RESEARCH ARTICLE

The economic impact of critical congenital heart disease to the health system and families in Colombia [version 1; peer review: 2 approved with reservations]

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Abstract

Background: Critical congenital heart disease (CCHD) make up a group of heart diseases present in newborns since the prenatal period and requiring early intervention through surgery or percutaneous interventions in the first year of life. Little is known about the societal economic impact associated with their care in low to middle income countries. We estimated direct medical costs, out-of-pocket expenditures and indirect costs of CCHD patients in Colombia.

Methods: The methodology to estimate costs involved four stages: identification, measurement, and assessment of resources consumed, and total cost calculation. Regarding medical costs, hospital and ambulatory costs were estimated for the patient's first year of life using clinical records of 73 patients and with thematic experts. A survey was carried out on 20 children's caregivers to determine the out-of-pocket expenses and indirect costs. For this estimation, a descriptive analysis was made on the survey taking into account the reported salary. All costs are expressed in US dollars (2017 exchange rates).

Results: The average direct medical hospital costs for CCHDs were \$25,835 and the ambulatory costs reached \$480. Indirect costs were \$1,303 and out-of-pocket expenses were \$2,058, which for families with an income lower than one monthly minimum wage (1 SMMLV) in

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2017 correspond to \$250. The impact on their budget was 57%.

Conclusions: CCHDs represent an important economic impact both for the Colombian General Social Health System and for families. This study made it possible to estimate the costs that are not easily visible and thus quantified.

Keywords

Economic impact, Cost of Illness, Cost Analysis, Hospital Costs, Out-of-pocket Expenditures, Indirect Expenditures

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Introduction

The term critical congenital heart disease (CCHD) refers to a group of structural malformations of the heart which are present from the prenatal period. They represent more than one third of all Congenital Heart Diseases (CHD)^{1,2}. Their incidence ranges from 1 in 15,000 to 1 in 26,000 live births and their prevalence corresponds to 147.4 cases per 100,000 live births³. CCHDs are an important cause of child mortality in the first month of life, and consequently it is necessary to start early surgical or interventional treatment in the first year of life to grant the patient's survival^{2,4}. 29.5% of newborns with CCHD are diagnosed late⁵, a fact associated with greater risk of morbidity and mortality⁶. The CCHD have a significant economic impact on the health systems and the patients' families; thereby, no studies on indirect costs are available, and few of them address direct medical costs at world level⁷.

The purpose of carrying out a costs study on a disease (COI "Cost of Illness"), is to perform a descriptive analysis to identify and measure the costs of a disease in monetary terms to estimate its economic burden on society^{8,9}. COI studies may take different perspective that determines the whole costs process; they estimate costs from the standpoint of society, the health system, the patient or the families. The perspective of society is the most comprehensive one and most widely recommended, since it includes all costs, independently from who incurs them. In consequence, besides direct medical costs, out-of-pocket expenses covered by the family, and indirect costs, are included. This society perspective allows to carry out a more complete analysis and include all costs of opportunity attributed to a disease. However, this perspective requires a larger collection and availability of information effort, a fact that hinders its use in COI for low prevalence diseases^{10,11}.

Overall, COI studies contemplate three cost categories: direct, indirect and intangible costs, the latter are rarely quantified, so it is accepted that only direct and indirect costs are considered. *Direct medical costs* are costs related with medical assistance, treatment, rehabilitation, etc.; *non direct medical costs* involve expenses in non-medical resources, i.e., transportation and nutrition outside the home. Lastly, *indirect costs* refers to the loss of productivity associated with the disease¹¹.

The micro-costing, and gross-costing approaches are used to estimate direct medical costs. The micro-costing is a "bottom-up" process whereby every resource is identified; measuring, assessment and calculation of the total cost are carried out; contrarily, the gross-costing estimates costs from a "top-down" approach, drawing an average of the total cost. The human capital approach and the friction cost approach estimate indirect costs. The loss of productivity is associated with the friction period, i.e., the time the patient takes to be replaced by another employee and for production to return to its former level¹².

From the economic standpoint, the needs of the population are endless but the resources to meet them are limited. Additionally, factors like technological development, cause health expenses to increase. The estimation of costs is important in making decisions, as it helps to define and prioritize policies and intervention

related with health assistance¹¹. Also, it provides information to make future costs projections in medical assistance and allocate resources according to budget limitations, thus granting the efficiency of policies^{8,11}.

In view of the importance of CCHD, the complexity presented in terms of costs for society and the scarce literature available on the subject, it was necessary for this study to estimate costs and hence approach the economic burden from a social perspective in Colombia. For this study we benefit from the information provided by Fundación Cardíofantail – Institute of Cardiology (FCI-IC) and its social program^{13,14}, to access a population that is difficult to study. Although many of the expenses incurred by the patients and their families are subsidized by FCI-IC's social program, the use of resources is real, which allowed to approximate to the real cost incurred by these CCHD patients in Colombia.

Methods

The method used to estimate the costs was micro-costing, as the information collected allowed to reach a level of detail in the identification and measuring of resources, and to carry out the assessment based on the national reference prices, and not only on those of a particular institution. Besides, this method identifies gaps in expenses and include only events related with CCHD. The calculations were made in Microsoft Excel 2016. We did not perform any statistical tests.

The seven CCHDs included in the estimation of costs were: Pulmonary Atresia, Tetralogy of Fallot, Tricuspid Atresia, Truncus Arteriosus, Hypoplastic Left Heart Syndrome, Total Anomalous Pulmonary Venous Drainage and Transposition of Great Arteries. The decision to include these CCHDs was made following the approaches and recommendations taken on the subject at world level, thus the prioritization of these cardiopathies in the United States was taken as reference¹⁵.

The cost estimation process followed four stages¹⁰: 1. Identification of resources; 2. Measuring the use of resources; 3. Assessment of resources; 4. Calculating total costs. This methodology was followed, and the "bottom-up" process, i.e., the estimation of direct medical hospital costs followed an identification of resources, measuring (frequency and quantity) and assessment of resources (as per Tariffs Manual ISS 2001+35%, market prices in Colombia)¹⁶, and lastly, the total costs calculation, based on clinical records for 73 patients with the seven CCHDs from the data base of the Institute of Congenital Heart Disease at FCI-IC. Direct annual medical costs were estimated for every CCHD, and for indirect costs and out-of-pocket expenses, 20 surveys were applied to caregivers of children with three Infrequent Congenital Heart Diseases since there is a higher level of complexity in the collection of information on CCHDs. The caregivers were a convenience sample identified from a cohort of the PINOCCHIO Program (Innovation Program in Uncommon Human Congenital Heart Diseases for Colombia). This cohort was built through three main sources at Fundación Cardíofantail – Instituto de Cardiología – FCI-IC: medical databases of patients from the Congenital Heart Disease Institute; Pediatric Cardiovascular surgery service; and patients

diagnosed during pediatric brigades carried out by FCI-IC social program “Give a life” in 12 cities in Colombia from the period 2010 to October, 2018. The eligibility criteria for the 20 caregivers of children were patients from the PINOCCHIO Cohort with detectable critical congenital heart diseases with the following diagnosis: Ebstein’s anomaly, Interrupted Aortic Arch and Pulmonary Valve Stenosis and which were less than two years of age at the time of the survey. The caregivers filled a case report form (CRF) whereby information on out-of-pocket expenses and indirect costs was obtained. Before caregivers started to answer the survey, a research assistant explained the objective, asked for signed informed consent, and were present to respond to any question. A copy of the CRF translated to English is attached as extended data.

The perspective taken in cost estimation was that of society. All costs are expressed in US dollars, firstly estimated in Colombian pesos for 2017, and then the average exchange rate for 2017 was applied (\$2,951.15 COP for one US dollar)¹⁷.

Estimation of direct medical costs

The collected information was filtered, and only the hospital events in which patients were admitted by causes directly related with CCHDs were selected. Then, a validation was made with experts on the amount of resources included in this data base to verify if they were similar to usual clinical practice. Then, an analysis of hospital events per patient was performed, including very detailed information on the resources used, grouped under large line items: surgical and non-surgical procedures, laboratories, diagnostic aids, blood bank, medication and surgical medical materials. Finally, a base case was drawn for a patient, including the average of hospital events (surgical and non-surgical) taking place in a year for every CCHD, weighing the frequency of these events and the average number of days in hospital for each.

The estimation of ambulatory costs was also carried out in four stages; the identification of resources for ambulatory events in the seven CCHD, was validated by clinical experts. To measure resources, we asked the same experts how many medical consultations, echocardiograms, EKGs and chest x-rays are performed on average on these patients in their first year of life. The assessment of resources was carried out based on Tariff Manual ISS 2001¹⁶; finally, the following formula was applied $Cost = frequency \text{ or } quantity * prices$.

The source for procedure prices was Tariff Manual ISS 2001¹⁶, a reference for tariffs used for contracting health services in Colombia. The adjustment of these tariffs for the Colombian market is done by taking the base tariff present in 2001 plus an increase of 35% (20% for the minimum cost and 50% for the maximum cost to perform a sensitivity analysis).

Estimation of out-of-pocket expenses and indirect costs

In order to estimate out-of-pocket expenses and indirect costs covered by the caregiver, a survey was applied to 20 caregivers of patients younger than two years with a CCHD, as detailed above. The survey included specific questions on monthly

income, occupation, days on leave from work and expenses associated with the disease to establish the impact of out-of-pocket expenses and the loss of productivity in the family budgets.

The methodology used to estimate the out-of-pocket expenses corresponds to a weighted average according to the records of very family. The line items included were: transportation costs (transfer to the city), payments caused by private medical consultation, payment of sliding scale fees¹⁸, co-payment expenses (contributions from patients), paperwork expenses, expenses caused by transfer to a temporary residence, on meals outside the home, purchase of medication and other as payment for care of other children, telephone calls, among others. A monthly average for out-of-pocket expenses was estimated considering the age of the patient, to then calculate the annual out-of-pocket expense undertaken by every family.

Regarding indirect costs, the number of days of leave was calculated for the caregivers who were economically active at the time of the survey and the equivalent cost of productive days lost was established by means of the human capital method, based on the net salary reported in the surveys. The average daily income was calculated based on the distribution of income of all caregivers, leading to a summation of the frequency in the level of expenses times the average daily income.

Out-of-pocket expenses and indirect costs were differentiated considering the need for a surgical procedure. Lastly, to understand the magnitude of these costs, the impact of these expenses on the annual budget of the families was calculated, differentiated by levels of income and measured in terms of the minimal wage in Colombia, which corresponded to \$250 monthly in 2017¹⁹.

Ethical approval

The ethical approval for the project was awarded by the Clinical Research Ethics Committee at FCI-IC (CEIC) for the supply and access to the database of patients with CCHD and for the collection of information on care providers. The informed consent was provided to be filled in by the CRF of patients from the PINOCCHIO Program Cohort, whereby information on out-of-pocket expenses and indirect costs of every care provided was obtained from those answering the survey.

Results

Direct medical costs

The average annual hospital cost for the seven CCHDs was \$25,835. Diagnosis of Truncus Arteriosus caused the highest cost, at \$42,419 and Anomalous Pulmonary Venous Drainage had the lowest cost, at \$16,904. Table 1 shows the results for the seven CCHDs, including inpatient costs with and without a surgical event, and the total cost (base cost), and the lowest and highest cost.

Table 2 shows the results of annual ambulatory costs. The highest ambulatory cost was found in the diagnosis for Hypoplastic Left Heart Syndrome, at \$719, and the lowest cost was

represented by the Tetralogy of Fallot, at \$357. The average ambulatory costs for the seven CCHDs was \$487.

Out-of-pocket expenses and indirect costs

The variables for the socio-demographic characteristics of the target population were collected by means of a survey applied to caregivers: 90% of patients with CCHD belonged to a socio-economic level equal or lower than 3 (in a scale ranging from 1 to 6, 1 being the lowest level²⁰); 55% were female, and the average age for patients was 11 months. Regarding the characteristics of caregivers, 40% had a secondary school level of education; 53% were employed, 42% reported having another occupation (housewife, unemployed); 75% stopped doing their most frequent activity to take up the patient's care and 54% had an income lower than one monthly minimum wage (SMMLV).

Indirect costs were estimated at \$1,303 per caregiver; the average number of days caregivers were on leave was 140 days. **Figure 1** shows the impact of indirect costs on the caregiver's budget depending on their level of income. The highest impact

is found among caregivers with incomes lower than one monthly minimum wage.

Besides, the differentiated indirect cost for patients undergoing surgery or not was estimated, where the difference between undergoing surgery and not is observed, reaching \$498. When the patient undergoing surgery the indirect cost was \$1,466 and in the other hand the cost was \$969.

The average out-of-pocket expense incurred by a family with a CCHD patient is \$171, approximately \$2,058 per year. The line item most frequently reported by the largest number of families was transportation costs in the city and meals outside the home (**Table 3**).

An estimate of out-of-pocket expenses was also made, differentiated for patients undergoing surgery where the difference was \$1,300. When the patient undergoing surgery, the out-of-pocket expenses was \$2,383.

To understand the economic impact imposed on families, it is important to estimate the impact on the family budget by income

Table 1. Inpatient costs for CCHDs (all in USD in 2017).

CCHD	Number of patients	Surgical event cost	Non-surgical event cost	Total cost	Lowest cost	Highest cost
Truncus Arteriosus	7	\$ 40,003	\$ 2,416	\$ 42,419	\$ 37,705	\$ 47,132
Transposition of Great Arteries	6	\$ 31,937	\$ 288	\$ 32,226	\$ 28,645	\$ 35,806
Tricuspid Atresia	9	\$ 23,879	\$ 1,286	\$ 25,164	\$ 22,368	\$ 27,960
Pulmonary Atresia	10	\$ 15,088	\$ 9,531	\$ 24,619	\$ 21,884	\$ 27,355
Hypoplastic Left Heart Syndrome	3	\$ 19,741	\$ 1,368	\$ 21,109	\$ 18,764	\$ 23,455
Tetralogy of Fallot	26	\$ 17,612	\$ 791	\$ 18,403	\$ 16,359	\$ 20,448
Total Anomalous Pulmonary Venous Drainage	12	\$ 16,266	\$ 638	\$ 16,904	\$ 15,026	\$ 18,783
Average CCHD	73	\$ 23,504	\$ 2,331	\$ 25,835	\$ 22,964	\$ 28,706

Table 2. Ambulatory costs for CCHDs.

CCHD	Base cost (USD 2017)	Lowest cost (USD 2017)	Highest cost (USD 2017)
Hypoplastic Left Heart Syndrome	\$ 719	\$ 639	\$ 799
Pulmonary Atresia	\$ 536	\$ 476	\$ 595
Tricuspid Atresia	\$ 536	\$ 476	\$ 595
Truncus Arteriosus	\$ 536	\$ 476	\$ 595
Transposition of Great Arteries	\$ 363	\$ 322	\$ 403
Anomalous Pulmonary Venous Drainage Total	\$ 363	\$ 322	\$ 403
Tetralogy of Fallot	\$ 357	\$ 317	\$ 397
Average for CCHDs	\$ 487	\$ 433	\$ 541

Figure 1. Impact of indirect costs on the caregiver's budget.

Table 3. Average monthly out-of-pocket expenses.

Out-of-pocket expenses	N	Frequency	Monthly average (USD 2017)	Weighted average (USD 2017)
Transportation costs (transportation in the city)	20	100%	\$ 46	\$ 46
Meals for caregiver outside the home	19	95%	\$ 34	\$ 32
Sliding scale fees	16	80%	\$ 6	\$ 5
Co-payment	15	75%	\$ 16	\$ 12
Purchase of medication	14	70%	\$ 14	\$ 10
Other expenses	10	50%	\$ 37	\$ 19
Paperwork expenses	8	40%	\$ 2	\$ 1
Payments caused by consultation to private doctors	7	35%	\$ 18	\$ 6
Expenses incurred by transfer to temporary accommodation	6	30%	\$ 134	\$ 40
Total				\$ 171

levels. For families earning less than one SMMLV (up to \$249) the out-of-pocket expense represented 57% of their budget, assuming an income of \$249, the widest interval range.

Discussion

The results of this study are consistent with a high economic impact of CCHDs on the health system and on families in Colombia. The consumption of resources that is made evident represents high average annual direct medical costs, both for inpatient and ambulatory services. Even out-of-pocket expenses undertaken by the families have a high impact on their budget, mainly in families with lower incomes.

The scarcity of literature on this subject makes this study relevant, not only in Colombia but also in other low and middle income countries. In addition, it is noteworthy to highlight the incorporation of estimates on direct costs and out-of-pocket

expenses, an economic burden for families which is rarely taken into account due to the difficulty in collecting this information. The estimation of costs in this study has been a comprehensive process, incorporating different sources of information to get an estimate of the data described, data bases, validation by experts, surveys to caregivers, complying with the cost estimation methodology involving the four stages previously described.

Studies on costs related with CCHDs mostly refer to the process of screening followed to detect these heart diseases, but not to the economic burden caused by these diseases as a whole. One study made by Peterson *et al.*¹⁵ approached this topic, although it focused on comparing inpatient events among CCHD patients diagnosed timely and late, and not on estimating the total financial burden assigned to the disease. However, inpatient costs for the first year of life were estimated for 2011 prices, which reached

\$100,200 in patients diagnosed late versus a cost of \$69,500 for patients timely diagnosed.

Regarding indirect costs and out-of-pocket expenses for CCHD, Connor *et al.*²¹ carried out a qualitative study that explored the perceptions of parents on the costs associated with having a child with congenital heart disease regarding direct medical and non-medical costs undertaken by the family, and how this economic burden resulted in more stress and emotional impact. Mughal *et al.*²² developed an observational study at a hospital in Lahore (Pakistan), and identified that 63.1% of the families contributed with the cost associated with the CHD patient's treatment, and 12.3% of them contributed at 100%. A study by Raj *et al.*²³, conducted in India, estimated costs associated with CHD. Although not focusing specifically on CCHD, it is the study that approaches this topic more closely. The main results indicated that the connection between total inpatient expenses and the family income was 0.93 times the annual family income; the average time lost due to leave of absence by the father was 35 days and the loss of working days was 15 days on average. A quantitative estimation on the indirect costs and out-of-pocket expenses associated with CCHDs was not found in the literature.

There were some limitations in this study, mainly related with the low number of cases available due to the low prevalence of most CCHDs, a fact that hindered the collection of more data which may have led to imprecision. This situation led to having to carry out the surveys within a period of nine months. In addition, since records did not provide enough information to identify ambulatory costs, the data had to be obtained by consensus with experts.

Conclusions

CCHDs represent a high economic impact on the health system and families, especially on those with lower incomes. Cost analysis is a relevant topic for the health system and it requires a systematic and complex process. In this case, different primary and secondary information sources were available: data bases, surveys to caregivers and validation with experts on the topic. In addition, the bottom-up approach as a way to estimate a more real cost can be highlighted.

One of the most relevant added values included in the analysis for this study was the social perspective, which includes the measuring of indirect costs and out-of-pocket expenses. This dimension led to a larger data collection effort, more so when dealing with low-prevalence diseases. The results deriving from this study show the impact of these costs on the budgets

of families when one of their members is diagnosed with a CCHD.

Data availability

Underlying data

In the following repository, you can find the dataset used for ambulatory costs, hospital costs and the survey data:

Open Science Framework: Underlying data of economic impact of CCHD in Colombia. DOI <http://dx.doi.org/10.17605/OSF.IO/9J8KT>²⁴

Extended data

The survey asked to 20 caregivers has been deposited in the following repository:

Open Science Framework: Survey asked to 20 caregivers of CCHD in Colombia. DOI <http://dx.doi.org/10.17605/OSF.IO/67S4G>²⁵

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

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The lack of Cost of Illness studies for Colombia makes this study particularly important. The societal perspective makes it even more valuable.

The paper has a good structure that covers briefly but clearly the basics on how costs are estimated. Although I would suggest having another read of the whole text and probably get it checked by an editor would help correct some usage errors that change the meaning of the text. For example, differentiated is a more mathematical term. Disaggregated could be a better word.

The first introductory paragraph mentions the prevalence CCHD with late diagnosis corresponds to a USA statistic which might be important to mention. It is likely that in a developing country could be different, probably higher.

The methods section could benefit from more clarity, particularly on the assumptions and decisions made. I am sure the study team had good reasons in making their decisions but is highly recommended that those reasons are clearly stated. For example, it is said that statistical tests were not performed without an explanation of why this was decided. Later in the text, seven CCHD conditions were selected based on a cost-effectiveness of screening for CCHD in the USA but is not stated why is this also considered a good selection for Colombia.

The time horizon used seems quite short, particularly because the criteria to select this period are not presented. Like other types of economic analyses in health, a clear description of the time horizon and the reasons for selecting a specific value should be included.

Hospital costs were estimated from observed data which is a valuable source. It is mentioned that observed use of services was validated by experts. There is no description of criteria used to categorise a person as an expert nor the methods used for the validation. Observed data may be considered data of higher quality relative to experts' opinion. It is not clear how discrepancies between both were solved. Moreover, the way the base case was constructed is poorly described. Moreover, a description of the contents and quantities of services included in the base cases is not

presented.

The sources of prices are a Tariff Manual used by an already extinct institution. It is not clear why the 2001 tariff is inflated for a specific value. This might be important because not all contracts among insurers and providers are negotiated using these prices. Furthermore, it is not clear that prices in this tariff manual are really reflecting the costs of production of health services.

Out of pocket expenditure is a big added value of the present study. The review of the survey used might have some problems, but I am not sure if this is because key questions got lost in translation. The interview is available in English. Probably the questions are well formulated in Spanish, although in English looks clunky. Despite this issue, I think is likely that monthly income might not be correctly estimated. Did the households income was estimated by adding the reported incomes? Routine household surveys could be used as an example of how income is estimated. It is not clear how the weighing of out-of-pocket expenses was made.

Regarding the results I would suggest presenting a weighted average for each condition instead of a simple average. I believe is important to describe what the non-surgical events include. Figure 1 describes the relation between reported income and indirect costs. Although the graph selected might not be the best option it presents interesting data. A chart or table with like this one relating out-of-pocket expenditure to the household income would be valuable.

It was mentioned in the introduction that some of the expenses were subsidised by the FCI foundation. That assertion makes necessary to present how much of the out-of-pocket expenditure was subsidised and to whom. The economic burden of these conditions seems to be extraordinarily high, probably sinking more into poverty families with children affected with CCHD. Is possible that subsidies received by the poorest families kept them in a better economic situation.

The discussion should reflect on some important aspects such as the short time horizon selected, the recall bias on the survey, the impact on the estimates of the differences in data sources or the lack of other statistical analyses. Also, what I consider the most valuable contribution of this study is the estimation of out-of-pocket expenditure, which the authors did not reflected upon in the discussion. This COI study finds an extremely high economic burden in the first year, which way above the 10% usually used as a threshold for driving families into poverty. A reflection on the financial protection that the Colombian Health System is providing would be interesting.

Finally, a reflection on how the producing estimates from a single source might affect the precision of the estimates. If the results presented here are to be used by decision-makers, what does an estimate from a single source that is a private provider in the capital city of the country might affect the estimates. Is it possible that costs in other cities might differ?

Unfortunately the methodological description is too shot and not entirely described. It is not possible to reproduce the results because the lack of descriptions in the methods.

My congratulations to the research team for a fantastic effort and looking forward to their discussion and considerations of the suggestions presented here.

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?

Yes

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?

No

Are the conclusions drawn adequately supported by the results?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Health economics, particularly in economic evaluation methods.

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.

Reviewer Report 11 February 2019

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The paper summarizes clinic-based medical cost estimates during the first year of life for 73 Colombian infants with one of seven specified CCHDs and also provides survey-based estimates of out-of-pocket medical and non-medical costs and productivity costs for 20 families.

The following text should be revised for clarity: "The human capital approach and the friction cost approach estimate indirect costs. The loss of productivity is associated with the friction period, i.e., the time the patient takes to be replaced by another employee and for production to return to its former level¹²." It should be made clear that those are two alternative approaches that can be used to estimate indirect or productivity costs and that the two methods typically yield very

different estimates. The second sentence refers specifically to the friction cost approach. Most (>90%) COI studies use the human capital approach, which typically yields substantially larger estimates of productivity costs since there is no time limit. Pike and Grosse (2018)¹ summarize the differences between those two approaches.

The study should not be described as following a micro-costing approach. A micro-costing or ingredients costing study assesses labor hours, consumables and supplies, depreciated equipment, floor space, utilities, et al. and the costs of each to calculate the cost of providing a service. Peterson et al. (2014)² used micro-costing to assess the costs of CCHD screening in a sample of hospitals in New Jersey, USA. The present paper used information on accounting costs, which is the approach typically used to estimate costs of health care services.

The authors are to be commended for seeking to undertake a COI study from the societal perspective, which includes the lost productivity of parents who take leave from employment to help their children obtain medical care. However, only a subset of societal costs have been estimated. First, the study did not include lost productivity associated with caring for a child with CCHD outside of medical encounters. Second, only considering time taken away from paid employment understates the loss of household services displaced by the need to provide care for a sick child. That approach is valid, but it is a partial estimate of the cost of informal care giving. Third, another limitation is the omission of the largest productivity cost using the human capital approach, namely the lost productivity associated with preventable deaths. Fourth, another limitation is that the analysis did not include estimates of costs for governmental programs such as public education. It is well-established that children with CCHDs are much more likely to require special education services. For example, see Oster et al. (2017)³.

The analysis of out-of-pocket costs likely double-counted medical costs by including both the charge and the amount paid by families, which is part of the overall charge. To avoid double-counting for the societal perspective COI analysis, medical expenses should be subtracted from the out-of-pocket costs reported. It is valid to include those costs in an analysis from the family perspective, in which case the clinic costs would not be included, thereby eliminating any potential double-counting.

References

1. Pike J, Grosse SD: Friction Cost Estimates of Productivity Costs in Cost-of-Illness Studies in Comparison with Human Capital Estimates: A Review. *Appl Health Econ Health Policy*. 2018; **16** (6): 765-778 [PubMed Abstract](#) | [Publisher Full Text](#)
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3. Oster ME, Watkins S, Hill KD, Knight JH, et al.: Academic Outcomes in Children With Congenital Heart Defects: A Population-Based Cohort Study. *Circ Cardiovasc Qual Outcomes*. **10** (2). [PubMed Abstract](#) | [Publisher Full Text](#)

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Are the conclusions drawn adequately supported by the results?

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Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Health economics, public health, newborn screening, birth defects

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.

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