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SCIENTIFIC REVIEW

Disability Weights for Pediatric Surgical Procedures: A Systematic Review and Analysis

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Abstract

Background Metrics to measure the burden of surgical conditions, such as disability weights (DWs), are poorly defined, particularly for pediatric conditions. To summarize the literature on DWs of children's surgical conditions, we performed a systematic review of disability weights of pediatric surgical conditions in low- and middle-income countries (LMICs).

Method For this systematic review, we searched MEDLINE for pediatric surgery cost-effectiveness studies in LMICs, published between January 1, 1996, and April 1, 2017. We also included DWs found in the Global Burden of Disease studies, bibliographies of studies identified in PubMed, or through expert opinion of authors (ES and HR).

Results Out of 1427 publications, 199 were selected for full-text analysis, and 30 met all eligibility criteria. We identified 194 discrete DWs published for 66 different pediatric surgical conditions. The DWs were primarily derived from the Global Burden of Disease studies (72%). Of the 194 conditions with reported DWs, only 12 reflected pre-surgical severity, and 12 included postsurgical severity. The methodological quality of included studies and DWs for specific conditions varied greatly.

Interpretation It is essential to accurately measure the burden, cost-effectiveness, and impact of pediatric surgical disease in order to make informed policy decisions. Our results indicate that the existing DWs are inadequate to accurately quantify the burden of pediatric surgical conditions. A wider set of DWs for pediatric surgical conditions needs to be developed, taking into account factors specific to the range and severity of surgical conditions.

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Introduction

The Lancet Commission on Global Surgery estimated that 5 billion people do not have access to surgical care, with the majority in low- and middle-income countries (LMICs) [1]. Surgical conditions contribute to at least 30% of the Global Burden of Disease [2]. Surgery has been shown to be cost-effective in LMICs [1, 3, 4], and a large portion of the economic losses related to surgical conditions can be averted by scaling up surgical services [5]. Among children, the burden of surgical disease is even more striking [6], with estimates that up to 85% of children in LMICs have a surgically treatable condition by the age of 15 [7]. Surgical conditions in children can lead to lifelong disabilities, with resultant effects on economic productivity and development in LMICs [8].

An accurate measurement of the burden of children's surgical conditions is critical in estimating the impact of children's surgical care and for informing policy development. Much of the effort of burden of disease measures has been led by the Global Burden of Disease (GBD) project as well as a number of complementary studies [9–13]. However, metrics specific for surgical conditions in children have largely relied on estimates from modeling studies, rather than primary data collection. Improved metrics to quantify the burden of surgical conditions has been identified as an area of much-needed research [8, 14–16].

The burden of disease is most commonly expressed in disability-adjusted life years (DALYs), a composite metric of years of life lost and years of life lived with disability [17]. An essential factor in the DALY formula is the disability weight (DW), an estimate of the severity of a health condition ranging from 0 (perfect health) to 1 (death). Small variations in DWs can lead to large discrepancies in DALY estimates, emphasizing the importance of the DW in burden of disease analysis. Although the GBD and other studies have defined DWs for an increasing number of health conditions, surgical disease remains underrepresented in these metrics, particularly among children [9–12, 15]. A significant drawback to DW values for surgical conditions is that most do not take into account factors such as the spectrum of disease severity. The objective of this study was to summarize the existing literature on DWs for pediatric surgical conditions, and to develop recommendations for improving their use to quantify the burden of surgical disease in children.

Materials and methods

Selection criteria and data sources

We conducted a systematic review of the literature including DWs for pediatric surgical conditions in LMICs. We followed the guidelines described in the 2009 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [18], and this review was prospectively registered in PROSPERO (CRD42017058458).

In collaboration with a clinical librarian, we conducted a search for all English-language publications indexed in MEDLINE (PubMed) based on search terms and MeSH headings meeting inclusion criteria relevant to this review. Our search included studies published between January 1996 and April 2017, with the full search list included in Table 1. We also included studies found in bibliographies identified in PubMed or through expert opinion of authors, including all GBD studies between 1993 and 2015. The search was limited to the pediatric population (defined as children less than 18 years of age), publications reporting economic analyses, DALY estimations and/or disability weights, and studies conducted in LMICs. We defined a surgical condition as one that could be treated with an operation, even if other noninvasive treatments were available. We excluded publications that did not examine surgical conditions or procedures, lacked disability weights, or were outside the pediatric population, as well as studies using non-primary data sources or other systematic reviews.

Publication selection and data extraction

Abstracts of identified publications were screened by two reviewers (SS and SL) to select those for full-text review. Disagreements were settled by discussion between the two reviewers and other co-authors (ES and HR). For each included publication, two reviewers (SS and ES) independently collected data including study location, age range of participants, and study population inclusion criteria. Disability weight information included the source, DW value, surgical condition, and whether the DW incorporated surgical factors. Discrepancies between the two reviewers regarding extracted data were resolved through discussion.

Disability weight reference tables

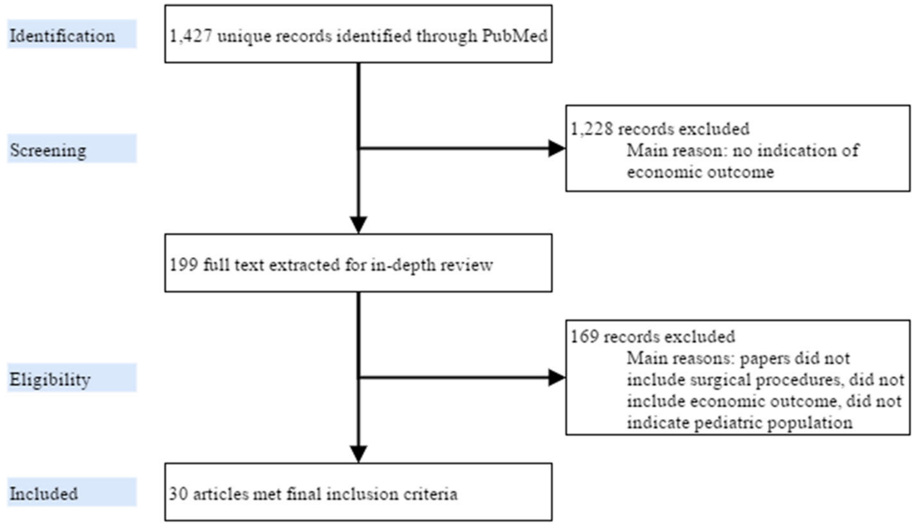
Reference tables of DWs for health conditions requiring surgical care were created by compiling values from all reviewed studies. Data summarized for each reviewed study included study setting and health condition category.

Table 1 Systematic review search terms

Set number	Search terms for PubMed	Results
1. Pediatrics	("Pediatrics"[Mesh] OR "Adolescent"[Mesh] OR "Child"[Mesh] OR "Infant"[Mesh] OR infant[tiab] OR neonat*[tiab] OR child[tiab] OR children[tiab] OR pediatric[tiab] OR adolescent*[tiab] OR teenage*[tiab] OR teen[tiab] OR teens[tiab] OR youth[tiab] OR youths[tiab] OR kid[tiab] OR kids[tiab])	3434047
2. Surgery	("Surgical Procedures, Operative"[Mesh] OR "surgery" [Subheading] OR "surgery"[tiab] OR "surgical"[tiab] OR "Hernia, Inguinal"[Mesh] OR "Testicular Hydrocele"[Mesh] OR "Cleft Lip"[Mesh] OR "Cleft Palate"[Mesh] OR "Cataract"[Mesh] OR "Anus, Imperforate"[Mesh])	3710845
3. LMICs	("Developing Countries"[Mesh] OR Africa[Mesh] OR "Central America"[Mesh] OR "American Samoa"[tiab] OR "Cambodia"[tiab] OR "China" [tiab] OR "Fiji"[tiab] OR "Indonesia"[tiab] OR "Kiribati"[tiab] OR "Korea"[tiab] OR "Lao"[tiab] OR "Malaysia"[tiab] OR "Marshall Islands"[tiab] OR "Micronesia"[tiab] OR "Mongolia"[tiab] OR "Myanmar"[tiab] OR "Palau"[tiab] OR "Papua New Guinea"[tiab] OR "Philippines"[tiab] OR "Samoa"[tiab] OR "Solomon Islands"[tiab] OR "Thailand"[tiab] OR "Timor-Leste"[tiab] OR "Tuvalu"[tiab] OR "Tonga"[tiab] OR "Vanuatu"[tiab] OR "Vietnam"[tiab] OR "Albania"[tiab] OR "Armenia"[tiab] OR "Azerbaijan"[tiab] OR "Belarus"[tiab] OR "Bosnia"[tiab] OR "Bulgaria"[tiab] OR "Georgia"[tiab] OR "Hungary"[tiab] OR "Kazakhstan"[tiab] OR "Kosovo"[tiab] OR "Kyrgyz Republic"[tiab] OR "Macedonia"[tiab] OR "Moldova"[tiab] OR "Montenegro"[tiab] OR "Romania"[tiab] OR "Serbia"[tiab] OR "Tajikistan"[tiab] OR "Turkey"[tiab] OR "Turkmenistan"[tiab] OR "Ukraine"[tiab] OR "Uzbekistan"[tiab] OR "Argentina"[tiab] OR "Belize"[tiab] OR "Bolivia"[tiab] OR "Brazil"[tiab] OR "Colombia"[tiab] OR "Costa Rica"[tiab] OR "Cuba"[tiab] OR "Dominica"[tiab] OR "Dominican Republic"[tiab] OR "Ecuador"[tiab] OR "El Salvador"[tiab] OR "Grenada"[tiab] OR "Guatemala"[tiab] OR "Guyana"[tiab] OR "Haiti"[tiab] OR "Honduras"[tiab] OR "Jamaica"[tiab] OR "Mexico"[tiab] OR "Nicaragua"[tiab] OR "Panama"[tiab] OR "Paraguay"[tiab] OR "Peru"[tiab] OR "St. Lucia"[tiab] OR "St. Vincent"[tiab] OR "Suriname"[tiab] OR "Venezuela"[tiab] OR "Algeria"[tiab] OR "Djibouti"[tiab] OR "Egypt"[tiab] OR "Iran"[tiab] OR "Iraq"[tiab] OR "Jordan"[tiab] OR "Lebanon"[tiab] OR "Libya"[tiab] OR "Morocco"[tiab] OR "Syria*"[tiab] OR "Tunisia"[tiab] OR "West Bank and Gaza"[tiab] OR "Yemen"[tiab] OR "Afghanistan"[tiab] OR "Bangladesh"[tiab] OR "Bhutan"[tiab] OR "India"[tiab] OR "Maldives"[tiab] OR "Nepal"[tiab] OR "Pakistan"[tiab] OR "Sri Lanka"[tiab] OR "Angola"[tiab] OR "Benin"[tiab] OR "Botswana"[tiab] OR "Burkina Faso"[tiab] OR "Burundi"[tiab] OR "Cameroon"[tiab] OR "Cabo Verde"[tiab] OR "Central African Republic"[tiab] OR "Chad"[tiab] OR "Comoros"[tiab] OR "Congo"[tiab] OR "Cote d'Ivoire"[tiab] OR "Ivory Coast"[tiab] OR "Eritrea"[tiab] OR "Ethiopia"[tiab] OR "Gabon"[tiab] OR "Gambia"[tiab] OR "Ghana"[tiab] OR "Guinea"[tiab] OR "Guinea- Bissau"[tiab] OR "Kenya"[tiab] OR "Lesotho"[tiab] OR "Liberia"[tiab] OR "Madagascar"[tiab] OR "Malawi"[tiab] OR "Mali"[tiab] OR "Mauritania"[tiab] OR "Mauritius"[tiab] OR "Mozambique"[tiab] OR "Namibia"[tiab] OR "Niger"[tiab] OR "Nigeria"[tiab] OR "Rwanda"[tiab] OR "São Tomé and Principe"[tiab] OR "Senegal"[tiab] OR "Seychelles"[tiab] OR "Sierra Leone"[tiab] OR "Somalia"[tiab] OR "South Africa"[tiab] OR "South Sudan"[tiab] OR "Sudan"[tiab] OR "Swaziland"[tiab] OR "Tanzania"[tiab] OR "Togo"[tiab] OR "Uganda"[tiab] OR "Zambia"[tiab] OR "Zimbabwe" OR "low resource"[tiab] OR "under-resourced"[tiab] OR "resource poor"[tiab] OR "under-developed"[tiab] OR "underdeveloped"[tiab] OR "developing country"[tiab] OR "developing countries"[tiab] OR "developing world"[tiab] OR "third world"[tiab] OR global[tiab] OR lmic[tiab] OR (low[tiab] AND middle[tiab] AND income[tiab])	1143071
4. Step 1	1&2&3	26,940
5. Econ	("Costs and Cost Analysis"[Mesh]) OR "economics" [subheading] OR "economic" [tiab] OR "disability" [tiab])	629873
6. Step 2	4&5	2049
7. Publication date	Filters: Publication date from 1996/01/01 to 2016/12/31	1422 (currently 1429)

Additional information included whether the DW applied to both children and adults, whether it was specific to pre-treatment or post-treatment states, methods of DW assignment, and DW source. If the DW source was assigned to one of the GBD studies, the specific year of study was noted. We also classified conditions and the corresponding DWs by type of specialty, including cardiovascular surgery, ear/nose/throat (ENT) surgery, general surgery, neurosurgery, ophthalmology, orthopedic surgery, plastic and reconstructive surgery, urology, and transplantation. In addition to DWs collected from the reviewed studies, we also included DWs for any other pediatric surgical condition noted in any of the GBD studies but not specifically cited in the reviewed studies.

Fig. 1 Flow chart using the PRISMA statement for the systematic review



Publication grading

To assess the quality of each report, we adapted criteria from a recently developed checklist by Shrime et al. used for cost-effectiveness analyses in global surgery [19]. We included three criteria pertinent to DW assignment:

1. does the report use DWs from the GBD studies (if available)? If the DW was unavailable, was it is calculated from available data using a multiplicative formulation?
2. subjective estimation of DWs is avoided and
3. the credibility of DW estimates is confirmed by comparing against other DWs and conditions of similar magnitude.

We chose specifically not to use the Drummond checklist [20], which is often used for quality grading of cost-effectiveness analyses of clinical trials, as these criteria are not applicable to the topic of DW assignment.

We examined each report to assess whether the authors included key elements, including DW source, method of DW estimation, and comparison of DWs against those of other conditions. Two observers independently graded each publication, with disagreements resolved by discussion.

Results

We identified 1427 publications through our initial search. A total of 199 of these were selected for full-text review based on title and abstract. A total of 30 publications were selected to be included in the final review (Fig. 1).

Of the 30 publications, 10 were limited to pediatric populations, while the other 20 spanned both adult and pediatric populations. Nine studies were conducted in African countries, with the rest originating in Asia ($n = 7$), Latin America ($n = 6$), or in multiple countries ($n = 8$). There were 66 surgical conditions with an assigned DW in a variety of surgical specialties, with the most common specialties including orthopedics (44%), and neurosurgery (14%) (Table 2). We identified a total of 194 distinct DWs for the 66 surgical conditions. The conditions with the most distinct DWs included cleft lip and palate (combined 14 DWs), hearing loss (15 DWs), and hydrocephalus (9 DWs). Of all reported conditions, the least commonly cited were for cardiovascular and urological conditions.

Most existing DWs did not account for disease heterogeneity or severity. For example, there were only two DWs for congenital heart disease, a broad spectrum of conditions ranging from minimally symptomatic (such as patent ductus arteriosus) to severe near-fatal (such as hypoplastic left heart syndrome). Furthermore, there was a wide variability in some DW estimates. For instance, the DW for cleft palate ranged from 0.10 from the GBD study to 0.38 elsewhere. Other conditions, such as gastroschisis and myelomeningocele cited DWs of 1.0, suggesting that the duration spent living with this condition is equivalent to death.

Disability weight sources

Overall, most DWs (72%) were derived from the GBD studies, followed by secondary studies using GBD data (13%), expert opinion (12%), and the Dutch Disability Weight Study (DDW) (3%) (Fig. 2). Of the 26 DWs for

Table 2 Reported disability weights

Surgical condition	GBD DW	DW from other sources
Cardiovascular		
Congenital heart anomalies	0.323 [21]	0.036 [22]
ENT		
Face bones fracture	0.223 [21]	
	0.173 [11]	
	0.067 [10]	
Hearing loss		
Mild[a]	0.000 [21]	0.04 [23, 24]
	0.005 [11]	
	0.01 [10]	
Moderate[a]	0.12 [21]	
	0.023 [11]	
	0.027 [10]	
Severe[a]	0.333 [21]	
	0.032 [11]	
	0.158 [10]	
Profound[a]	0.333 [21]	
	0.031 [11]	
	0.204 [10]	
Complete[a]	0.033 [11]	
	0.215 [10]	
Otitis media		
Chronic infection	0.023 [21]	
Deafness	0.229 [21]	
General surgery		
Cystic echinococcosis		0.239[b] [25, 26]
Appendicitis	0.463 [21]	
Esophageal atresia	0.85 [21]	
Intestinal atresia		0.834 [27]
		0.9 [28]
Inguinal hernia		0.0 [29]
		0.096 [30]
		0.3 [30]
		0.5 [31]
Grade H2		0.1 [29]
Grade H3a, b, c		0.5 [29]
Grade H4a		0.8 [29]
Grade H4b, H4c		1.0 [29]
Hirschsprung's disease		0.72 [28, 32]
After colostomy		0.17 [27]
Before colostomy		0.526 [27]
Anorectal malformations (imperforate anus)		0.85 [32, 33]
Mild		0.28 [27, 28]
Severe		0.81 [27, 28]
Anorectal atresia	0.85 [21]	
Abdominal wall defects	0.85 [21]	
Mild		0.421 [27]

Table 2 continued

Surgical condition	GBD DW	DW from other sources
Severe		0.758 [27]
		0.92 [28]
Gastroschisis		1 [34]
Neurosurgery		
Dermoid cyst		0.013 [35, 36]
Skull fracture	0.431 [21]	0.18 [37]
	0.073 [11]	
	0.071 [10]	
Erb's palsy		0.308[a] [37]
Lipomyelomeningocele		0.337 [35, 36]
		0.44 [35, 36]
Atretic thoracic meningocele		0.398 [35, 36]
Hydranencephaly		1 [35, 36]
Hydrocephalus		0.4 [38]
		0.74 [27, 28]
		1 [35, 36]
With IQ 70-84		0.09 [39]
With epilepsy		0.113 [39]
With motor deficits		0.17 [39]
With IQ 50-69		0.29 [39]
With visual or auditory deficits		0.33 [39]
With IQ $\leq$50		0.62 [39]
Epilepsy	0.113 [21]	
	0.42 [11]	
Less severe	0.263 [10]	
Severe	0.657 [11]	
	0.552 [10]	
Spina bifida	0.593 [21]	
Mild		0.336 [27]
		0.46 [28]
Severe (paraplegia)		0.765 [27]
		0.87 [28]
Myelomeningocele		0.334 [22]
		0.425 [36]
		1 [36]
Ophthalmology		
Corneal wound		0.1 [40]
Pterygium		0.1 [40]
Foreign body removal		0.4 [40]
Cataract		0.6 [40]
Low vision	0.17 [21]	
Blindness	0.6 [21]	
Bilateral		0.9 [40]
Glaucoma		
Low vision	0.17 [21]	
Blindness	0.57 [21]	
Trachoma		
Low vision	0.17 [21]	

Table 2 continued

Surgical condition	GBD DW	DW from other sources
Blindness	0.581 [21]	
Orthopedic		
Shoulder dislocation		0.074 [41, 42]
Hip dislocation (congenital)		0.074 [41, 42]
Macrodactyly		0.102 [37]
Bone cysts		0.18 [37]
Club foot		0.2 [38, 40]
Fractures		
Hand	0.1 [21]	
	0.016 [11]	
	0.014 [10]	
Foot	0.077 [21, 41, 42]	
	0.033 [11]	
	0.026 [10]	
Clavicle, scapula, or humerus	0.153 [21]	
	0.053 [11]	
	0.035 [10]	
Femur (other than neck)	0.372 [21]	
	0.053 [11]	
	0.042 [10]	
Radius or ulna	0.18 [21]	
	0.05 [11]	
	0.043 [10]	
	0.12–0.24 [9]	
Ankle	0.196 [21, 41, 42]	
	0.07 [11]	
	0.055 [10]	
Patella, tibia, or fibula	0.271 [21]	
	0.07 [11]	
	0.055 [10]	
Sternum or ribs	0.199 [21]	
	0.15 [11]	
	0.103 [10]	
Vertebral column	0.266 [21]	
	0.132 [11]	
	0.111 [10]	
Pelvis	0.247 [21]	
	0.194 [11]	
	0.182 [10]	
Neck of femur	0.372 [21]	
	0.388 [11]	
	0.402 [10]	
Humerus		0.153 [41, 42]
Tibia		0.271 [41, 42]
Femur		0.372 [41, 42]
		0.4 [40]
Other fractures	0.003 [11]	
	0.005 [10]	

Table 2 continued

Surgical condition	GBD DW	DW from other sources
Amputations		
Finger	0.102 [21]	
	0.03 [11]	
	0.005 [10]	
Toe	0.102 [21]	
	0.008 [11]	
	0.006 [10]	
Thumb	0.165 [21]	
	0.013 [11]	
	0.011 [10]	
One arm	0.308 [21]	
	0.12 [11]	
	0.118 [10]	
One leg	0.3 [21]	
	0.164 [11]	
	0.173 [10]	
Below the knee	0.24–0.36 [9]	
Both arms	0.359 [11]	
	0.383 [10]	
Both legs	0.494 [11]	
	0.443 [10]	
Primary or stump revisions		0.3 [41, 42]
Plastic and reconstructive		
Cleft lip	0.05 [21, 38, 43, 44]	0.098 [45–48]
		0.18 [4, 18, 28, 49]
		0.2 [40]
		0.122 [22]
		0.076 [27]
Cleft palate	0.103 [21]	0.101 [43, 44]
		0.264 [27]
		0.3 [40]
		0.231 [11, 12, 14, 15, 17, 45–50]
		0.38 [28]
		0.122 [22]
Cleft lip and palate		0.231 [49]
		0.264 [27]
		0.44 [45]
Skin graft		0.4 [40]
Burns		0.4 [40]
<20%	0.002 [21]	
	0.018–0.096 [11][c]	
	0.016–0.141 [10][c]	
>20%	0.255 [21]	
	0.127–0.438 [11][c]	
	0.135–0.455 [10][c]	
Urology		
Undescended testes		0.22 [28]
		0.115 [27]

Table 2 continued

Surgical condition	GBD DW	DW from other sources
Hypospadias		
Mild		0.0 [27]
		0.12 [28]
Severe		0.275 [27]
		0.5 [28]
Transplant		
End-stage renal disease, with transplantation	0.027 [11]	
	0.024 [10]	
End-stage renal disease, on dialysis	0.098 [21]	
	0.571 [10]	
	0.573 [11]	
Cirrhosis of the liver	0.33 [21]	
	0.194 [11]	
	0.178 [10]	

[a]Additional GBD DW for hearing loss severities with ringing; mild, with ringing: 0.038b, 0.021c; moderate, with ringing: 0.058b, 0.074c; severe, with ringing: 0.065b, 0.261c; profound, with ringing: 0.088b, 0.277c; complete, with ringing: 0.092b, 0.316c

[b]DW used is a proxy for liver cancer

[c]Range of DW reported only. GBD reports DW within this range according to surface area and location of burns

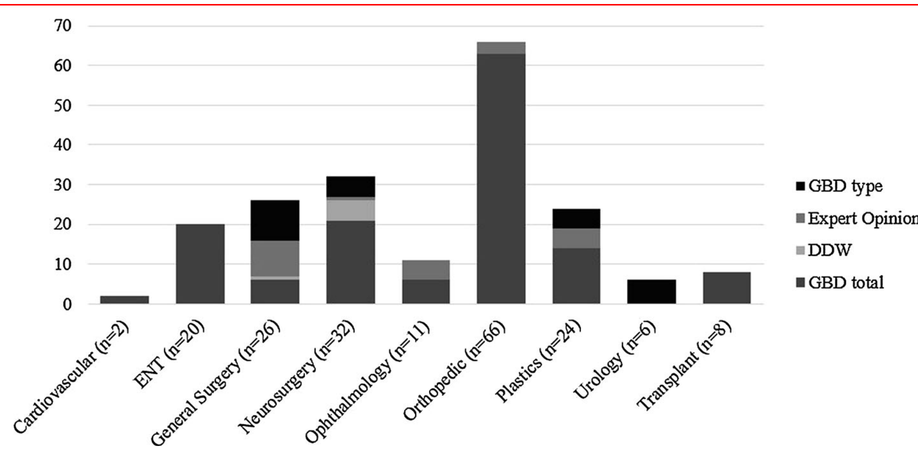


Fig. 2 Disability weight estimation source

general surgery, six were derived from the GBD studies and nine were obtained by expert opinion. Nearly all of the DWs cited in orthopedics and ophthalmology were derived from the GBD studies (63 out of 66 DWs) and the remainder by expert opinion. Over half of the DWs for plastics, ophthalmology, and neurosurgery were derived from the GBD studies. All DWs for ENT, cardiovascular and transplant conditions were also derived from the GBD study.

Disability weight values and incorporation of surgical factors

Of all surgical conditions encountered, only 18% ($n = 12$) incorporated measures of disease severity, and only 18% ($n = 12$) accounted for a range of postoperative disability states (Table 3).

Quality of evidence (Table S1)

Of the 30 included publications, 17% ($n = 5$) scored positively to all three quality questions using our modified Shrime checklist (Table S1). Twenty-four publications

Table 3 Pediatric surgical condition pre-treatment and post-treatment disability weights

	Disability weight	
	Pre-treatment	Post-treatment
ENT		
Hearing loss	0.333 [21]	0.120
General		
Gastroschisis	1 [34]	0.2
Neurosurgery		
Epilepsy	0.113 [21]	0.072, 0.319
Ophthalmology		
Cataract	0.6 [21]	0.488
Orthopedic		
Amputation		
One arm	0.118 [10]	0.039
Both arms	0.383 [10]	0.123
One leg	0.173 [10]	0.039
Both legs	0.443 [10]	0.088
Plastic and reconstructive		
Cleft lip	0.098 [45–48]	0.16
Cleft palate	0.231 [11, 12, 14, 15, 17, 45–50]	0.015
	0.44 [21, 22, 45]	0.015
Cleft lip or palate	0.122 [22]	0.021
Burns	0.002 [21]	0.001

Post-treatment DW notes: Epilepsy: treated seizure free, treated non-seizure free

NP not provided, *ENT* ear, nose, and throat

(80%) used DWs from the GBD studies or variants of GBD methods if the DW weight was unavailable in any GBD study. Subjective estimation of DWs was avoided in 20 of the studies (67%), while only five studies (17%) confirmed the validity of the DWs against other DWs of similar magnitude.

Discussion

Main findings

As the global health agenda increasingly recognizes the importance of surgical care for children, accurate assessment of the burden of surgical disease is essential to guide health care policy [1]. Current methods to assess the burden of disease depend in large part on DW assignment. The DW metric is more than just of academic interest, as even small variations in DW values have a significant impact on burden of disease estimates. We found many challenges in the DW assignment for surgical conditions, including lack of granularity, wide variability in estimates, and lack of inclusion of surgically significant variables such as disease severity and treatment efficacy. These gaps offer research opportunities to improve our understanding of the burden of surgical disease and ultimately improve the health care of children in LMICs.

Global burden of disease studies

It is important to understand the concept of DWs within the context of the GBD studies. For over 25 years, the GBD project along with several supporting groups has provided valuable data on (a) the prevalence of different diseases in the world, (b) their etiological factors and disability weights, and (c) their population burden [9]. Since the original GBD 1990 study which developed a framework for integrating this information on the health of populations [51], regular revisions have been undertaken, with each revision providing an update for the entire series based on incorporation of newly identified data, transitioning from disease-based to health-state DW definitions, and refinement of methods [52, 53].

Disability weight assessment

Disability weights are intended to quantify social preferences for different states of health [17, 54, 55]. These values are expected in part to reflect comparisons of functional disability between different health states [55, 56]. Several methods of DW estimation have been used, including psychometric exercises, standard-gamble, or trade-off methods [57]. Despite ongoing modifications of methods, the use of DWs for pediatric surgical conditions remains problematic for several fundamental reasons, as discussed below.

Problem 1: lack of pediatric surgical DWs

The DW values for many pediatric surgical conditions remain absent or are assigned at a generalized level, making estimates of the burden of surgical conditions challenging and sometimes almost meaningless [15]. For example, the GBD 2004 study [21] included DWs for only 7 congenital surgical conditions, themselves adapted from the GBD 1990 study [9, 58]. In the absence of validated DWs for a sufficient number of conditions, we have found in this review that much of the surgical literature has used DW proxies, many of which have non-applicable levels of disability. For example, the cited DW for liver cancer has been used for cystic echinococcosis, and the DW for arm amputation has been used for Erb's palsy [25, 26, 59]. Alternatively, DWs have also been estimated by expert opinion, often using ballpark disability descriptions based on other surgical conditions [4]. In one study, for example, the DWs for several inguinal hernia severity grades were estimated with general disability descriptions and informed by one co-author, not an expert panel [29]. As these DWs are usually developed by a small number of experts in a limited number of settings, they are intrinsically subjective and tend to either underestimate or overestimate the actual burden of surgical conditions [19].

Problem 2: lack of disease severity and granularity of DWs for surgical conditions

Currently defined DWs for most children's surgical conditions are overgeneralized, resulting in inaccurate representation of the spectrum of disability associated with surgical conditions. Within the GBD study there has been an attempt to incorporate disease severity for a subset of causes, with severity defined at Level 4 of the GBD hierarchy. However, the number of surgical causes which have associated severity levels remains low even in the latest GBD update. For example, there is no post-treatment severity stratification for the DWs of anorectal malformations, which clinically can have a wide severity range depending on the underlying defect, operative complications, and overall surgical quality. Having such few DW values summarize the broad range of pediatric surgical conditions leads to almost meaningless estimates of the burden of health conditions in children and renders it difficult to systematically quantify the value of surgical intervention.

Although the DWs of many treated non-surgical health conditions are increasingly being assigned, existing DWs for many surgical conditions do not often reflect differences between treated and untreated disease, leading to difficulty assessing the value of surgical therapies. Furthermore, the lack of post-treatment DWs provides an inaccurate picture of disability lived with a condition even if treated. Although there are methods to estimate residual disability after treatment [56], the resulting DW is a rough estimate rather than an empirically generated value through rigorous methods, such as pairwise comparisons.

Problem 3: lack of external and internal validation

All DW calculations should be referenced against other health conditions of similar magnitude as well as internally across studies to confirm their validity [19]. Our review found few publications which validated the DWs by comparison of health states with DWs of the same magnitude. This lack of validation poses a significant challenge in comparing outcome measures. As well, this lack of validation leads to unrealistic assessments of disability associated with some surgical conditions. For example, the DW assigned for undescended testes exceeds many DWs of amputations. Similarly, some DWs are nonsensical, such as extreme DW values of 0 (equivalent to perfect health) for inguinal hernia and 1 (equivalent to death) for meningomyelocele and gastroschisis [34, 36]. Our review found that 33% of the studies used subjective estimation of DWs without any comparison to other health conditions.

Problem 4: limited settings for disability weight studies

We found a limited number of countries from which the DWs originated, leading to lack of generalizability of findings. Our review found 30 publications from only 20 countries, with two studies focusing on unspecified Sub-Sahara African countries. We note that in the GBD 2010 study, results from pairwise comparisons of DWs from non-fatal states were similar in Indonesia, Tanzania, Peru, and the USA [11]. However, others have noted that these DW correlations give an exaggerated impression of cross-country agreement and conceal the need for validation of individual country DWs [60]. Extension of DWs for pediatric surgical disease across global settings as additional data becomes available should improve our understanding of burden of disease in different countries.

Limitations

Some of the key limitations in the study include challenges with the precise definition of a pediatric surgical condition and the potential for missing publications with other DWs. Regarding the former, we included DWs for all health conditions that could be treated surgically even if other noninvasive treatment options were available. For example, hearing loss may or may not be treated surgically in children depending on the severity of the condition or availability of resources. However, to remain inclusive in our selection process, we did not exclude these conditions. Regarding the potential for missing publications, our search criteria was broadly defined to attempt to capture DWs from numerous types of studies, including economic analyses and standard-gamble approaches. We believe this approach captured the majority of DWs for pediatric surgical conditions, particularly since we included all GBD studies.

Conclusions (the way forward)

The use of DWs is critical for estimating the burden of disease and guide policy development to improve the surgical care of children around the world. There are large gaps in the existing DWs for surgical. Although the objective of this manuscript was to provide an overview assessment of DWs used for pediatric surgery, there are several new opportunities for research. Our specific recommendations include:

1. Expand the number of assigned DWs to include all pediatric surgical conditions with a large burden of disease and conditions which have been identified as essential for a functional health system by expert bodies (such as the WHO or Global Initiative for Children's Surgery). Although the lack of DWs is in large part due to limited available primary data on surgical conditions, the recent efforts of the Global Initiative for Children's Surgery are expected to address this data gap [32]. We view the lack of DWs in children's surgery as a vast research opportunity, and DW estimates for all high-burden pediatric surgical conditions across all specialties need to be developed to optimally assist policy development.
2. Incorporate surgical-specific factors into methods for DW assignment, such as range of disease severity, treatment efficacy, pre- and post-treatment states, and impact of surgical complications. The most rigorous methodology for DW assignment has evolved in the GBD studies from visual analogue scales to the most recent technique of pairwise comparisons. Utilizing the most recent refinement of pairwise comparisons, health states for pediatric surgical conditions could be defined based on untreated disease severity levels, thereby creating severity-specific DWs, and based on post-treatment disability, thereby incorporating residual disability.
3. A key aspect to pairwise comparison is writing lay descriptions for health states. We are advocating for stratified health states based on pre-surgical severity and post-treatment efficacy. Thus, lay descriptions for each stratification will need to be developed for pediatric surgical conditions. These descriptions should be constructed with attention to wording choices and may vary by location or audience. For example, rating of the health states may differ by culture, interviewee (i.e., mothers may value health differently than physicians), and country. Further research regarding these nuances should be conducted.

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Compliance with ethical standards

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