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RESEARCH ARTICLE

Cost analysis of outpatient services for major external structural birth defects: An ingredient approach in selected hospitals in Kiambu County, Kenya [version 1; peer review: 1 approved with reservations]

George N. Agot , Joseph K. Wang'ombe, Marshal M. Mweu

School of public health, College of health sciences, University of Nairobi, Nairobi, Kenya, 30197-00100, Kenya

V1 First published: 07 May 2021, 10:359
<https://doi.org/10.12688/f1000research.52521.1>
Latest published: 07 May 2021, 10:359
<https://doi.org/10.12688/f1000research.52521.1>

Abstract

Background: Major external structural birth defects are known to exert an enormous economic burden on individuals and health services; however, they have been vastly unappreciated and underprioritized as a public health problem in settings where cost analyses are limited. **Objective:** The objective of this study was to conduct a cost analysis of outpatient services for major external structural birth defects in selected hospitals in Kiambu County, Kenya. **Methods:** A hospital-based cross-sectional study design was adopted in four hospitals where an ingredient approach was used to retrospectively gather data on cost drivers for interventions consisting of castings, bracings, and tendonectomies for the under-fives from health care providers' perspectives for a one-year time horizon (January 1st, 2018, to December 31st, 2018). The hospitals were selected for providing outpatient corrective and rehabilitative services to the under-fives. Prevalence-based morbidity data were extracted from outpatient occupational therapy clinic registers, whereas staff-time for the hospitals' executives comprising the medical superintendents, chief nursing officers, orthopedic surgeons, and health administrative officers were gathered through face-to-face enquires from the occupational therapists being the closest proxies for the officers. Following a predefined inclusion criterion, 349 cases were determined, and associated cost drivers identified, measured, and valued (quantified) using prevailing market prices. The costs were categorized as recurrent, and unit economic costs calculated as average costs, expressed in U.S Dollars, and inflated to the U.S Dollar Consumer Price Index from January 2018 to December 2018. **Results:** The unit economic cost of all the cases was estimated at \$1,139.73; and \$1,143.51 for neural tube defects, \$1,143.05 for congenital talipes equinovarus, and \$1,109.81 for congenital pes planus. **Conclusions:** The highest economic burden of major external structural birth

Open Peer Review

Approval Status ?

1

version 1

07 May 2021

?
view

1. Vincent Okungu , University of Nairobi,
Nairobi, Kenya
The END Fund, Nairobi, Kenya

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defects in the county was associated with neural tube defects, followed by congenital pes planus despite having the fewest caseloads.

Keywords

Major external structural birth defects, costing analysis, outpatient services, ingredient approach, Kenya

This article is included in the **Health Services** gateway.

Corresponding author: George N. Agot (nyadimogeorge@gmail.com)

Author roles: **N. Agot G:** Conceptualization, Data Curation, Formal Analysis, Funding Acquisition, Investigation, Methodology, Project Administration, Resources, Software, Supervision, Validation, Visualization, Writing – Original Draft Preparation, Writing – Review & Editing; **K. Wang'ombe J:** Conceptualization, Supervision, Validation; **M. Mweu M:** Conceptualization, Supervision, Validation

Competing interests: No competing interests were disclosed.

Grant information: The author(s) declared that no grants were involved in supporting this work.

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How to cite this article: N. Agot G, K. Wang'ombe J and M. Mweu M. **Cost analysis of outpatient services for major external structural birth defects: An ingredient approach in selected hospitals in Kiambu County, Kenya [version 1; peer review: 1 approved with reservations]** F1000Research 2021, **10**:359 <https://doi.org/10.12688/f1000research.52521.1>

First published: 07 May 2021, **10**:359 <https://doi.org/10.12688/f1000research.52521.1>

Introduction

Major external structural birth defects (MESBDs) are defined as physical abnormalities of intrauterine origin present from birth, detectable visually, and having significant health and development impacts¹⁻⁴. These defects are potentially fatal and those children who survive beyond infancy require substantial economic resources to deal with lifelong disabilities⁵⁻⁹. Worldwide, approximately 134 million births reportedly occur yearly, of which 7.9 million (6%) are born with at least a major birth defect, mostly affecting the central and musculoskeletal systems^{1-4,10}. Although about 3.3 million of these children die before they are five years old, the 3.2 million who survive may be disabled for life if sufficient resources are not dedicated to corrective and rehabilitative health services^{1,2,8}.

MESBDs continue to occur exerting an enormous economic burden on individuals and health services in developing countries; however, they have been vastly ignored and unappreciated as public health problems due to limited estimation of the associated costs attributed to the scantiness of, and inaccurately profiled data on cost drivers^{1,2,9,11}. Even though these defects remain a “silent” global public health problem, the highest-burden is shouldered by developing countries due to high prevalence of modifiable risk factors coupled with deficient expertise in economic evaluation studies^{1-4,6,8,9,12-15}. Hospital charges for new-born children born with some forms of birth defect have been reported as four to eight times higher than those without any form of the defects¹⁶. Cost analysis is a partial economic method of evaluating health care programs used to compare the costs of at least two alternative interventions; however, cost studies are still useful even in the absence of comparative interventions as they can establish baseline economic costs of a health intervention¹⁷⁻¹⁹.

Children surviving beyond infancy could require restorative health services to reduce the adverse impacts associated with MESBDs^{1,2}. These interventions are described as corrective and rehabilitative outpatient services consisting of castings, bracings, and tendonectomies whose monetary value is referred to as economic or opportunity costs^{17,18,20-24}. The resources used in the provision of such services could be quantified through micro-costing (bottom-up) using an ingredient approach to gather data on the cost drivers by a step-down full costing technique, activity-based costing, time and motion, surveys, and manager interview techniques^{17,18,20,25-27}. Alternatively, these inputs may be quantified by gross costing (top-down) using historical input outlay^{17,18,20,25-27}.

The range, contexts, and extents of cost elements are determined by economic viewpoints consisting of health care providers', individuals', or societal perspectives that are informed by policy decisions known as the study objectives, and/or questions^{17,18,20,26}. The existing market prices and opportunity costs (forgone benefits) are used to value the inputs in the monetary units categorized as recurrent and capital costs^{17,18,20-24}. These costs are assigned to direct, indirect, and intermediate cost centers using a step-down accounting technique^{17,18,20-24}. The costs of the inputs are sometimes shared among the cost centers thus referred to as overhead (shared or joint) costs which are proportionally

allocated to the respective cost centers for estimating final economic costs^{17,18}. Capital costs determined for more than a one-year time horizon should be considered for the differential-time discounting unlike recurrent costs^{17,18,20-24}. Similarly, statistical and/or sensitivity analysis should be conducted to ascertain the robustness of the evaluation findings because of potential uncertainties arising from sample size determination and data collection methods for the cost drivers¹⁷.

The advancements in medical and surgical interventions are known to reduce the severity of lifelong physical disabilities related to MESBDs; however, their costs are catastrophic and prohibitive to many households and public health care systems globally^{1,2,9,15}. Even though substantial resources are usually allocated to health care systems for the provision of corrective and rehabilitative health services for MESBDs, their costs are seldom estimated due to the rarity and stochasticity of the defects, scantiness of the cost data, inaccurately profiled cost information and inadequate costing expertise in developing countries^{19,20,28,29}. The scarcity of local epidemiological data and differences in the epidemiological study design (prevalence/incidence-based) also impede the accuracy of the profiled cost data in developing countries^{19,20,28,29}.

Economic costs increase the extent to which health services, individuals, and society are affected by MESBDs because of the forgone benefits of not investing in the next best alternative^{16-18,20-24,30}. Corrective and rehabilitative health care services for MESBDs is critical in reducing the severity of lifelong disabilities and improving the quality of life for the affected children, as well as the economic productivity of the affected families^{1,2,9}. Thus, cost analysis is of public health importance in influencing and informing health planning, policy decisions, resource allocations, informing further economic evaluations and assessing health system performance^{17,18,20,22,23,31,32}. Consequently, the objective of this study was to conduct a cost analysis of the outpatient services for MESBDs from the providers' viewpoints using an ingredient approach in selected hospitals in Kiambu County, Kenya.

Methods

Study settings, and designs

The study was conducted in four hospitals consisting of three county referral hospitals (Kiambu, Thika, and Gatundu), and PCEA Kikuyu orthopedic (faith-based) selected for providing corrective and rehabilitative outpatient health services to children born with MESBDs in the county. The three county referral hospitals were purposively selected being the only public hospitals providing these services in the county, whereas PCEA Kikuyu orthopedic hospital (faith-based) was selected by simple randomization using sealed envelopes between two faith-based hospitals for providing the same services in the county. A hospital-based cross-sectional study design was adopted to generate cost data from prevalence-based local morbidity data gathered using an ingredient approach to estimate the economic costs of corrective and rehabilitative outpatient health services from health care providers' perspectives. This was however the best choice of study design for measuring the unit economic cost of health services as an attribute of the

population, and thus provided a snapshot of the burden associated with the 'silent' public health problem and allowed for generalization of the study results in similar hospital settings in the region. Even though incidence data were readily available and easily accessible for the costing activity, prevalence-based data were excessively preferred to improve the accuracy of the profiled cost data and the estimation of the unit economic costs. This was an economic evaluation study, therefore was reported as per the CHEERS (checklist for consolidated health economic evaluation reporting standards) guidelines²⁷.

Study population and eligibility for participation

The study population consisted of all children aged under five years old born to resident women of Kiambu County between January 1st, 2014, and December 31st, 2018. Cases were defined as live births with at least one clinically obvious major external structural birth defect referenced/or described by assistant occupational therapists and/or orthopedic surgeons and presented to the occupational therapy clinics for care from January 1st, 2018, to December 31st, 2018. Caregivers of children born with MESBDs were likely to seek outpatient corrective and rehabilitative health services at the study hospitals whether the children were born in or out of the county. Thus, the eligibility criterion defined above could minimize systemic bias and ensure the reliability of the study results.

Study perspective, time horizon, and discount rate

The data for cost drivers were gathered retrospectively from health care providers' perspective for a one-year time horizon between January 1st, 2018, and December 31st, 2018 for purposes of maintaining similar currency conversion, and inflation rates. Discounting for differential timing did not suffice in this study because the value of the resources considered for the analysis were categorized only as current costs.

Unit economic costs, currency conversion, and study assumption

The total (annual) economic costs were calculated for the defects (349 cases) for computing the unit economic costs as an average of the total costs expressed in Kenya Shillings (KES). The unit economic costs were calculated by dividing by the total annual costs by the number of cases using the following formula: -

$$\text{Unit economic costs(\$)} = \frac{\text{Total economic costs(KES)}}{\text{Total number of cases}}$$

Further, the unit economic costs were converted to United States Dollars (\$) at an existing exchange rate of KES 98.00 equivalent to \$1.00 in December 2018 using the following formula: -

$$\text{Unit economic costs(\$)} = \frac{\text{Total economic costs(KES)}}{\text{KES 98.00}}$$

This study assumed that the existing currency exchange rate of KES 98.00 in 2018 reflected the Purchasing Power Parity (PPP) globally. Thus, the unit economic costs for corrective and rehabilitative services for the defects were inflated to the U.S Dollar (\$) Consumer Price Index (CPI) from January 2018 to December 2018³³.

Data collection process

Before data collection began, the Principal Investigator (PI) recruited and trained four nursing graduates as research assistants (RAs) to ensure that the data abstraction process that spanned from August 1st, 2019 to September 30th, 2019 was carried out in a standardized manner. We adopted an ingredient approach to retrospectively gather the prevalence-based data on caseloads for the cost analysis of corrective and rehabilitative outpatient health services. The cost ingredients of bracing, tendonectomy, and casting interventions quantified consisted of caseloads (morbidity data) by type of MESBDs, the number of braces, the number of bracings, the number of bracing review visits, the number of casting materials, the number of castings, the number of casting review visits, the number of tendonectomies, and the number of tendonectomies review visits considered as direct recurrent costs; and staff emoluments, staff-time, building space for rental, and utility charges categorized as overhead recurrent costs. The prevalence-based morbidity data were retrospectively drawn from outpatient occupational therapist registers; described as medical records containing information on health services provided to children with major external structural birth defects. The information captured in these registers includes dates of clinic visits, outpatient numbers, names of the patients, patients' age, residence, diagnoses, and therapeutic interventions, among others. Following a predefined inclusion criterion, 349 cases were determined, and associated cost drivers identified, measured, and valued (quantified) using prevailing market prices and entered in a predetermined secondary data abstraction tool. On the other hand, staff-time for the hospitals' executives comprising the medical superintendents, chief nursing officers, orthopedic surgeons, and health administrative officers were gathered through face-to-face enquires from the occupational therapists being the closest proxies for the officers mentioned above. The ingredient technique and prevalence-based data were chosen for possibly of generating detailed and improved accuracy of the profiled costing data. The data gathered comprised the following: -

Caseloads/morbidity data: Following a predefined exclusion criterion, 349 cases of MESBDs were considered for the cost analyses.

Casting: Castings used to stabilize the affected feet consisted of Plaster of Paris (POP) bandages, orthopedic cotton bandages, and glycerine valued at local market prices. A set of these materials valued at \$3.9 were used to cast two cases of club foot. The study computed the number of castings and the number of revisits after the procedure for all cases of CTEV treated using these strategies.

Braces: Braces consisted of leather foot covers, rubber soles, and metallic rods used to stabilize cases of club foot. Braces were sourced from the local markets as ready-made products, therefore, were valued at \$15.31 using prevailing market prices. The study also enumerated the number of braces and the number of revisits after the procedure for all cases of CTEV treated using this strategy.

The number of tendonectomies: This was a procedure performed by surgeons to extend the Achilles tendon in club

feet. This is largely an outpatient specialized procedure, therefore, existing market hospital charges for outpatient specialized surgical procedures were used to value the cost of tendonectomies estimated at \$51.02. Similarly, the number of tendonectomies and the number of revisits after the procedure were computed for all the cases of CTEV that adopted this treatment strategy.

Emolument for personnel: The personnel comprised assistant occupational therapists and support staff whose emolument were estimated based on the respective schemes for staff with at least ten years of work experience^{34–36}. Emoluments for assistant occupational therapist consisted of basic salary, house allowance, commuter allowance, health risk allowance, and health extraneous allowance, whereas, for the support staff comprised basic salary, house allowance, and commuter allowance^{34–36}. The monthly salary and benefits for an assistant occupational therapist at “Grade 10” was valued at \$1,224.50, whereas support staff at “Grade 14” was valued at \$173.50.

Renting building space: Occupational therapy outpatient clinics were identified within the respective study hospitals whose plinth floor surface areas were measured in square feet and valued based on the existing local market rates for renting building spaces. The total renting space for the four hospitals was estimated at 3,593.63 square feet and valued at \$0.37 local market value.

Utility charges: Utilities included electricity; and water and sewerage estimated at \$25.51 and \$30.61 per month, respectively.

Supervisory staff-time: The staff-time for the medical superintends/directors, orthopedic surgeons, chief nursing officers/directors, and health administrative officers/directors were identified and measured by consensus through face-face inquiries made to the assistant occupational therapists being the closest proxies for the above-mentioned officers. The staff-time for medical directors and orthopedic surgeons was measured as a single specialized medical practitioner’s consultation valued at \$20.40 and quantified for five days a week for one calendar year. The staff-time for nursing directors and administrative directors on the other hand was measured as a single general medical practitioner’s consultation valued at \$10.20 for five days a week for one calendar year.

Ethical considerations

We obtained ethical approval from Kenyatta National Hospital (KNH)-University of Nairobi (UoN) Ethics Review Committee (Ref. No: KNH-ERC/A/44). Data collected were de-identified using anonymous codes and entered in a laptop secured by an alphanumeric coded key known only to the PI to maintain confidentiality.

Patient consent for publication: Patients were not directly involved because data was drawn from the medical registers, thus consent was not required.

Minimization of biases

Case ascertainment, information, and systemic biases were expected in this study; therefore, the PI began by predefining

an eligibility criterion (case definitions) for participation in the study and predetermining a secondary data abstraction tool for purposes of reducing case ascertainment biases. On the other hand, information biases were reduced by training the data collectors on secondary data extraction techniques from the outpatient occupational therapy clinics and entering data into the abstraction tools to ensure the process was conducted in a standardized manner. Further, all the registers for the entire one-year study period (2018) were reviewed and listed all the cases of external structural birth defects to reduce ascertainment and information biases in this study. Systemic bias was also reduced by excluding cases of delayed milestones, and/or developmental conditions due to management intervention similarities.

Data processing and statistical analysis

Following data collection, filled secondary data abstraction tools were manually checked daily for accuracy and completeness and subsequently entered in a Microsoft Excel spreadsheet (Microsoft Office Professional Plus 2019) by two independent data managers to reduce potential errors. The PI cross-checked and validated the computerized dataset against predetermined data abstraction tools for analyses. Descriptive qualitative categorical variables were summarized in frequency tables, proportions, and percentages to show their distributions, whereas continuous variables were summarised and presented in means (averages). Costs assigned to the direct cost center consisted of; (i) the names and numbers of specific MESBDs (caseloads/morbidity data), (ii) the number of primary and review visits for castings and/or bracings and/or tendonectomies (iii) the number of assistant occupational therapists and their emoluments (iv) the number of support staff and their emoluments, and (v) the floor plinth area for renting. Overheads on the other hand comprised; (i) the number of orthopaedic surgeons and associated staff-time (ii) the number of medical superintendents/directors, and associated staff-time (iii) the number of chief nursing officers/directors and associated staff-time (iv) the number of health administrative officers/directors and associated staff-time, and (v) utility charges for electricity, water and sewerage. All the costs were categorized as recurrent and allocated to the direct cost center whereas overhead costs were shared proportionally among the respective types of MESBDs and allocated to the final costs center for estimation of the economic costs. The unit economic costs were calculated as average costs expressed in U.S Dollars and inflated to the U.S Dollar Consumer Price Index from January 2018 to December 2018 for all types of MESBDs collectively, and individual types of MESBDs. Capital costs did not suffice in this cost study because movable, and fixed capital resources were not considered for valuation because no inventory records existed for furniture, examination couches even though they appeared to have lost more than half of their economic half-lives, and capital donations in kind, whereas motor vehicles, motorcycles, and bicycles were not used either as direct, indirect, or intermediate costs for corrective and rehabilitative health services for the under-fives with MESBDs. The occupational therapy clinics on the other hand, also as fixed capital costs, were exceedingly small portions of the respective hospitals’ floor plinths, hence valued as recurrent costs using prevailing local market prices for building space rental.

Inflation factor and statistical uncertainties: All the resources were categorized as recurrent costs and inflated to the U.S Dollar (\$) Consumer Price Index (Calculator) for a one-year time horizon from January 2018 to December 2018³³. This computation adjusted the unit economic costs to purchasing power parity as a factor of inflation, and minimized statistical uncertainties due to cost data scantiness and collection methods^{32,33}. Consumer Price Index measures the mean changes in market prices over some time in which consumers pay for goods and services such as health services for MESBDs, thus was preferred because of being the best optimal measure of inflation in this study³³.

Results

Distribution of cases by category

Of 349 cases 305 (87.39%) comprised congenital talipes equinovarus (CTEV) comprising unilateral congenital talipes equinovarus 300 (85.96%), bilateral congenital talipes equinovarus 3 (0.86%), unilateral congenital talipes equinovarus with germ valgus 1 (0.29%), and congenital talipes equinovarus with spina bifida 1 (0.29%). Additionally, the study observed 35 (10.03%) cases of congenital pes planus (CPP), and 9 (2.58%) cases of neural tube defects (NTD) consisting of hydrocephalus

5 (1.43%), spina bifida 3 (0.86%), and spina bifida with hydrocephalus 1 (0.29%) (Table 1).

Resource quantification for casting materials

Resource quantification for casting materials costing \$3.9 used for two procedures consisted of Plaster of Paris Bandage (7.6cm×2.7m×2pcs), orthopedic cotton bandage (15cm×3m×1pc), and glycerine oil (100 millilitres×1pc) costing \$1.84, \$1.22, and \$0.82, respectively. A set of casting materials valued at \$3.9 were used to cast two cases of club foot (Table 2).

Estimation of unadjusted economic costs

The annual economic costs were estimated and adjusted for inflation factors using the consumer price index calculator (CPI). Of the total unadjusted annual cost for all cases of the observed MESBDs (\$392,436.49), almost two-thirds (71.48%) of resource inputs were accounted for by emoluments of occupational therapists, whereas administrative staff-time accounted for about one-quarter (18%) (Table 3).

Of the total unadjusted economic costs, overhead (shared) costs for these defects consisted of staff-time, staff emoluments, and utilities were estimated at \$376,553.05 (Table 4). Of the

Table 1. Proportions of cases of major external structural birth defects.

Groups of MESBDs	Specific types of MESBDs	Frequency (%)
Musculoskeletal system defects	Unilateral congenital talipes equinovarus	300 (85.96)
	Bilateral congenital talipes equinovarus	3 (0.86)
	Congenital pes-planus	35 (10.03)
	Unilateral congenital talipes equinovarus with spina bifida	1 (0.29)
	Unilateral congenital talipes equinovarus with germ valgus	1 (0.29)
Central nervous system defects	Spina bifida	3 (0.86)
	Spina bifida with hydrocephalus	1 (0.29)
	Hydrocephalus	5 (1.43)
Total cases		349 (100.00%)

MESBDs, major external structural birth defects; %, percent

Table 2. Identification, measurement, and valuation of casting resource inputs.

Inputs for two castings	Item description	Quantity	Unit costs (\$)
Plaster of Paris Bandage	7.6cm ×2.7m×2pcs	1	1.84
Orthopaedic Cotton Bandage	15cm ×3m	1	1.22
Glycerine Oil	100mls	1	0.82
Sub-total			\$3.9

cm, centimetres; m, meters; mls, millilitres; pcs, pieces; \$, USD

Table 3. Quantification of annual inputs for 349 cases.

Resource inputs	Item description	Quantity	Unit costs (\$)	Total costs (\$)	Percent (%)
Outpatient bracing	Leather foot cover, rubber sole, and a metallic rod	50 procedures	@15.31 per procedure	765.50	0.20
Outpatient tendonectomies	Orthopaedic surgical procedure	14 procedures	@51.02 per procedure	714.28	0.18
Outpatient castings	Orthopaedic medical procedure	1089 procedures	@1.94 per procedure	2,112.66	0.54
First and review visits for castings	First and revisits	1089 visits	@10.2 per visit	11,107.80	2.83
First and revisits for all the defects	First and revisits	116 visits	@10.2 per visit	1,183.20	0.30
Estimated renting building space in square feet	898.44 (28.75×31.25) square feet @ \$0.37 per hospital per month at four hospitals	898.44 @ \$0.37 per month for 4 hospitals for 12 months	@ \$0.37× 898.44×4×12	15,956.29	4.07
Emoluments	19 occupational therapists at the four study hospitals per month	19 @ \$1,224.5 per month for 12 months	@\$1,224.5× 19×12	279,072.00	71.11
Emoluments	Four support staff at the four study hospitals	4 @ \$173.5 per month for 12 months	@\$173.5×4×12	8,328.00	2.12
Staff-time	Four medical superintendents at the four study hospitals	4 @ \$ 20.4 per day for 24 days for 12 months	@\$20.4×4× 24×12	23,501.00	5.99
Staff-time	Four chief nursing officers at the four study hospitals	4 @ \$10.2 per day for 24 days for day for 12 months	@\$10.2×4× 24×12	11,750.50	2.99
Staff-time	Four health administrative officers at the four study hospitals	4 @ \$10.2 per day for 24 days for day for 12 months	@\$10.2×4× 24×12	11,750.50	2.99
Staff-time	Four orthopaedic surgeons at the four study hospitals	4 @ \$20.4 per day for 24 days for day for 12 months	@\$20.4×4× 24×12	23,501.00	5.99
Utilities	Water and sewerage	Estimated @ \$ 25.51 per month for four hospitals	@\$25.51×4×12	1,224.48	0.31
Utilities	Electricity	Estimated @ \$ 30.61 per month for four hospitals	@\$30.61×4×12	1,469.28	0.37
Total (\$)				392,436.49	100.00%

@ at; \$, USD; %, percent

total overhead costs (\$376,553.05), emoluments for the occupational therapists accounted for three-thirds (74.11%), whereas staff-time costs accounted for 18.72% (Table 4).

Overheads were allocated proportionally (percentage-based) among the three specific cases: congenital talipes equinovarus (87.39%), congenital pes planus (10.03%), and spina bifida (2.58%) (Table 5).

Of the total unadjusted annual economic costs for major external structural birth defects; congenital talipes equinovarus was estimated at \$343,959.87, whereas congenital pes planus and neural tube defects were estimated at \$38,322.97 and \$10,153.67, respectively (Table 6).

Estimation of adjusted economic costs: The unadjusted unit economic costs on the other hand were estimated at \$1,124.46 for all the defects, \$1,127.74 for congenital talipes equinovarus, \$1,094.94 for congenital pes planus, and \$ for neural tube defects \$1,128.19 (Table 7). The annual and unit economic costs for all MESBDs observed were adjusted for the inflation factor to \$397,765.72. The annual economic costs for congenital talipes equinovarus were estimated at \$348,630.80,

whereas congenital pes planus and neural tube defects were estimated at \$38,843.39, and \$10,291.56, respectively (Table 7). Adjusted unit economic costs on the other hand were estimated at \$1,139.73 for all types of MESBDs, \$1,143.05 for CTEV, \$1,109.81 for CPP, and \$1,143.51 for NTD (Table 7).

The study showed relatively similar unit economic costs of the defects despite wide variations among the caseloads for specific types of MESBDs (Figure 1).

Discussion

To our knowledge, this was the first study to estimate the unit economic costs of MESBDs from health care providers' economic perspective among the under-five-year-old children in Kiambu County, Kenya. Substantial public health resources are continually allocated to the health care systems for care of children with MESBDs, however, the unit economic costs of care are barely known because they are rarely estimated mainly in the developing countries^{9,29}. Sufficient access and utilization of corrective and rehabilitative health services remain an important public health intervention for improving the quality of life for birth defect-affected children globally¹⁻⁴. Even though limited cost data, inadequate costing expertise, and the rarity of

Table 4. Distribution of overhead costs for specific birth defects.

Inputs	Item descriptions	Total annual costs (\$)	Percent (%)
Staff-time	Medical, orthopaedic surgeons nursing, and health administrative directors	70,503.00	18.72%
Renting building space	Estimated in square feet	15,956.29	4.24%
Emoluments	Occupational therapists	279,072.00	74.11%
Emoluments	Support staff	8,328.00	2.21%
Utilities	Water and sewerage	1,224.48	0.33%
Utilities	Electricity	1,469.28	0.39%
Total (\$)		376,553.05	100.00%

\$, USD; %, percent

Table 5. Proportional overhead costs allocated to the specific birth defects.

Specific MESBDs	Defects with/without co-defects	Cases (%)	Overheads (\$)
Congenital talipes equinovarus	Bilateral congenital talipes equinovarus, unilateral congenital talipes equinovarus, unilateral congenital talipes equinovarus with germ valgus, and unilateral congenital talipes equinovarus with spina bifida	305 (87.39%)	329,069.71
Congenital pes planus	Congenital pes planus	35 (10.03%)	37,768.27
Neural tube defects	Spina bifida, hydrocephalus, and spina bifida with hydrocephalus	9 (2.58%)	9,715.07
Total (\$)		349 (100.00%)	376,553.05

\$, USD; %, percent

Table 6. Resource inputs for the sub-groups of the defects.

Resource inputs for CTEV with co-defects (n=305)				
Resource inputs	Item description	Quantity	Unit cost (\$)	Annual costs (\$)
Castings	Unilateral CTEV	1028	1.94	1,994.32
	Bilateral CTEV	27	1.94	52.38
	Unilateral CTEV with germ valgus	21	1.94	40.74
	Unilateral CTEV with spina bifida	11	1.94	21.34
First and review visits	Unilateral CTEV castings	1028	10.20	10,485.60
	Bilateral CTEV castings	27	10.20	275.40
	Unilateral CTEV with germ valgus castings	21	10.20	214.20
	Unilateral CTEV with spina bifida castings	11	10.20	112.20
Bracings	Unilateral CTEV	50	15.31	765.50
Review visits	CTEV and co-defects bracings	21	10.20	214.20
Tendonectomies	Unilateral CTEV	14	51.02	714.28
Overheads	CTEV	305	1,078.92	329,069.71
Total (\$)				343,959.87
Resource inputs for congenital pes planus (n=35)				
Castings	Congenital pes planus	2	1.94	3.90
Visits for castings	Congenital pes planus	2	10.20	20.40
Review visits	Congenital pes planus	52	10.20	530.40
Overheads	Congenital pes planus	35	1,079.09	37,768.27
Total (\$)				38,322.97
Resource inputs for neural tube defects (n=9)				
First and review visits	Hydrocephalus	26	10.20	265.20
	Spina bifida	16	10.20	163.20
	Spina bifida with hydrocephalus	1	10.20	10.20
Overheads	Spina bifida with hydrocephalus	9	1,079.45	9,715.07
Total (\$)				10,153.67

CTEV, congenital talipes equinovarus; n, sub-total number of observations

Table 7. Unadjusted and adjusted economic costs (\$).

Caseloads (Morbidity data)		Unadjusted costs		Adjusted costs (inflated to CPI)	
Type of cases	Frequency (n)	Annual total economic costs (\$)	Unit economic costs (\$)	Annual total economic costs (\$)	Unit economic costs (\$)
All MESBDs	349	392,436.49	1,124.46	397,765.72	1,139.73
CTEV	305	343,959.87	1,127.74	348,630.80	1,143.05
CPP	5	38,322.97	1,094.94	38,843.39	1,109.81
NTD	9	10,153.67	1,128.19	10,291.56	1,143.51

MESBDs, major external structural birth defects; CTEV, congenital talipes equinovarus; CPP, pes planus; NTD, neural tube defects; CPI, consumer price index calculator

Figure 1. Bar graph for adjusted unit economic costs of major external structural birth defects.

defects have been attributed to the lack of knowledge on their costs, it is of public health and economic interest to estimate the opportunity costs of health care services for MESBDs^{9,29}. Worldwide, the results of this study could provide a baseline unit economic costs for the corrective and rehabilitative health services, inform efficient allocation of health resources, stimulate, and inform cost studies especially the costs' arms of full economic evaluation analyses^{17,22,31}.

The study encountered 349 cases consisting of 305 (87.39%) cases of CTEV, 35 (10.03%) cases of CPP, and nine (2.58%) cases of NTD. Congenital talipes equinovarus consisted of 300 (85.96%) cases of unilateral CTEV, three (0.86%) cases of bilateral CTEV, one (0.29%) case of unilateral CTEV with germ valgus, and one (0.29%) case of CTEV with spina bifida. Neural tube defects on the other hand comprised five (1.43%) cases of hydrocephalus, three (0.86%) cases of spina bifida, and one (0.29%) case of spina bifida with hydrocephalus. Despite variations in the number of cases (caseloads) observed for each of the defects mentioned above, this study showed a relatively similar unit economic cost for each defect in the county. The unit economic costs for NTD were approximated at \$1,143.51, whereas CTEV, and CPP were valued at \$1,143.05, and \$1,109.81, respectively. Notably, the unit economic cost of providing corrective and rehabilitative outpatient health services for these defects collectively was approximately estimated at \$1,139.73.

Despite defects of the central nervous system contributing the least number (9) of cases compared to congenital talipes equinovarus (305), and congenital pes planus (35), its unit economic costs was relatively equivalent to the costs of the latter two types of the MESBDs observed in the county (Figure 1). Although some forms of neural tube defects are potentially fatal, the children who survive beyond infancy require substantial economic resources to deal with the related adverse health impacts^{1,2,9,15}. The results of this study were indeed consistent with other research findings in the region and across the world that the greatest burden of disease associated with MESBDs is usually accounted for by the defects of the central nervous system^{11,15,37}. The economic burden of spina bifida is usually substantial throughout the life of the affected individuals ascribed to the experienced high medical care expenditures in the early years of life with the defect and later reduced milestone development usually associated with spina bifida^{11,38}. Our study similarly showed that neural tube defects followed by congenital pes planus accounted for the highest disease burden associated with MESBDs being shouldered by the health care systems in Kiambu county.

Even though our study estimated the economic costs of these defects among the under-five-year-old children, our findings mimicked results of other studies such as in Germany where similarly high staggering economic costs were encountered among the general population with various forms of NTD

between 2006 and 2009¹¹. Worldwide, spina bifida has singly been observed to account for the highest-burden of disease among other types of MESBDs^{11,15}. Significant economic costs have been reported among new-born children with NTD during their first years of life, whereas, high healthcare expenditures have been observed during childhood, adolescents, and adulthood compared to the children without NTD globally¹¹. In Germany, the average annual health expenditure of persons with spina bifida was estimated at €4,532, with inpatient health services contributing €1,358 (30.0%), outpatient health services €644 (14.2%), rehabilitation health services €29 (0.6%), drug therapy €562 (12.4%), and other remedies €1,939 (42.8%)¹¹. In the United States among children aged between 1–17 years old, medical expenditures on spina bifida were estimated to cost 13 times as much as those on children without spina bifida³⁷.

Notably, annual direct economic costs of different forms of major birth defects were estimated at \$2.6 billion in 2004 in the United States of America^{5,39}. Nonetheless, our study also endeavored to find and estimated the annual direct economic cost of MESBDs at \$397,765.72. The defects encountered at the study hospitals consisted of neural tube defects, congenital talipes equinovarus, and congenital pes planus. Despite different socioeconomic and demographic characteristics in Kenya and U.S, being developing, and developed countries respectively, this was indeed a remarkable empirical effort to estimate the direct economic costs of MESBDs in Kiambu County. Cost studies were pioneered in the United States of America by Dorothy Rice in 1967, and have since been undertaken widely in Europe and Australia unlike in middle-and low-income economies¹⁹. Low undertakings of cost studies, particularly in low-and middle-income economies have been attributed to the scarcity of data on the burden of these defects^{20,28,29}. Thus, the variations of annual direct economic costs could have been due to differences in the availability of the cost data, costing expertise, health services access, and utilization (economies of scale)^{5,17,19,39}. Despite variations observed in the estimates of the economic costs, these findings point to the continuous disease burden associated with MESBDs in the county underpinning efficiency in resource utilization, and allocation for MESBDs in public and faith-based health facilities.

The few cases of NTD observed in this study could be attributed to a proportion of the carers of children with NTD seeking alternative therapies due to the associated adverse psychosocial effects experienced by the affected families^{38,40–42}. Thus, the economic costs of NTD would be exponential compared to other forms of the defects observed in the study if all carers would have sought for care from the respective study hospitals. Nevertheless, the estimated costs demonstrated the potential catastrophic burden of the ‘silent’ economic problem in the region, thus underscores more scientific efforts to understand the magnitude of MESBDs regionally⁹. The observations made by this study have contradicted the epidemiological and economic fallacy that MESBDs are not of public health priority relative to other health events especially in resource-constrained countries^{1–4}. Nevertheless, some limitations were inherent in

this study; first and foremost, medical records used to draw the cost data were not designed for economic evaluation studies, whereas some of the defects were likely to delay childhood milestone development prolonging the demand for corrective and rehabilitative outpatient service possibly leading to more economic expenditures. The researchers also experienced difficulties in distinguishing the extent of the cost drivers for congenital talipes equinovarus occurring with spina bifida, congenital talipes equinovarus occurring with germ valgus, and spina bifida occurring with hydrocephalus, potentially due to inaccuracies of the profiled cost data.

Conclusions

This study estimated the economic costs of outpatient corrective and rehabilitative health services for MESBDs in Kiambu County in Kenya. Despite the fewest caseloads for the NTD, the study showed that NTD was associated with the highest burden of disease followed by CPP in the county. Despite CTEV proportionally contributing the highest caseload for the defects, it essentially accounted for the lowest burden of the disease associated with MESBDs in the county. This observation thus points to adverse developmental, and psychosocial impacts among the affected children and their families who are not able to access corrective and rehabilitative services. Similarly, these findings suggest a possible reduced economic productivity among the affected families arising from direct and indirect costs associated with major external structural birth defects. Therefore we would like to recommend further studies on the direct and indirect economic costs of MESBDs among children of school-going age to understand the impacts, and establishment of functional occupational therapy clinics in the ten sub-county hospitals to increase access of these services within Kiambu County.

Data availability

Harvard Dataverse: Costing analysis of outpatient services for major external structural birth defects: An ingredient approach in selected hospitals in Kiambu County, Kenya. <https://doi.org/10.7910/DVN/AULBDG>⁴³

Data are available under the terms of the [Creative Commons Zero “No rights reserved” data waiver](#) (CC0 1.0 Public domain dedication).

Acknowledgment

We would like to acknowledge National Commission for Science, Technology, and Innovation (Ref. No. NACOSTI/P/19/75586/28325), Kiambu County Commissioner (Ref. No. ED. 12 (A)/1/VOL.11/107), and Kiambu County Health Department (Ref. No. KIAMBU/HRDU/AUTHO/2019/03/06/AgotGN) for permitting us to carry out this study in the county. We would also like to acknowledge the Medical Superintendents/Directors of the four hospitals for granting us the permission and support during data collection. Lastly, we would like to acknowledge the efforts of all data collectors in making this exercise a reality.

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<http://www.doi.org/10.7910/DVN/AULBDG>

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<https://doi.org/10.5256/f1000research.55815.r88083>

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Vincent Okungu

¹ University of Nairobi, Nairobi, Kenya

² The END Fund, Nairobi, Kenya

The study design is confusing:

1. The design is not cross-section but rather retrospective ingredient-based approach using secondary data from purposively selected hospitals.
2. Authors need to itemize cost drivers; identify major cost drivers for each intervention.
3. What are cost centers?
4. The study is purely concerned with operational costs/recurrent. I fail to see any element of economic costing.
5. Authors need to clearly define economic costing, and how it fits with this type of study.
6. Why consider inflation factor when the time horizon is just one year? Doesn't make sense to me.

Is the work clearly and accurately presented and does it cite the current literature?

Yes

Is the study design appropriate and is the work technically sound?

Partly

Are sufficient details of methods and analysis provided to allow replication by others?

Partly

If applicable, is the statistical analysis and its interpretation appropriate?

Partly

Are all the source data underlying the results available to ensure full reproducibility?

Yes

Are the conclusions drawn adequately supported by the results?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Health economics & policy

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 17 Aug 2021

George Agot

1. We adopted an ingredient-based approach as a technique for gathering resources inputs for economic evaluation studies, both partial and full, however, cross-sectional design was specified to demonstrate the dimension of data collection. i.e., data was collected at a point in time similar to the retrospective dimension of data collection.

2. The interventions are clearly specified in this study as castings, bracings, and tendonectomies whose cost drivers are specifically stated as the number of the interventions, the number of first visits, the number of revisits among others including quantification of the resource inputs for casting, bracings and tendonectomy procedures. Staff emolument was identified as the major cost driver for these interventions thus will be included in the results section of the revised version of this article.

3. The cost centers are indeed not specified, however, they will be included in the revised version of this article to illustrate the resources assigned to the direct, intermediate, and indirect cost centers and their allocations to the final cost center.

4. This study is characteristically a cost analysis study in my opinion demonstrated by the methods used to determine the monetary value of the resource inputs, for example, the prevailing market prices and opportunity costs (staff-time) where applicable. Operational costs are usually established by financial cost analysis techniques to determine the actual expenditures on specific resource inputs for a service/good; thus not to determine the opportunity (economic) costs. Nevertheless, both financial and economic costs can be described as recurrent financial costs and recurrent economic costs respectively.

5. As explained in number four above, this study is purely an economic cost analysis based on the attributes stated above. However, I would like to point out that economic cost analysis and economic costing analysis have different meanings to some extent in my view. Economic cost analysis determines the opportunity costs (the best-forgone alternative choice) of the expenditures already incurred whereas, economic costing analysis endeavors

largely to determine the opportunity cost of expenditures likely to be incurred.

6. Discounting for differential timing is indeed not required in this study owing to the one-year time horizon as specified in the introduction section of this article. This explanation will be corrected in the methods and results section in the revised version of this article. However, computer price index (CPI) calculation was adopted particularly in this study as a strategy for uncertainty analysis attributed to the scantiness of the cost data and data collection methods.

Competing Interests: No competing interests

Author Response 20 Aug 2021

George Agot

To further clarify point number four, operational costs are essentially determined by accounting procedures as actual expenditures.

Competing Interests: No competing interests

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