, chapter 4 explored a population-based surgical BoD due to congenital conditions. This novel experiment combined a random household survey design with a photographic portfolio of common visible congenital conditions, contrasting results presented both as prevalence values and as DALYs, thus highlighting the qualitatively different information generated through the DALY metric.

Expanding the basic GBD concepts to surgical interventions introduced the concept of “met need” measured in *averted* DALYs, which requires reliable residual disability values following surgical intervention. This task was tackled in chapter 5, which sought to measure the impact of pediatric surgical work in a defined population: a large self-enclosed refugee camp in northern Kenya. Here as in subsequent studies, the residual post-operative surgical disability was estimated using McCord and Chowdry’s method of modulating post-intervention DALYs by the risk of permanent disability and the probability of successful treatment (16). This generated a first-time quantification of the actual impact of targeted surgical interventions in children, both overall and per patient. The study went however beyond standard DALYs. In this classic setting of delayed access to care (most children had virtually no access to surgical care before entering

the camp) the study quantified for the first time the “burden of waiting” for surgical care, in *delayed averted* DALYs. It also expressed the surgical burden and activity within the population in non-DALY terms, revealing a very poor effective coverage (under 15%) in the camps. Finally, the study used basic cost-effectiveness analysis (CEA) to document the high cost-effectiveness of the range of pediatric surgical procedures performed in the camp: between \$40 - \$90/DALY, which is vastly superior to the WHO standard typically applied (33).

Once the new methodology had been successfully applied to individual populations, the stage was set for comparative institutional studies, aimed at revealing the differences in surgical BoD alleviation between high- and low-and-middle income countries. Chapter 6 was specifically designed to use the newly derived DW values from chapter 3 in the native settings where these values had been generated. This permitted a unique comparison of impact in averting surgical BoD between two pediatric surgical units in countries literally at the opposite ends of the Human Development Index (34). While the individual types of surgical procedures predominant in each institution reflected various socio-economic factors, the study highlighted the higher averted DALYs (*met need*) both overall and per procedure in the low-resource setting.

While the comparison of met DALYs between the two economically disparate sites highlighted broad variations in practice and surgical need, contrasting delayed (*prevalent*) averted DALYs can provide a unique window into access to care at each site. This was the aim of chapter 7, which measured for the first time in the literature the “burden of waiting” in DALYs. The study identified precisely the types of procedures or specialties for which this burden of waiting is greatest, at both sites. And while the delayed averted burden was predictably much higher in the low-resource setting, the estimation of a BoD secondary to delayed access to care in the high-resource setting offers a novel way to empirically support patient waitlist discussions in the surgical specialties.

The next step (proceeding in fact in parallel with the previous ones) was to use the entire spectrum of BoD measurement and impact valuation methods developed in order to describe and audit the large-scale ongoing work of global surgical providers. SmileTrain, world’s largest cleft charity, provided a fitting dataset for such an exploration. Do the millions of dollars donated yearly “to ensure that every child born with a cleft—anywhere in the world—has the opportunity

to live a full and productive life” (35) translate into a commensurate impact on burden of cleft disease, is cost-effective, and truly benefits economically the communities in which it is implemented? This was the subject of chapters 8 and 9, two studies based on a 600,000 patient global database provided by the SmileTrain organization. In chapter 8 the surgical need met through the charity was first calculated for each global region, highlighting the major global impact that a surgical charity can have. This “happy story” message however was dampened by the estimates of delayed averted burden, effective coverage, and backlog in cleft care. Delayed access to cleft surgery resulted in up to half a million delayed DALYs (prevalent or “lost” DALYs of children having suffered from the disease before their condition was treated). As for the numbers of children operated yearly in each region, though it was impressive (SmileTrain has now provided over one million cleft surgeries globally (35)), it is still smaller than the number of new cases of cleft lip and palate (the “incident need”) born yearly within some regions. Thus as the surgical work impacts this incident need, the prevalent need (or backlog) continues to grow. One of the most sobering findings of the study was, in fact, its first-time estimation of backlog in cleft surgeries – a staggering million cases globally, with over 300,000 children waiting for surgery in both Africa and South-East Asia alone. On the economic side, however, the news was better: repairing cleft lips and palates could be done at the very affordable price of USD 70-130 / DALY, well in keeping with other surgical procedures in LMICs, and less expensive, for instance, than treating a person with HIV/AIDS (23).

Finally, chapter 9 explored further the economic benefits of the same surgical charity. It estimated an economic impact between USD 5,500 – 50,000 per person, translating into an immense USD 3 to 28 billion globally over a 10-year period of time. Most notably, these gains were realized very efficiently against costs averaging between 1 – 7% of the gains. The Lancet Commission on Global Surgery stated that addressing surgical needs can increase the GNP of individual nations by up to 2-3% (25), and this last chapter points in the same direction.

Overall, from Part II it can be concluded that the theoretical framework as introduced in Part I could be applied to providing a variety of global health care indicators applicable and meaningful to surgical specialties.

Limitations

As any work in a new field, the current project suffers from two main types of limitations: limitations in scope (“acts of omission”), and threats to validity and reliability (“acts of commission”). We will start with the latter, which must be duly recognized before any significant conclusions are drawn.

Regarding threats to validity and reliability and sources of uncertainty, all BoD work is fraught with significant limitations, introduced at each step in the calculations. Many of these limitations are in fact discussed in detail in chapter 1, which offers a critique of the DALY methodology. As described there, all the DALY studies in this thesis suffer from assumptions and value choices centering on three issues: universal vs. regional life expectancy, use of age weighing and future discounting, and the very concept of disability weights. This set of limitations was addressed in all relevant chapters by providing sensitivity analyses and presented ranges rather than distinct values, highlighting the degree of uncertainty propagated across the project. The concept of disability weight itself, central to the current project, has been met with multiple critiques and rebuttals for decades (36,37). Fundamentally, it undertakes the philosophically challenging and questionable task of assigning value to suffering, and opens itself to questions of context, culture, and myriads of socio-economic factors. Moreover, DWs are not necessarily stable over time, and may vary significantly even within one health state with its infinite potential levels of severity. Like the DALYs themselves, arguably the greatest weakness of DWs is at the more fundamental level of semantics: are respondents grading the clinical handicap associated with a given health state (the “disabling impairment”), or the pain and social stigma of the condition (the actual “disability” (38)), or both?

Having acknowledged from the onset these methodological limitations, the empirical studies of part II (chapters 3-9) still built on these assumptions, and in fact added some new ones. The central methodological study deriving new DWs (chapter 3) noted that each valuation method used for DW derivation comes with its own value choices, and the decision as to how to combine the DWs derived by each method into one unitary value introduced further uncertainty.

Applying the DALY framework to populations (as in chapters 4 and 5) added significant limitations in sampling methods, survey validity, and non-random choice of procedures undertaken based on availability of skills. Estimating met need, and thus residual disability, is by itself a very approximate process based on expert opinion, rather than actual observation, of complication rates. Moreover, such complication rates with their ensuing residual impairments vary widely between high- and low-resource settings, and even between providers and sites in similar settings. Also, calculating exact averted DALYs for health states requiring more than one surgical intervention (as in the case of staged procedures) is both difficult and inexact. Finally however, it must be borne in mind that the DALY framework was in the first place designed for estimating overall burden of disease, *not* the impact of intervention.

The novel concept of delayed averted BoD relies on measurement of the delay itself, which is imprecise as the ideal timing for surgical intervention for any congenital condition often varies with expert opinion and with local facilities such as anesthetic support. Furthermore, it is difficult to assess delays in care predicated by associated anomalies, predilection for combined multi-system interventions to minimize anesthetic use, and patient / caregiver preference.

In this work surgical backlog has been estimated in a novel way using the mean age of the patients operated, with the implication that choice of intervention among patients with the same condition is independent of age. This assumption is generally, but not universally, correct. One example of such an exception are surgical interventions in children with cleft palate, where many surgeons do not offer palate closure above a set age (typically 10 years) in light of limited chance of speech recovery in older children.

Effective coverage is generally a simple and “fool-proof” concept, but it clearly varies widely geographically - and thus any attempts to generalize the data obtained must be viewed with caution. Moreover, in the case of the SmileTrain work, the prevailing assumption was that *all* cleft interventions in the countries where SmileTrain worked were funded and provided by that organization. This assumption is inaccurate, as other cleft providers contribute a small but variable minority of the surgical procedures.

Finally, cost-effectiveness analyses and economic valuations arguably introduce some of the greatest levels of uncertainty. Here uncertainties in DALYs are compounded by the challenges of estimating direct and indirect costs of care, and in the studies of the cleft surgeries these costs were, of necessity, replaced by artificial reimbursement rates. As for the economic valuations, there are multiple options available for estimating such data across countries, and the chosen option of simple multiplication of averted DALYs by the GNI is naturally crude.

The other major set of limitations in the current work pertains to “errors of commission” – gaps that have not been covered or were incompletely addressed. There are several examples of such gaps in the project. For instance, the disability weights were estimated using four deliberately chosen valuation methods, with many other, potentially equally appropriate, methods being omitted. Also, cleft BoD estimates omit deaths from the disease and focus only on disability. While deaths from cleft disease are indeed rare, they are definitely not nil (39).

Cost-effectiveness analyses in our studies were very simplistically undertaken, using SmileTrain per-case reimbursements as a surrogate of actual direct and indirect costs. This is clearly inaccurate, as costs are often lower (though occasionally higher) than the agency’s reimbursements, depending on hospital type and national costs of care. The omitted presence of other cleft surgery providers in each country of SmileTrain activity did not affect the averted DALYs, CEA, or economic impact, but accurate backlog and effective coverage data would require including the elusive total number of cleft surgeries performed in each region.

Directions for future research

A good research study should raise more questions than it has answered – and our work is certainly no exception. The process of improving and applying the BoD framework to a surgical specialty is complex, and the current project is only a beginning. The inquiry started with the critique of the existing methods and then suggested some new ones - but ultimately no good, all-round, metric for burden of surgical disease burden has been found. While backlog and effective coverage are useful adjuncts, they clearly don’t appear to be replacing the DALYs. The quest for an ideal metric continues.

The methodology used for estimating DWs in pediatric surgery has generated useful and valid results. The method would however benefit from validation in other specialties, using either the same four valuation methods or any of the single individual ones shown to yield similar results. This strategy offers a simple and efficient way of generating specialty-specific DWs for use in quantifying the burden of surgical disease in any specialty area. The cultural aspect of DWs is also not settled – and duplicating the study in other socio-economic settings would likely prove very useful.

When estimating residual disability after surgical interventions, we have used very granular extrapolated criteria for success and complications. A key study would be needed to validate this technique by comparing it to the “gold-standard” – actual long-term follow-up of patients post-operatively.

The population study included in the project was quite limited in extent and validity, and it invites further similar population studies with larger household sample sizes and wider sampling of other sets of diseases. Such an expanded study may be combined with a standardized country-wide tool, such as the SOSAS tool (40).

The comparative study of two pediatric surgical centers was significantly affected by the uniqueness of each institution chosen, significantly limiting generalizability. Replicating such studies in other centers and with other predominant pathologies is necessary.

One of the novel approaches in BoD studies introduced by this work is the hitherto underdeveloped concept of surgical backlog, which is simple and very easily applicable. The age cohort method is attractive but will require empirical validation though studies where this method can be compared to direct population-based estimates of backlog.

Within the existing DALY framework as applied to surgical care, the emerging concept of delayed or prevalent averted burden is also novel, requiring further exploration and application to other specialties. This concept could also be expanded in high-resource settings where it provides first-hand empirical evidence of morbidity (rather than mortality) in surgical wait list systems designed to prioritize surgical services.

Beyond the extended application and validation of the key findings of this work, the research agenda in global pediatric surgery remains extensive. The existing work published through the Lancet Commission on Global Surgery offers a good, if challenging, point of departure. Much of the data generated to support the main statements and recommendations of the Commission will need to be contextualized and applied to children’s surgery. Following are some suggested research deliverables needed to achieve this goal, both from the LCoGS and outside it:

- Identify number of children globally who don’t have prompt access to surgical care;
- Estimate number of additional surgical interventions globally needed to provide adequate surgical care to children;
- Estimate numbers of surgical and anesthesia providers required to adequately staff the needed surgical interventions;
- Develop reliable markers of quality of pediatric surgical care and easy-to-use indices of care;
- Identify “bellwether procedures” for children’s surgery, as markers of hospitals able to provide essential pediatric surgical services;
- Estimate percentages of pediatric population, regionally and globally, at risk of catastrophic and impoverishing out-of-pocket expenditures for surgical care;
- Identify major barriers to surgical care of children and reasons for delayed access.

“The fields are ripe with the harvest, but the workers are few” (Luke 10:2). As the global academic community affirms its commitment to equitable health care for the world’s children, the research agenda ahead is growing, presenting global health researchers with ample opportunities for significant and meaningful projects.

Conclusions

The current thesis focused on the burden of disease encountered and addressed in pediatric surgery. It started by identifying the weaknesses in the current GBD system and the gaps that needed to be filled. It then proceeded in identifying a variety of existing and newly developed measures, both incremental and disruptive, which would render the GBD framework better

suited for measuring the impact of surgical endeavors. These measures were then uniquely applied and tested in a variety of clinical settings in low- and high-income countries, and their specific benefits identified. Many other areas of investigation however remained unanswered and will require further studies.

Despite some significant limitations, the work has demonstrated the process of applying the GBD framework to pediatric surgery, and constitutes therefore a model for similar BoD projects in other surgical specialties. Through studies such as the current one and those which will follow, global health care workers will be increasingly empowered with scientific data for concerted advocacy and fund allocation decisions in limited-resource areas.

And then hopefully, the surgeon returning from the medical mission will have a clear idea of the impact that she has had.

References

1. Murray CJL, Vos T, Lozano R, Naghavi M, Flaxman AD, Michaud C, et al. Disability adjusted life years (DALYs) for 291 diseases and injuries in 21 regions, 1990-2010: A systematic analysis for the Global Burden of Disease Study 2010. *Lancet*. 2012;380(9859):2197–223.
2. Salomon J a, Vos T, Hogan DR, Gagnon M, Naghavi M, Mokdad A, et al. Common values in assessing health outcomes from disease and injury: disability weights measurement study for the Global Burden of Disease Study 2010. *Lancet*. 2012 Dec 15;380(9859):2129–43.
3. Williams A. Calculating the global burden of disease : time for a strategic reappraisal?? *Health Econ*. 1999;8:1–8.
4. Bickler S, Ozgediz D, Gosselin R, Weiser T, Spiegel D, Hsia R, et al. Key concepts for estimating the burden of surgical conditions and the unmet need for surgical care. *World J Surg*. 2010 Mar;34(3):374–80.
5. Murray CJL. Quantifying the burden of disease : the technical basis for disability-adjusted life years. *Bull World Health Organ*. 1994;72(3):429–45.
6. Gosselin R a, Heitto M. Cost-effectiveness of a district trauma hospital in Battambang, Cambodia. *World J Surg*. 2008 Nov;32(11):2450–3.
7. Gosselin R a, Maldonado A, Elder G. Comparative cost-effectiveness analysis of two MSF surgical trauma centers. *World J Surg*. 2010 Mar;34(3):415–9.
8. McCord C, Chowdhury Q. A cost effective small hospital in Bangladesh: what it can mean for emergency obstetric care. *Int J Gynaecol Obstet*. 2003 Apr;81(1):83–92.
9. WHO. *The World Health Report 2002*. Geneva; 2002.
10. UNDP Human Development Report Office. *Human Development Report 2015 - Statistical annex*. 2015. p. 210–72.
11. SmileTrain Annual Report 2014. 2014.
12. Chao TE, Sharma K, Mandigo M, Hagander L, Resch SC, Weiser TG, et al. Cost effectiveness of surgery and its policy implications for global health: a systematic review and analysis. *Lancet Glob Heal*. 2014 Jun;2(6):334–45.
13. Meara JG, et al. Global Surgery 2030: evidence and solutions for achieving health, welfare, and economic development. *Lancet* 2015 6736(15):569-624.

14. Reidpath DD. Measuring health in a vacuum: examining the disability weight of the DALY. *Health Policy Plan.* 2003 Dec 1;18(4):351–6.
15. Anand S, Hanson K. Disability-adjusted life years : a critical review. *J Health Econ.* 1997 Dec;16(6):685–702.
16. Oliver M. Defining impairment and disability: issues at stake. In: Barnes C, Mercer G, editors. *Exploring the divide.* Leeds: The Disability Press; 1996. p. 29–54.
17. Wilson J, Hodges A. Cleft Lip And Palate Surgery Carried Out By One Team In Uganda: Where have all the palates gone? *Cleft Palate Craniofac J.* 2011 May 12;49(3):1–7.
18. Groen RS, Samai M, Petroze RT, Kamara TB, Cassidy LD, Joharifard S, et al. Household Survey in Sierra Leone Reveals High Prevalence of Surgical Conditions in Children. *World J Surg.* 2013 Mar 26;37:1220–6.

Summary

Introduction

Global surgery is a relatively new surgical discipline. Its focus is not just on international surgery, but on equity of resources and advocacy - establishing it therefore not only as a professional and academic discipline, but extending its sphere of action into social activism and health care policy. One primary focus of global surgery is the study of the global burden of surgical disease, which is measured in disability-adjusted life-years (DALYs). The DALY is a “health gap” measure, a summary measure of health incorporating both mortality and morbidity data. Its basic formula is:

$$\text{DALY} = \text{YLL} + \text{YLD} = N * \text{LE} + I * \text{LE} * \text{DW},$$

where YLL = years of life lost, YLD = years lived with disability, N= number of deaths, LE = life expectancy at onset of disease, I = number of incident cases in the population over period of time, and DW = disability weight.

Within surgery, the burden of disease has been divided into *met burden* (averted DALYs), *unmet burden* (avertable DALYs), and *unmeetable burden*. Furthermore, estimates of residual DALYs due to disability (morbidity) following surgical intervention have allowed researchers to calculate the actual impact of surgical interventions for various conditions, as well as the cost-effectiveness of surgery, expressed in \$/DALY.

There is however no data available in the literature on the met and unmet burden of *pediatric* surgical disease, or its cost-effectiveness. Part of the reason for this was the absence of disability weights for even common pediatric surgical conditions, which are needed to calculate DALYs.

The aim of this work was to first develop a burden of disease framework for pediatric surgery, then estimate the burden of pediatric surgical disease by generating disability weights for pediatric surgical conditions. These DWs were then applied in various settings both in high-income countries (HICs) and especially low-and middle-income countries (LMICs), and used to measure the impact and cost-effectiveness of pediatric surgery.

Discussion

The thesis includes 2 major parts. Part I reviews and critiques the current theoretical framework for surgical burden of disease measurement, offering several alternative metrics usable in pediatric surgery. Part 1 includes **chapters 2 and 3**, which discuss various metrics of burden of disease from their perspective of suitability to surgical disease, and propose refined or alternative metrics for use in global surgical care. Part II builds up on the aforementioned theoretical framework through several empirical studies which explore its implications and applications. This includes generating DWs for pediatric surgery, then building the evidence for the burden of surgical disease in children and the cost-effectiveness of its treatment.

Chapter 4 focuses on estimating DWs for common pediatric surgical conditions. It details the methodology used to derive these DWs and the results obtained in two widely divergent health care settings (HIC and LMIC).

Using the DWs generated in chapter 3, the magnitude of the pediatric surgical burden of disease in various African settings is surveyed and estimated in **chapter 5** (using population-based data) and **chapter 6** (using institutional data). DALY-based estimates were complemented in these two chapters by measures of effective surgical coverage and surgical backlog, and data on the cost-effectiveness of interventions.

Moving from estimating total burden of disease to estimating averted burden (or "met need") through surgical intervention, **chapter 7** expands the work started in chapter 3 by using the newly-derived DW values to measure averted burden of disease in the two (LMIC and HIC) pediatric surgical settings where the weights had been derived.

The previously undescribed concept of *delayed averted burden* "lost" through various delays in accessing surgical care is examined in **chapter 8** by comparing the results between an LMIC and HIC setting. This study shows the applicability of the delayed averted burden not only for estimating the impact of delayed access to care in LMICs, but also for surgical wait list burden in high-income countries. **Chapter 9** applies the same concept in the context of one disabling condition (cleft lip and/or palate), as chapter 5 also applied it to one isolated population world's largest refugee camp).

The empirical valuation of surgical interventions on children in LMICs was then undertaken using the \$/DALY metric in two settings: the global work of a large surgical charity (chapter 9) and the work of another, smaller, charity in a defined population (chapter 6). Finally, **chapter 10** uses the same large data set of chapter 9 to estimate the global economic impact of one large surgical charity.

Conclusion

The work described basically applies, for the first time, the burden of disease framework to the specialty of pediatric surgery. It seeks first to define and improve this framework, generates critical factors in the burden of disease calculations, then applies them in a variety of settings to produce an essential corpus of data necessary for all future endeavors in the field of global pediatric surgery.

Samenvatting

Introductie

Global surgery (letterlijk: mondiale chirurgie) is een relatief jonge chirurgische discipline. De focus ligt niet alleen op de mondiale chirurgie, maar ook op het promoten van een rechtvaardige en toegankelijke gezondheidszorg wereldwijd. Global surgery laat zich kenmerken als een professie en academische discipline, maar draagt ook bij aan het op de politieke agenda zetten van verbetering van de gezondheidszorg en ontwikkeling met betrekking tot internationale gezondheidszorg.

Het meten van ziektelast of „Burden of Disease“ is een kernactiviteit van Global Surgery. Dit wordt uitgedrukt in DALYs (disability adjusted life-years). De DALYs is een maat voor de totale hoeveelheid gezondheid die verloren gaat in een bevolking door ziekte, opgebouwd uit twee componenten: het verlies aan levensjaren en een verlies in kwaliteit van leven in jaren geleefd met ziekte. In formule:

$$DALY = YLL + YLD = N * LE + I * LE * DW$$

waarbij YLL = verloren levensjaren, YLD = jaren geleefd met ziekte, N = aantal sterfgevallen, LE = levensverwachting bij optreden van de ziekte, I = aantal incident gevallen in de bevolking, DW = wegingsfactor voor ziekte.

Binnen de chirurgie, wordt onderscheid gemaakt naar ziektelast die wordt voorkomen door de chirurgie (averted DALYs), en ziektelast die niet is voorkomen (unmet burden), en ziektelast die niet *kan* worden voorkomen (unmeetable burden). Verder worden er ramingen gemaakt van de rest-morbiditeit na een operatie om onderzoekers te helpen de werkelijke impact van chirurgische ingrepen voor verschillende condities te berekenen, evenals de kosten-effectiviteit uitgedrukt in \$ / DALY.

Er zijn echter geen gegevens beschikbaar in de literatuur over de voorkomen en onvervulde ziektelast van pediatrische chirurgie ziekte, of over de kosten-effectiviteit ervan. Een van de redenen hiervoor was de afwezigheid van wegingsfactoren voor de ernst van veel voorkomende aandoeningen in dit vakgebied die nodig zijn voor de berekening van DALYs.

Het eerste doel van dit proefschrift was een conceptueel kader te ontwikkelen voor ziektelast van aandoeningen in de pediatrische chirurgie, en vervolgens om de ziektelast meetbaar te

maken door wegingsfactoren voor de betreffende aandoeningen te ontwikkelen. Die wegingsfactoren zijn vervolgens gebruikt om de effecten en kosteneffectiviteit van pediatrische chirurgie te meten, in landen met hoge inkomens (HIC) maar vooral lage-en midden-inkomens landen (LMICs).

Discussie

Het proefschrift bestaat uit 2 delen. Deel I beschrijft en bekritiseert het huidige theoretisch kader voor het meten van ziektelast van chirurgische aandoeningen, en presenteert alternatieve maten die geschikt zijn voor gebruik in pediatrische chirurgie. Deel 1 bevat **de hoofdstukken 2 en 3**, waarin de bruikbaarheid van traditionele ziektelast maten wordt besproken voor aandoeningen in de chirurgie, en voorstellen worden gedaan voor verfijning of alternatieve maten.

Deel II bouwt voort op de eerder genoemde theoretisch kader met empirische studies die de implicaties en toepassingen verkennen. In dit deel worden de wegingsfactoren vastgesteld voor aandoeningen binnen de pediatrische chirurgie, om vervolgens gegevens te verzamelen over de ziektelast die kinderen met deze aandoening ervaren en de kosteneffectiviteit van de behandeling van deze aandoeningen.

Hoofdstuk 4 richt zich op het schatten van wegingsfactoren voor veel voorkomende pediatrische chirurgische aandoeningen. Het beschrijft hoe de wegingsfactoren zijn vastgesteld en hoe de resultaten van pediatrische chirurgie verschillen tussen landen met een hoog inkomen en landen met lage of midden inkomens.

Gebruik makend van de wegingsfactoren voor ziekten uit hoofdstuk 3, is berekend hoeveel ziektelast aandoeningen in het gebied van de pediatrische chirurgie genereren in verschillende Afrikaanse landen op basis van populatie gegevens (**hoofdstuk 5**) en institutionele data (**hoofdstuk 6**). De ziektelast metingen vanuit het DALY kader zijn in die berekeningen aangevuld met informatie omtrent chirurgische dekking en chirurgische achterstand, en gegevens over de kosten-effectiviteit van interventies.

Aanvullend hierop beschrijft **hoofdstuk 7** hoeveel ziektelast in lage- en midden inkomens landen en in hoge inkomens landen wordt voorkomen met pediatrische chirurgie.

Hoofdstuk 8 vergelijkt voor lage- en midden inkomens en voor hoge inkomens landen de consequenties van vertragingen in de toegang tot chirurgische zorg, waardoor een deel van de te voorkomen of afgewende ziektelast (*averted burden*) niet gerealiseerd wordt. Deze studie laat zien dat *delayed averted burden* een toepasbaar concept is voor het inschatten van de gevolgen van de vertraagde toegang tot zorg in lage en midden inkomens landen, maar ook voor de gevolgen van wachtlijsten voor chirurgie in landen met hoge inkomens. Hoofdstuk 9 past *delayed averted burden* toe in een specifieke setting, namelijk bij de behandeling van schisis (hazenlip en / of gespleten gehemelte) in een geïsoleerde populatie ('s werelds grootste vluchtelingenkamp). Ten slotte is er een economische evaluatie uitgevoerd van chirurgische ingrepen bij kinderen in lage en midden inkomenslanden, voor twee instellingen: een instelling die mondiaal actief is (hoofdstuk 9) en een andere, kleinere instelling die haar activiteiten tot een specifieke populatie heeft beperkt (hoofdstuk 6). **Hoofdstuk 10** berekent tot slot de wereldwijde economische impact van een grote, mondiaal actieve liefdadigheidsinstelling voor chirurgie, gebruik makend van de gegevens uit hoofdstuk 8.

Conclusie

Dit proefschrift past voor het eerst het ziektelast concept toe in de pediatrie chirurgie. Kritische factoren voor verfijning en uitbreiding van het ziektelast kader voor dit veld zijn eerst gedefinieerd, en daarna toegepast om in meerdere contexten gegevens te genereren die richting geven aan toekomstige inspanningen op het gebied van de pediatrie chirurgie.

PhD Portfolio

Name PhD candidate: Dan Poenaru
PhD period: 2012-2016
Promotors: Prof.dr. J. L. Severens
Prof.dr. D. Tibboel
Co-promotor: Dr. E. A. Stolk

PhD training

Teaching

University of British Columbia Branch for International Surgery course: "Global Disability: A Surgical Care Mandate" Sept.-Nov. 2014

McGill University Advanced Curriculum in Pediatric Global Health course: "Global burden of (pediatric) surgical disease" Oct. 2013

University of British Columbia Branch for International Surgery course: "Global Disability: A Surgical Care Mandate" Sept.-Nov. 2013

University of British Columbia Branch for International Surgery course: "Global Disability: A Surgical Care Mandate" Sept.-Nov. 2012

University of British Columbia Branch for International Surgery course: "Surgical Care in International Health" Mar.-April 2012

Presentations at conferences

Oral presentations

American Academy of Pediatrics National Conference. How Far Are We from Optimally-Resourced Care in Africa?: A Pilot Survey from the Global Pediatric Surgery Network (GPSN) Collaborative (Washington DC, Oct. 2015)

Canadian Association of Pediatric Surgeons,: Global Pediatric Surgery (panel talk) (Niagara Falls, ON Sept. 2015)

Sainte Justine Hospital Global Plastic Surgery Day: The burden of surgical disease in low-resource settings: discovering it, measuring it, and addressing it (Montreal, QC, Aug. 2015)

Global Surgery Rounds: The burden of surgical disease in low-resource settings: discovering it, measuring it, and addressing it (John Radcliffe Hospital, Oxford, UK, July 2015)

Global Surgery Conference: The burden of surgical disease in low-resource settings: discovering it, measuring it, and addressing it (Maine Medical Center, Portland, ME, July 2015)

British Association of Paediatric Surgeons Congress: The global burden of paediatric surgical disease: discovering it, measuring it, addressing it. (Cardiff, UK July 2015)

Bethune Round Table for International Surgery,: Pediatric Plastic Surgery in Global Health: A Scoping Review. (Calgary, AB, July 2015)

Pan-African Paediatric Surgical Association : Pediatric Plastic Surgery in Global Health: A Scoping Review. (Cairo, Egypt, Nov. 2014)

Pan-African Paediatric Surgical Association: Epidemiology of pediatric trauma in Ethiopia: data from an institutional trauma database. (Cairo, Egypt, Nov. 2014)

Pan-African Paediatric Surgical Association: Averted burden of pediatric surgical disease in a Ugandan referral institution. (Cairo, Egypt, Nov. 2014)

Canadian Association of Pediatric Surgeons: Mortality rates associated with pediatric surgical conditions in low and middle-income countries in Africa: a call to action. (Montreal, QC, Sept. 2014)

Canadian Association of Pediatric Surgeons: The burden of waiting: DALYs accrued from delayed access to pediatric surgery in Kenya and Canada. (Montreal, QC, Sept. 2014)

Bethune Round Table,: Pediatric surgery outcomes in low-and middle-income African countries: a scoping review of the recent literature (Hamilton, ON, June 2014)

Bethune Round Table: Quantification of Global Burden of Pediatric Surgical Disease Using Disability Weights: Implications for Priority Setting. (Hamilton, ON, June 2014)

Fifth Annual CUGH Conference: Understanding the burden of surgical congenital anomalies in Kenya: an international mixed-methods approach (Washington DC, May 2014)

American Academy of Pediatrics National Conference,: Measuring the disability averted through surgery for 14 pediatric congenital conditions in a Kenyan hospital (Orlando, FL, Oct. 2013)

American Academy of Pediatrics National Conference: Too little, too late: A pediatric surgeon's struggle with providing access to care in Africa (Orlando, FL, Oct. 2013)

McGill University Surgery Grand Rounds: Global burden of surgery: what's in it for surgeons (Montreal, QC, Oct. 2013)

McGill University Pediatrics Grand Rounds: Too little, too late: A pediatric surgeon's struggle with providing access to care in Africa (Montreal, QC, Sept. 2013)

ASAP Global Surgery Conference: Analysis of the averted global burden of cleft disease through a large cleft charity and its economic valuation (Durham, NC, Sep. 2013)

Global Health Metrics and Evaluation: Establishing Disability Weights for Congenital Pediatric Surgical Disease: A cross-sectional, multi-modal study (Seattle, CA, June 2013)

McMaster-Brock Global Health Forum: Quantification of Global Burden of Pediatric Surgical Disease Using Disability Weights through International Partnership (Hamilton, ON, Jan. 2013)

College of Surgeons of East, Central, and Southern Africa Annual Scientific Meeting: Getting the job done: analysis of the effectiveness of the SmileTrain program in alleviating the global

burden of cleft disease (Addis Ababa, Ethiopia, Dec. 2012) College of Surgeons of East, Central, and Southern Africa Annual Scientific Meeting: Burden of congenital disease in Kenya: a population-based study (Addis Ababa, Ethiopia, Dec. 2012)

College of Surgeons of East, Central, and Southern Africa Annual Scientific Meeting: Burden of surgically correctable disabilities among children in the Dadaab refugee camp (Addis Ababa, Ethiopia, Dec. 2012)

Bethune Round Table for International Surgery: Quantifying the Burden of Pediatric Surgical Disease due to Delayed Access to Care (Toronto, ON, June 2012)

McMaster University Department of Surgery Chair's Rounds: "Measuring the burden of pediatric surgical disease" (Hamilton, ON, June 2012) Pan-African Paediatric Surgical Association Congress: Burden of congenital disease in Kenya: a population-based study (Cape Town, South Africa, Mar. 2012)

Pan-African Paediatric Surgical Association Congress: Burden of surgically correctable disabilities among children in the Dadaab refugee camp (Cape Town, South Africa, Mar. 2012)

Poster presentations

Bethune Round Table for International Surgery,. Ready, Set, Go! Follow-up from the 2014

Bethune Round Table Global Surgery Research Working Group (Halifax, NS, June 2016)

Bethune Round Table for International Surgery,. Ready, Set, Go! Deferred surgical intervention among pediatric patients in Tanzania: reasons for delays in presentation and surgical care (Halifax, NS, June 2016)

Canadian Association of Pediatric Surgeons: Trauma in Ethiopian children: factors affecting severity of injury (Niagara Falls, ON, Sept. 2015)

Pan-African Paediatric Surgical Association: "Partnership through Fellowship: The Bethany Kids – McGill University Pediatric Surgery Fellowship Exchange" (Cape Town, South Africa, Mar. 2012)

Pan-African Paediatric Surgical Association: "Getting the job done: analysis of the effectiveness of the SmileTrain program in alleviating the global burden of cleft disease" (Cape Town, South Africa, Mar. 2012)

ASAP Global Surgery Conference: "Getting the job done: analysis of the effectiveness of the SmileTrain program in alleviating the global burden of cleft disease" (San Diego, CA, Nov. 2011)

ASAP Global Surgery Conference: "Burden of congenital disease in Kenya: a population-based study" (San Diego, CA, Nov. 2011)

ASAP Global Surgery Conference: "Burden of surgically correctable disabilities among children in the Dadaab refugee camp" (San Diego, CA, Nov. 2011)

Bethune Round Table for International Surgery: Prevalence of congenital defects in Kenya: a population-based study" (Montréal, PQ, May 2011)

Books

Ameh E, Bickler SW, Lakhoo K, Nwomeh BC, Poenaru D, eds . Paediatric Surgery: a comprehensive text for Africa. 2nd ed. Seattle, WA: Global HELP, 2016 (*submitted*)

Muzira A, Ameh E, Ozgediz D, Poenaru D. Essential Pediatric Surgery. In Park A, Price R., eds. Global Surgery: The Essentials. Springer (*in print*)

Bickler SW, Ozgediz D, Poenaru D. Academic Global Surgery: Current Research Needs and Priorities. In M Swaroop, S Krishnaswami, eds. Academic Global Surgery. Springer International Publishing, 2016

Ameh E, Bickler SW, Lakhoo K, Nwomeh BC, Poenaru D, eds. Paediatric Surgery: a comprehensive text for Africa. Seattle, WA: Global HELP, 2011

List of scientific publications

Included in thesis

Poenaru D, Pemberton J, Frankfurter C, Cameron BH, Stolk E. Establishing disability weights for congenital paediatric surgical conditions: a prospective, cross-sectional, multi-modal approach. Health Meas Res (*submitted*)

Poenaru D, Pemberton J, Cameron BH. The Burden of Waiting: DALYs accrued from delayed access to pediatric surgery in Kenya and Canada. J Pediatr Surg 50:765-70, 2015

Poenaru D, Pemberton J, Frankfurter C, Cameron BH. Quantifying the Disability Averted through Pediatric Surgery: a cross-sectional comparison of a pediatric surgical unit in Kenya and Canada. World J Surg 39:2198-2206, 2015

Poenaru D, Ozgediz D, Gosselin R. Burden, need, or backlog: a call for improved metrics for the global burden of surgical disease. Int J Surg 12:483-6, 2014

Gosselin R, Ozgediz D, Poenaru D. A square peg in a round hole? Challenges with DALY-based “burden of disease” calculations in surgery and a call for alternative metrics. World J Surg 37:2507-11, 2013

Wu VK, Poenaru D, Poley MJ. Burden of Surgical Congenital Anomalies in Kenya: A Population-Based Study. J Tropical Pediatrics 59:195-202, 2013

Wu V, Poenaru D. Burden of surgically correctable disabilities among children in the Dadaab refugee camp. World J Surg 37:1536-43, 2013

Poenaru D. Getting the job done: analysis of the effectiveness of the SmileTrain program in alleviating the global burden of cleft disease. World J Surg 37:1562-70, 2013

Poenaru D, Lin D, Corlew S. Economic Valuation of the Global Burden of Cleft Disease Averted by a Large Cleft Charity. World J Surg 40:2053-9, 2015

Not included in this thesis

Corlew S, Alkire B, Poenaru D, Meara J, Shrimme M. Economic valuation of the impact of a large surgical charity using the Value of Lost Welfare approach. BMJ Global Health 1:e000059, 2016

Ford K, Poenaru D, Moulot O, Tavener K, et al. Gastroschisis: bellwether for neonatal surgery capacity in low resource settings? J Pediatr Surg 51:1262-7, 2016

Saxton AT, [Poenaru D](#), Ozgediz D, Ameh Emmanuel A, Farmer D, Rice HE. Economic analysis of children's surgical care in low-and middle-income countries: A systematic review. PLOS ONE 11:e01654480, 2016

[Poenaru D](#). The global burden of paediatric surgical disease in low-resource settings: discovering it, measuring it, and addressing it. J Pediatr Surg 51:216-24, 2016

Ozgediz, Langer M, Kissa P, [Poenaru D](#). Pediatric surgery as an essential component of global child health. Sem Pediatr Surg 25:3-9, 2016

GBD 2013 Disease and Injury Incidence and Prevalence Collaborators. Global, regional, and national comparative risk assessment of 79 behavioural, environmental and occupational, and metabolic risks or clusters of risks in 188 countries, 1990–2013: a systematic analysis for the Global Burden of Disease Study 2013. The Lancet 386: 2287-2323, 2015

GBD 2013 Disease and Injury Incidence and Prevalence Collaborators. Global, regional, and national incidence, prevalence, and YLDs for 301 acute and chronic diseases and injuries for 188 countries, 1990-2013: a systematic analysis for the Global Burden of Disease Study 2013. The Lancet 386:743-800, 2015

Livingston MH, DCruz J, Pemberton J, Ozgediz D, [Poenaru D](#). Mortality of pediatric surgical conditions in low and lower middle income countries in Africa. J Pediatr Surg 50:760-764, 2015

GBD 2013 Mortality and Causes of Death Collaborators. Global, regional, and national age– sex specific all-cause and cause-specific mortality for 240 causes of death, 1990 – 2013 : a systematic analysis for the Global Burden of Disease Study 2013. Lancet 385:117-71, 2015

Ozgediz DE, [Poenaru D](#). The burden of pediatric surgical conditions in low and middle income countries: a call to action. J Pediatr Surg 47:2305-11, 2012

Other publications

LeBlanc D, Nyagetuba KM, Frehun A, Baird R, Hansen E, Andrawes S, [Poenaru D](#). Management of bladder exstrophy using the Mainz II procedure in a resource-limited setting. Eur J Pediatr Surg (submitted)

Butler MW, Ozgediz D, [Poenaru D](#), et al. Global Paediatric Surgery Network Collaborative. The Global Paediatric Surgery Network: A model of subspecialty collaboration within global surgery. W J Surg 39:335-342, 2015

Nasir AA, Ameh EA, [Poenaru D](#). Completion publication of abstracts presented at the scientific meetings of the Pan-African Paediatric Surgical Association. Afr J Paed Surg 10:246-51, 2013

Wood JH, Nthumba P, Stepita-Poenaru E, [Poenaru D](#). Pediatric Surgical Site Infection in the Developing World: A Kenyan Experience. Ped Surg Int 28:523-7, 2012

Cornegé-Blokland E, Jansen HE, de Jong-de Vos van Steenwijk CCE, [Poenaru D](#). Quality of life of children with spina bifida in Kenya is not related to the degree of the spinal defects. Tropical Med Int Health 16:30-36, 2011

MacLeod JBA, Jones T, Aphivantrakul P, Chupp M, [Poenaru D](#). Evaluation of Fundamental Critical Care Course in Kenya: Knowledge, Attitude & Practice. J Surg Res 167:223-230, 2011

Gathura E, [Poenaru D](#), Bransford R, Albright L. Outcomes of ventriculoperitoneal shunting in Sub-Saharan Africa. J Neurosurg Pediatr 6:329-335, 2010

Nthumba PM, Stepita-Poenaru E, Poenaru D, Bird P, Allegranzi B, Pittet D, Harbarth S. Surgical Site Infections: A Cluster-Randomized, Cross-over Study of the Efficacy of Plain Soap and Water Versus Alcohol-Based rub for Surgical Hand Preparation. *Br J Surg* 97:1621-1628, 2010

Margaron F, Poenaru D, Bransford R, Albright L. Timing of Ventriculoperitoneal Shunt Insertion Following Spina Bifida Closure in Kenya. *Childs Nerv Syst* 26:1523-1528, 2010

Ngock GF, Sparks SR, Poenaru D. Crocheting pin in the falciform ligament: a rare cause of recurrent right hypochondriac pain. *East Centr Afr J Surg* 15:147-149, 2010

Margaron FC, Poenaru D, Northcutt A. Pediatric cancer spectrum in Kenya: a histopathologic review. *Pediatr Surg Int* 26:789-94, 2010

Poenaru D, Borgstein E, Numanoglu A, Azzie G. Caring for children with colorectal disease in the context of limited resources. *Seminars Pediatr Surg* 19:118-127, 2010

Munyi N, Poenaru D, Bransford R, Albright L. Encephalocele – a single institution African experience. *East Afr J Med* 86:51-54, 2009

Jansen H, Blokland E, de Jong C, Greving J, Poenaru D. Quality of life of African children with spina bifida: results of a validated instrument. *Cerebrospinal Fluid Res* 6 Suppl 2:S25, 2009

van't Veer T, Meester H, Poenaru D, Kogei A, Augenstein KB. Quality of life of families of children with spina bifida in Kenya. *Tropical Doctor* 38:160-162, 2008

Bauman N, Poenaru D. Hydrocephalus in Africa: a surgical perspective. *Ann Afr Surg* 2:30-37, 2008

Nthumba PM, Carter LL, Poenaru D. Ambiguous genitalia in rural Africa and the complexities of management: Which way forward? *East Centr Afr J Surg* 13:51-59, 2008

Meester H, Van't Veer T, Poenaru D, Bransford R. Spina bifida care in Kenya through a network of mobile clinics. *African J Paed Surg* 3:9-12, 2006

Park J, Minor S, Taylor RA, Vikis E, Poenaru D. Why are women deterred from general surgery training? *Am J Surg* 190:141-6, 2005

Bourque M, Poenaru D, Bransford R. Unusual variants of spina bifida – four additional cases. *African J Paed Surg* 1:89-93, 2005

Dwight P, Poenaru D. Entero-enteric fistula following mild necrotizing enterocolitis. *Eur J Pediatr Surg* 15:137-9, 2005 Cheng G, Soboleski D, Daneman A, Poenaru D, Hurlbut D. Sonographic pitfalls in the diagnosis of enteric duplication cysts *AJR* 184:521-5, 2005

Jeruto A, Poenaru D, Bransford R. Clean intermittent catheterization: overview of results in 194 patients with spina bifida. *African J Paed Surg* 1:20-3, 2004

Dwight P, Poenaru D. Spontaneous duodenal blow-out in an infant: a case report. *J Pediatr Surg* 39:1593-4, 2004

Snelling CMH, Poenaru D, Drover JW. Minimum post-operative antibiotic duration in advanced appendicitis in children: a review. *Pediatr Surg Int* 11-12:838-845, 2004

Lingard L, Garwood S, Poenaru D. Tensions influencing operating room team function: does institutional context make a difference? *Med Educ* 38:691-9, 2004

Basford TJ, Poenaru D, Silva M. Comparison of delayed complications of central venous catheters placed surgically or radiologically in pediatric oncology patients. *J Pediatr Surg* 38:788-92, 2003

Minor S, Poenaru D., Park J. A study of career choice patterns among Canadian medical students. *Am J Surg* 186:182-8, 2003

Minor S, Poenaru D. The in-house education of clinical clerks in Surgery and the role of housestaff. *Am J Surg* 184:471-5, 2002

Gmora S, Poenaru D, Tsai E. Neostigmine for the treatment of pediatric acute colonic pseudo-obstruction. *J Pediatr Surg* 37:E28, 2002

Dueck A, Poenaru D, Jamieson MA, Kamal IK. Unilateral ovarian agenesis and Fallopian tube maldescent. *Pediatr Surg Int* 17:228-9, 2001

Poenaru D, Fitzgerald P, et al. Training general surgery residents in pediatric surgery: a Canadian survey. *J Pediatr Surg* 36:706-10, 2001

Dueck A, Poenaru D, Pichora D. Cost factors in Canadian pediatric trauma. *Can J Surg* 44:11721, 2001

Amjadi K, Poenaru D, Soboleski D, Hurlbut D, Kamal I. Anterior rectal duplication: a diagnostic challenge. *J Pediatr Surg* 2000; 35: 613-4

Valadka S, Poenaru D, Dueck A. Long-Term Disability Following Trauma in Children. *J Pediatr Surg* 35:684-7, 2000

Poenaru D. Objectives-based self-assessment of surgical residents. *Acad Med* 2000, 75:543

Chanda MN, Jamieson MA, Poenaru D. Congenital perineal lipoma presenting as "ambiguous genitalia". *J Pediatr Adolesc Gynecol* 2000, 13:71-4

O'Brien K, Poenaru D. Penetrating sledding injuries to the lower torso. *CMAJ* 1999; 160:353

Soboleski D, Mehta S, Kamal I, Dexter D, Poenaru D. Hemangioendothelioma of the adrenal gland in a 4-month old male infant. *Am J Roentgenol* 1999;172:235-7

Dueck A, Poenaru D, Kamal I. Hypertension following minor trauma: a rare presentation of pheochromocytoma. *Pediatr Surg Int* 1999; 15:508-9

Bozinovski J, Poenaru D, Paterson W, Kamal I. Esophageal aperistalsis following fundoplication in a patient with trisomy 21. *Pediatr Surg Int* 1999; 15:510-1

Poenaru D, Jacobs D, Kamal I. Unusual findings in the inguinal canal: a report of 4 cases. *Pediatr Surg Int* 1999; 15:515-6

Davison P, Poenaru D, Kamal I. Esophageal atresia: primary repair of rare long gap variant involving distal pouch mobilization. J Pediatr Surg 1999, 34:1881-3

Starreveld Y, Poenaru D. Ventriculo-peritoneal shunt knot -a rare cause of bowel obstruction and ischemia. Can J Surg 1998; 41:239-240

Fung K, Poenaru D, Soboleski DAA, Kamal IM. Impact of magnetic resonance imaging on the surgical management of cystic hygromas. J Pediatr Surg 1998; 33:839-841

Poenaru D, Woo M. Undergraduate pediatric surgery objectives: goal and reality. J Pediatr Surg 1998; 33:852-855

Poenaru D, Davidson L, Donnely M, Tekian A. Is a mandatory general surgery rotation necessary in the surgical clerkship? Am J Surg 1998; 175:515-517

Anderson MAB, Poenaru D, Kamal IM. Calcified catheter cast: a rare complication of indwelling central venous catheters in infants. Pediatr Surg Int 1998; 13:610-612

Uroz-Tristan J, Garcia-Urgelles X, Poenaru D, et al. Torsion of vermiform appendix: value of ultrasonographic findings. Eur J Pediatr Surg. 1998; 8:376-7

Seshadri PA, Poenaru D, Park A. Laparoscopic splenic cystectomy. J Pediatr Surg 1998 ; 33:1439-40

Poenaru D, Roblin N, Bird M, et al. The pediatric bowel management clinic: initial results of a multi-disciplinary approach to functional constipation in children. J Pediatr Surg 1997; 32: 843

Poenaru D, Morales D, Richards A, O'Connor HM. Running an objective structured clinical examination on a shoe-string budget. Am J Surg 1997; 173:538-541

Abou-Khalil S, Smith BM, MacLean JD, Poenaru D, et al. Acute cholecystitis and cholangitis caused by Echinococcus granulosus. Am J Gastroenterol 1996; 91:805-7

Kimmins MH, Poenaru D, Kamal I. Traumatic gastric transection: a case report. J Pediatr Surg 1996; 31:757-758

Uroz-Tristan J, Poenaru D, Urgelles XG, et al. Segmental cecal dilatation with absent appendix: a case of failure of appendiceal regression? Eur J Pediatr Surg 1996; 6: 373-374

Poenaru D, Uroz-Tristan J, Murphy S, Bensoussan A-L. Imperforate anus, malrotation and Hirschsprung's disease: a rare association. Eur J Pediatr Surg 1995; 5:187-9

Marras T, Poenaru D, Kamal I. Perinatal management of nasopharyngeal teratoma. J Otolaryngol 1995; 24:310-312

Poenaru D, Yazbeck S, Murphy S. Primary spontaneous pneumothorax in children. J Pediatr Surg 1994; 29:1183-5

Poenaru D, Uroz-Tristan J, Leclerc S, Murphy S, St-Vil D, Youssef S, Blanchard H. Minimally conjoined omphalopagi: a consistent spectrum of anomalies. J Pediatr Surg 1994; 29:1236-8

Uroz-Tristan J, Poenaru D, Martinez Lagares F, Leclerc S, Sanchis Solera L. Selective splenic artery embolization or use of polyglycolic acid mesh in children with severe splenic trauma. Eur J Pediatr Surg 1994; 4:1-3

Poenaru D, Homsy YL, Peloquin F, Andze G. The value of laparoscopy in the diagnosis and treatment of non-palpable testicular cryptorchism. Prog Urol 1994; 4:206-13

Poenaru D, Yazbeck S. Anal fistulas in infants: etiology, features, management. J Pediatr Surg 1993; 28:1194

Uroz-Tristan J, Mogueya SA, Poenaru D, Martinez Lagares F, et al. Intralobar pulmonary sequestration with multiple arterial blood supply. Cir Pediatr 1994; 7:102-4

Poenaru D, Laberge JM, Neilson IR, Guttman FM. A new prognostic classification for esophageal atresia. Surg 1993; 113:426

Poenaru D, Homsy Y, et al. The laparoscopic management of the impalpable abdominal testis. Urology 1993; 42:574-8

Poenaru D, Laberge JM, Neilson IR, Nguyen LT, Guttman FM. Over 25-year experience with end-to-end versus end-to-side repair for esophageal atresia. J Pediatr Surg 1991; 26:472

Poenaru D, Szilagyi A, Zabad F, Lamoureux E, Lough JO. Hepatocellular carcinoma: comparison of clinical features among ethnic groups in an area of low prevalence. Am J Gastroenterol 1991; 86:487

Poenaru D, De Santis M, Christou NV. Imipenem vs. tobramycin-antianaerobe antibiotic therapy in intra-abdominal infections. Can J Surg 1990; 33:415

Poenaru D, Christou NV. Clinical outcome of seriously ill surgical patients with intra-abdominal infection depends on both physiologic (APACHE II Score) and immunologic (DTH Score) alterations. Ann Surg 1990; 213:130

Poenaru D, Laberge JM, Jequier S, Blanchard PW, Doody DP. Ultrasound diagnosis of delayed-onset congenital diaphragmatic hernia associated with group B streptococcal infection. Pediatr Surg Int 1988; 3:66

Vinograd I, Filler RM, England SJ, Smith C, Poenaru D, Bahorik A, Kent G: Tracheomalacia: An experimental animal model for a new surgical approach. J Surg Res 1987; 42:597

About the author

Dan Poenaru is a pediatric surgeon working in Africa and Canada. Following medical school studies in Toronto, he trained in general surgery at McGill University, then in pediatric surgery at the Université de Montréal. He has earned a Masters in Health Professions Education (University of Illinois in Chicago), and is currently completing a Masters in International Development (William Carrey University, California). He has practised academic surgery for 10 years in Kingston, Canada, 8 years in Kijabe, Kenya, and 2 years in Addis Ababa, Ethiopia. He now divides his professional time between Montréal, Canada, and various sites in Africa.

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Dan is happily married to Dita who is a nursing clinician, educator, and missions mobilizer; they have two adult children in Canada.