

# 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 10-year period.

**Methods** Anonymized data of all primary cleft lip and cleft palate procedures in the Smile Train database were analyzed and disability-adjusted life years (DALYs) calculated using country-specific life expectancy tables, established disability weights, 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 and 2011. 547,769 records contained complete data available for the study; 58 % were cleft lip and 42 % cleft palate. Averted DALYs ranged between 1.46 and 4.95 M. 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 [1–3]. 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.

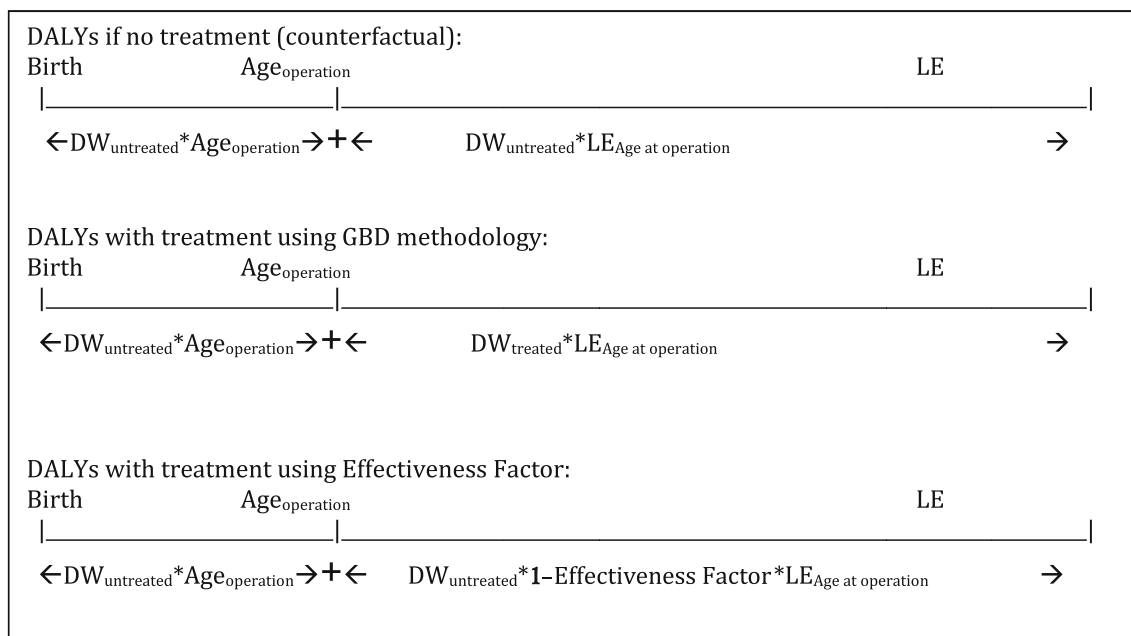
This study uses a framework derived from previous work [4–11] to examine the economic value of the programs of a non-governmental organization. Smile Train (ST) supports the surgical care of patients with cleft lip and palate (CLP) in low- and middle-income countries (LMICs). ST identifies and trains 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

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**Fig. 1** Diagram of age milestones and calculations used for estimating averted DALYs

specialty surgical hospital platform described by Shrimet 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 difference between these two figures, is presented as the economic effect of the program. The difference in DALYs is also presented.

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:

$$\text{BoD}_{\text{without intervention}} = \left( DW_{\text{untreated}} * \text{age}_{\text{operation}} \right) + \left( DW_{\text{untreated}} * LE_{\text{Age at treatment}} \right),$$

where  $DW_{\text{untreated}}$  = disability weight for CLP untreated (both  $DW_{\text{untreated}}$  and  $DW_{\text{treated}}$  from the GBD study [16] and  $LE_{\text{Age at treatment}}$  = 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:

$$\text{BoD}_{\text{with intervention}} = \left( DW_{\text{untreated}} * \text{age}_{\text{operation}} \right) + \left( DW_{\text{treated}} * LE_{\text{Age at operation}} \right)$$

DALYs incurred with the intervention were also estimated by the method published by McCord et al. [11] and widely followed in the surgical literature [18–21]. 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 and 94 % for cleft lip and between 25 and 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  $DW_{\text{treated}}$ :

$$\text{BoD}_{\text{with intervention}} = (\text{DW}_{\text{untreated}} * \text{Age}_{\text{operation}}) + (\text{DW}_{\text{untreated}} * (1 - \text{EF}) * \text{LE}_{\text{Age at operation}})$$

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.

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 Fig. 2.

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 and 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.

Total averted DALYs achieved by method of calculation and year are shown in Fig. 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.

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 9907 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.

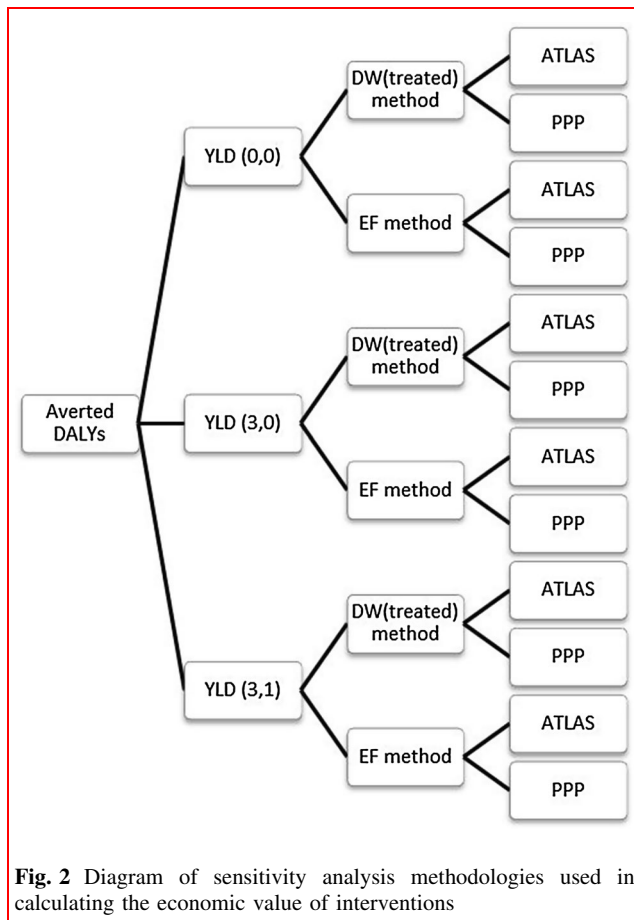
The overall cost for the interventions studied was estimated at 197 million USD, which represents between 0.7 and 6.6 % of the estimated economic gain, depending on the valuation method used. In terms of cost-effectiveness, this represents between \$40 and 135/DALY.

## 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 are viewed in the context of an estimated 25 million DALYs for overall surgical BoD in Africa [26], and the mean averted DALYs per patient are similar to those estimated for hydrocephalus and inguinal hernia [6, 27].

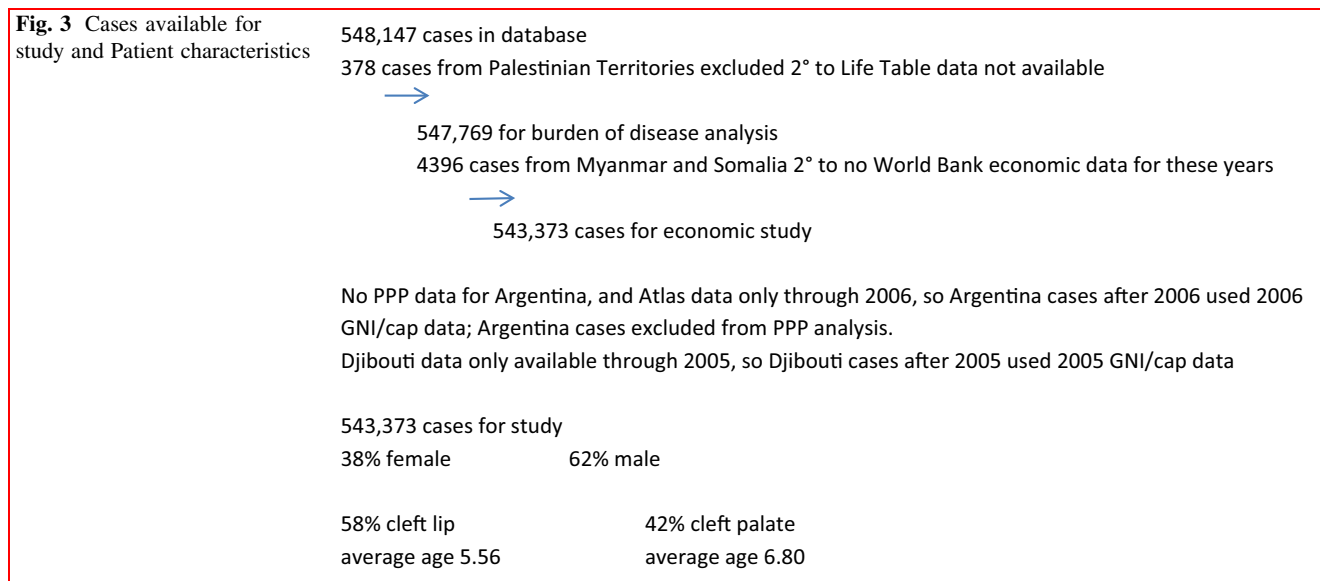
The economic value of improved health resulting from the surgical interventions was determined using GNI per capita, based on the premise that each individual theoretically contributed an equal share. A first assumption was that CLP affects equally all sectors of society regardless of socioeconomic stratum,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

**Table 1** Averted burden of disease in DALYs per patient and total

	0.0	3.0	3.1
Sum of averted DALYs using Eff factor			
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 versus 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 versus 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

	Atlas 0.0	PPP 0.0	Atlas 3.0	PPP 3.0	Atlas 3.1	PPP 3.1
Average economic gain per pt using DW untreated versus treated (USD)						
CP	32,216	82,405	13,693	35,303	16,478	42,629
CL	10,362	27,639	4,391	11,843	5102	13847
Total	19,538	50,634	8297	21,693	9878	25,932
	Atlas 0.0	PPP 0.0	Atlas 3.0	PPP 3.0	Atlas 3.1	PPP 3.1
Average economic gain using effectiveness factor (USD)						
CP	17,227	44,064	7322	18,877	8811	22,795
CL	9907	26,426	4198	11,323	4878	13,239
Total	12,981	33,832	5510	14,495	6529	17,251
	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
Sum of economic gain using DW untreated versus treated (USD)						
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 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
Sum of economic gain using effectiveness factor (USD)						
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	9,449,625,14

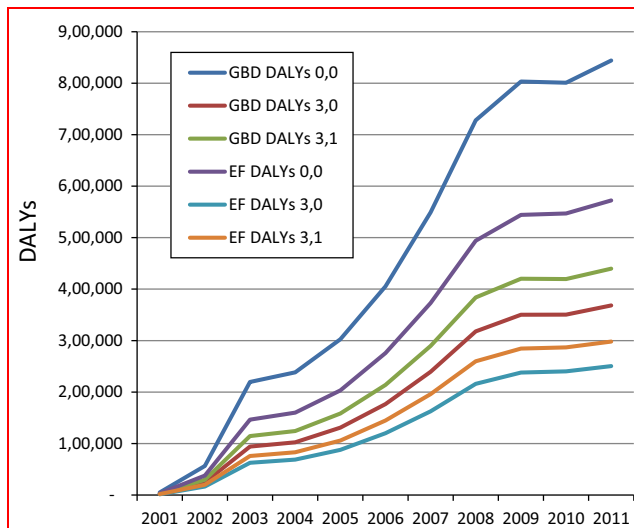


willingness to pay, value of a statistical life, or direct income studies may be the focus of future studies.

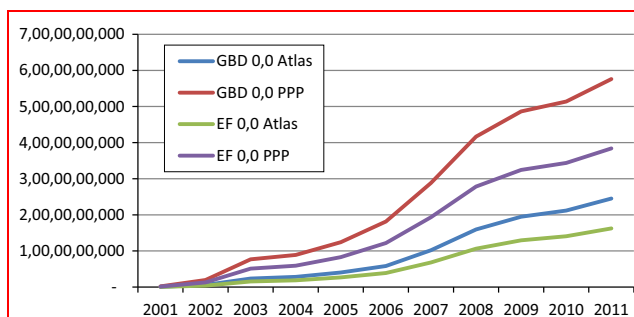
The GNI data indicate a very substantial economic impact, in keeping with other reports on surgery in limited resource settings. Using a similar methodology, Alkire et al. estimated the economic impact of treating CLP in

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



**Fig. 4** Averted BoD through cleft lip and palate surgery by method and year



**Fig. 5** Economic impact by year and calculation method without age weighting or discounting

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 compound 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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