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RESEARCH

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Do free caesarean section policies increase inequalities in Benin and Mali?

Marion Ravit^{1,4*} , Martine Audibert², Valéry Ridde^{1,3}, Myriam De Loenzien¹, Clémence Schantz¹ and Alexandre Dumont¹

Abstract

Background: Benin and Mali introduced user fee exemption policies focused on caesarean sections (C-sections) in 2005 and 2009, respectively. These policies had a positive impact on access to C-sections and facility based deliveries among all women, but the impact on socioeconomic inequality is still highly uncertain. The objective of this study was to observe whether there was an increase or a decrease in urban/rural and socioeconomic inequalities in access to C-sections and facility based deliveries after the free C-section policy was introduced.

Methods: We used data from three consecutive Demographic and Health Surveys (DHS): 2001, 2006 and 2011–2012 in Benin and 2001, 2006 and 2012–13 in Mali. We evaluated trends in inequality in terms of two outcomes: C-sections and facility based deliveries. Adjusted odds ratios were used to estimate whether the distributions of C-sections and facility based deliveries favoured the least advantaged categories (rural, non-educated and poorest women) or the most advantaged categories (urban, educated and richest women). Concentration curves were used to observe the degree of wealth-related inequality in access to C-sections and facility based deliveries.

Results: We analysed 47,302 childbirths (23,266 in Benin and 24,036 in Mali). In Benin, we found no significant difference in access to C-sections between urban and rural women or between educated and non-educated women. However, the richest women had greater access to C-sections than the poorest women. There was no significant change in these inequalities in terms of access to C-sections and facility based deliveries after introduction of the free C-section policy.

In Mali, we found a reduction in education-related inequalities in access to C-sections after implementation of the policy (p -value = 0.043). Inequalities between urban and rural areas had already decreased prior to implementation of the policy, but wealth-related inequalities were still present.

Conclusions: Urban/rural and socioeconomic inequalities in C-section access did not change substantially after the countries implemented free C-section policies. User fee exemption is not enough. We recommend switching to mechanisms that combine both a universal approach and targeted action for vulnerable populations to address this issue and ensure equal health care access for all individuals.

Keywords: Caesarean section, Low-income countries, Mali, Benin, Health policy, User fees, Health equity, Maternal health

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Background

The number of maternal deaths has decreased worldwide since 1990; however, 275,000 women still died giving birth in 2015, and almost half of these deaths occurred in sub-Saharan Africa [1]. A systematic review concluded that poorer women or women living in rural areas have less access to skilled delivery than richer women or those living in urban areas [2]. Inequalities also exist in terms of access to caesarean sections (C-sections), a major life-saving intervention needed by 3.6 to 6.5% of pregnant women [3]. Although C-section rates have increased in most low- and middle-income countries, these rates have typically increased faster for women in the richest quintile than for those in the poorest one [4]. A recent study of 2003–2013 showed that C-sections were extremely rare (less than 1%) among rural poorer women in seven out of 11 studied West and Central African countries, and only one country had a C-section rate higher than 2%. In contrast, eight of these countries had C-section rates higher than 4% among richer urban women [5].

In the 1980s, many African countries introduced user fees at the point of service to improve the quality of health services and access to primary health care. This change was supported by the Bamako Initiative (BI), which aimed to promote community financing of health services [6, 7]. User fees at the point of health service became a major barrier to health care access, especially for vulnerable populations [8]. This barrier is especially relevant for maternal health [9]. The fear of having to pay for excessive expenses may even lead some women to deter or delay their decision to seek care when they are dealing with obstetric complications [10]. Because C-sections are an expensive intervention, their access is directly influenced by household wealth and other non-financial factors [11], and the procedure can lead to catastrophic expenses [12, 13].

Some African countries have implemented user fee exemption policies for maternal health care services to improve access to maternal health services. In the 2000s, Benin and Mali, two western African countries with very high maternal mortality rates (respectively 405 and 587 per 100,000 live births), a low human development index ranking (167 and 175 out of 188,) and a high fertility rate (4.9 and 6.4 births per women) [14], decided to remove user fees only for women who receive a C-section.

In April 2009, the government of Benin introduced a national user fee exemption policy concerning all C-sections in selected public and private hospitals that offer emergency obstetric care. Hospitals receive 100,000 CFA (US\$166) per C-section, which covers pre-operative laboratory tests, medications, surgery kits, blood, hospitalization for 7 days and transportation to a hospital if the woman is referred [15, 16]. The state is the principal financier of the reform [16].

The government of Mali introduced a user fee removal reform on January 1, 2005; this reform concerns all C-sections in the public sector and covers the surgical procedure and pre-operative examinations, the surgical kit and postoperative treatment (a standardized set of products and medications), and hospitalization. Structures receive 30,000 FCFA (US\$50) for a simple C-section and 42,000 FCFA (US\$70) for a complicated C-section in addition to a surgical kit [17, 18].

In contrast to other financing mechanisms, such as conditional cash transfers or targeted vouchers, the fee exemption policy for C-sections in Mali and Benin concerns all women and does not target poorer, rural or non-educated women. This policy has had a positive impact on access to C-sections and facility based delivery (FBD) among favoured and unfavoured women [19]. The impact of user fee exemptions on socioeconomic inequality is still highly uncertain [20]. In Mali, a patient survey study conducted 5 years after implementation of the free C-section policy to estimate the distribution of C-sections across socioeconomic groups showed that wealthier women clearly had greater access to C-sections than poorer women [21]. Some studies showed that user fee removal was far from sufficient to ensure equity in access to maternal health care, and in some cases, it increased existing inequality in access [22–26].

Two recent studies used a robust approach via the difference-in-differences method to assess the effects of pregnancy-related fee removal on inequalities in access to maternal health services in Ghana, Senegal, Kenya and Burkina Faso [22, 27]. Only one of these studies evaluated the impact of the fee exemption policy on inequalities in access to C-section and found that the reform had its greatest impact on rural and less educated women [27]. However, this study did not determine whether inequalities were reduced after the policy was implemented in the studied countries compared to countries without a fee exemption policy.

The aim of this study was to observe whether there was an increase or a decrease in urban/rural and socioeconomic inequalities in access to C-sections and facility based deliveries after the free C-section policy was introduced in Mali and Benin.

Methods

We observed the evolution of inequalities in access to C-sections and facility based deliveries in the two countries through an observational study using repeated cross-sectional surveys.

Data available

We selected three Demographic and Health Surveys (DHSs) in each country covering a period over 15 years: (1) Benin: 2001, 2006 and 2011–2012 and (2) Mali: 2001, 2006 and 2012–13.

DHSs are funded by the U.S. Agency for International Development (USAID) and have been conducted approximately every 4 or 5 years in more than 90 countries (<https://dhsprogram.com>) since the beginning of the 1980s. These household surveys are nationally representative with large sample sizes (usually between 5000 and 30,000 households) that provide a wide range of information on, for example, child health, education, domestic violence, HIV prevalence and maternal health. These surveys are free and available on demand.

In DHSs, interviews were conducted with women aged 15 to 49 years old who spent the night before the interview in the surveyed household. Women were interviewed on their pregnancies in the last 5 years prior the survey. We selected information on the last birth for each of the surveyed women (if a woman delivered more than one child during this period, we selected the last birth only). Data on household characteristics (demographic, socioeconomic and environmental conditions) and on the last pregnancy, including information on the use of maternal health services, were extracted from relevant questionnaires. During the interview, the woman was asked whether her child was born by C-section. As recommended, for greater accuracy, C-section cases among births that occurred at home were recoded as vaginal birth deliveries [28]. Furthermore, on the DHSs, socioeconomic status was evaluated using principal component analysis [29, 30]. The wealth index is a composite measure of a household's cumulative living standard. The calculation is based on the household conditions and assets, such as televisions, telephones, vehicles, materials used for housing construction, and types of water access and sanitation facilities. The wealth index is calculated separately for each survey, which allows measuring the wealth of each household relative to others. For each woman, we used the household relative wealth index and wealth quintile available in each DHS survey.

Measures and exposure variables

We studied the trends in inequalities in women's access to C-sections and FBD in Benin and Mali. C-sections were the primary outcome because we assumed that policy implementation had a direct positive impact on women's access to C-sections. FBD was chosen as a secondary outcome to assess whether the policy helped improve access healthcare facilities. An FBD was defined as a birth that occurred in a public or private healthcare facility (hospital, health centre or clinic). Other births (taxi, car or home) were coded as non-FBD.

We studied the evolution of inequalities in terms of zone of residence, education and wealth. Zone of residence is a binary variable (urban or rural) that corresponds to the areas of residence of the woman during

the interview. Concerning education, we considered a woman as educated if she received at least a primary education. Finally, wealth was measured by the wealth index described below and the wealth quintile (from the poorest to the richest).

Other variables used in the analysis were the maternal age by category (<18 years, 18–34 years, and 35 years or more), parity (primipara or multipara) and the number of newborns (singleton pregnancy or multiple pregnancy).

We identified three study periods according to the policy introduction date in each country (Fig. 1). Study periods 1 and 2 corresponded to births before implementation of the free C-section policy, and study period 3 corresponded to the period afterwards. Period 1 included all births that occurred within 5 years prior to DHS 2001 in both countries. Since the policy was implemented in 2005 in Mali, period 2 included only the births that occurred from June 2001 to December 2004 prior to DHS 2006. The other births that occurred after December 2004 were included in period 3. For the same reason, and since the policy was implemented in 2009 in Benin, period 2 in Benin included births from DHS 2006 and DHS 2011–12 that occurred from December 2001 to April 2009. Period 3 in Benin included only births that occurred from April 2009 to March 2012 prior to DHS 2011–12.

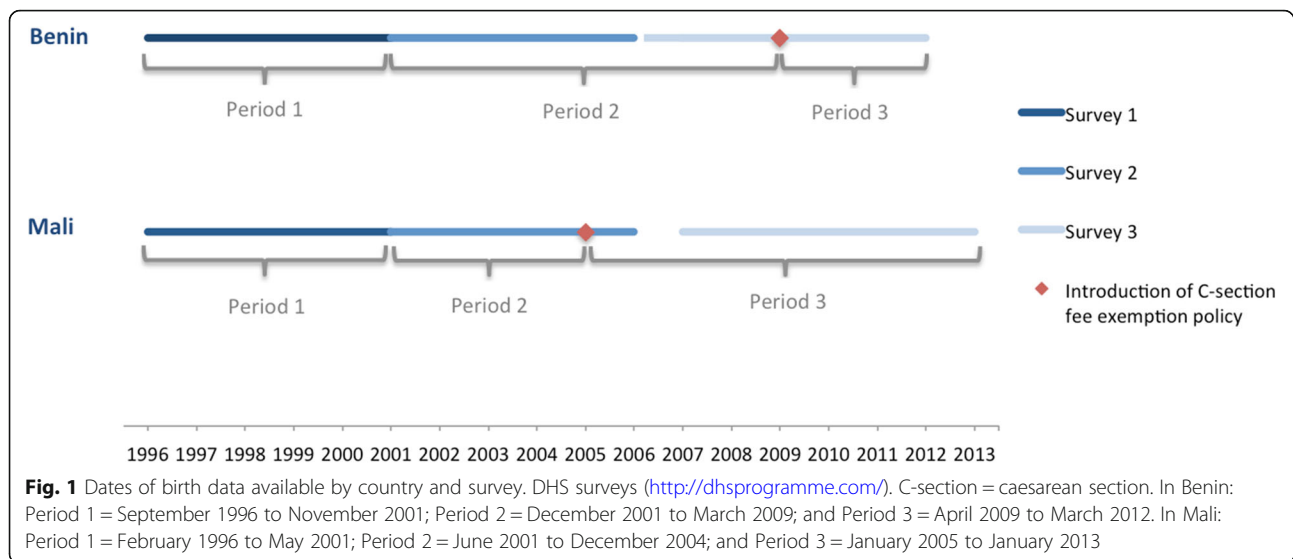
Statistical analysis

To study the trends in inequalities, we performed a multivariate logistic regression with a two-way interaction (time*categories) using the following form:

$$\text{Logit}[P(Y_{igt})] = \alpha + \delta \text{category}_i + \gamma \text{time}_i + \beta \text{group} * \text{category}_i + X_i$$

where Y is an indicator of whether woman i gave birth by C-section; category is a dummy variable indicating whether the woman belongs to the least or most advantaged category; time is a dummy variable indicating whether the birth occurred before (period 1 or 2) or after adoption of the policy (period 3); and X_{ict} is a vector of individual-level covariates.

In this case, β measures whether the change in inequalities regarding access to C-section rates between the most and the least advantaged categories is significantly different between periods (i.e., p -value of the interaction test is <0.05). Multivariate logistic regression models were adjusted based on maternal age, education, zone of residence, wealth quintile of household, parity, and multiple pregnancy, as well as sampling weight, clustering and strata. We analysed two outcomes (C-sections and FBD) and two types of inequalities (urban-rural and education-related inequalities) for each country. For each of the logistic regressions, we performed a Hosmer-Lemeshow



goodness-of-fit test, which allowed us to assess the model fit after fitting a logistic regression model taking survey design into account [31].

Adjusted odds ratios (aORs) associated with β from these regression were used to measure urban-rural and socioeconomic inequalities in access to C-sections. We first used an aOR, which is commonly applied in health and social sciences to measure inequalities [32, 33]. The OR here represents the odds of having a C-section (proportion of women who had a C-section divided by the proportion of women who did not have a C-section) in the least advantaged categories (rural, non-educated or poorest) divided by the odds of having a C-section in the most advantaged category (urban, educated or richest). An OR below 1 indicates inequalities in favour of the least advantaged categories (they are more likely to have C-section than the most advantaged categories), a value over 1 indicates inequalities in favour of the most advantaged categories (they are more likely to have a C-section than the least advantaged categories), and an OR equal to 1 indicates an equal distribution of C-sections among all women. Using logistic regression models, we calculated aORs on variables selected a priori as potentially affecting C-sections (area of residence, maternal age, education level, wealth quintile of household, parity, multiple pregnancy) and considered sampling weight, clustering and strata.

Third, to complete our analyses of socioeconomic inequalities, we used concentration curves (CCs) [34, 35] to present the degree of socioeconomic inequality in access to C-section. CCs plot the cumulative percentage of C-section rates (y-axis) and the cumulative percentage of women ranked by household wealth index (available in the DHS survey) in the order of poorest to richest

women (x-axis). If every woman, regardless of her wealth status, has the same access to C-section, the CC is a 45-degree line running from the bottom left-hand corner to the top right-hand corner, called the line of equity. If the CC is above the line of equality, it means that C-sections are more concentrated among the poor than among the rich. By contrast, if the CC is below the line of equity, the richest women have greater access to C-sections than the poorest women. The closer the CC is to the line of equity, the less important the inequalities in access to C-section are. We performed a CC by period for each country. We tested whether the concentration index (area under the curve) was significantly different between periods (Z-test).

No imputation of missing data was performed. Tests were two-tailed, and $p < 0.05$ was considered statistically significant. We managed the data with SPSS version 20 (SPSS Inc., Chicago, IL), and analyses were performed using Stata version 13.0 software (Stata Corp., College Station, TX, USA).

Results

We used the three DHSs for each country to analyse 47,302 women who delivered a live-born child in the 5 years prior to the interview (23,266 women in Benin and 24,036 women in Mali).

Figure 2 illustrates the evolution of C-section rates between study periods in Benin and Mali. In these two countries, regardless of the period, the C-section rates were higher for women in the most advantaged categories (urban, educated or richest) than for those in the other categories.

Table 1 presents the characteristics of women by period and country.

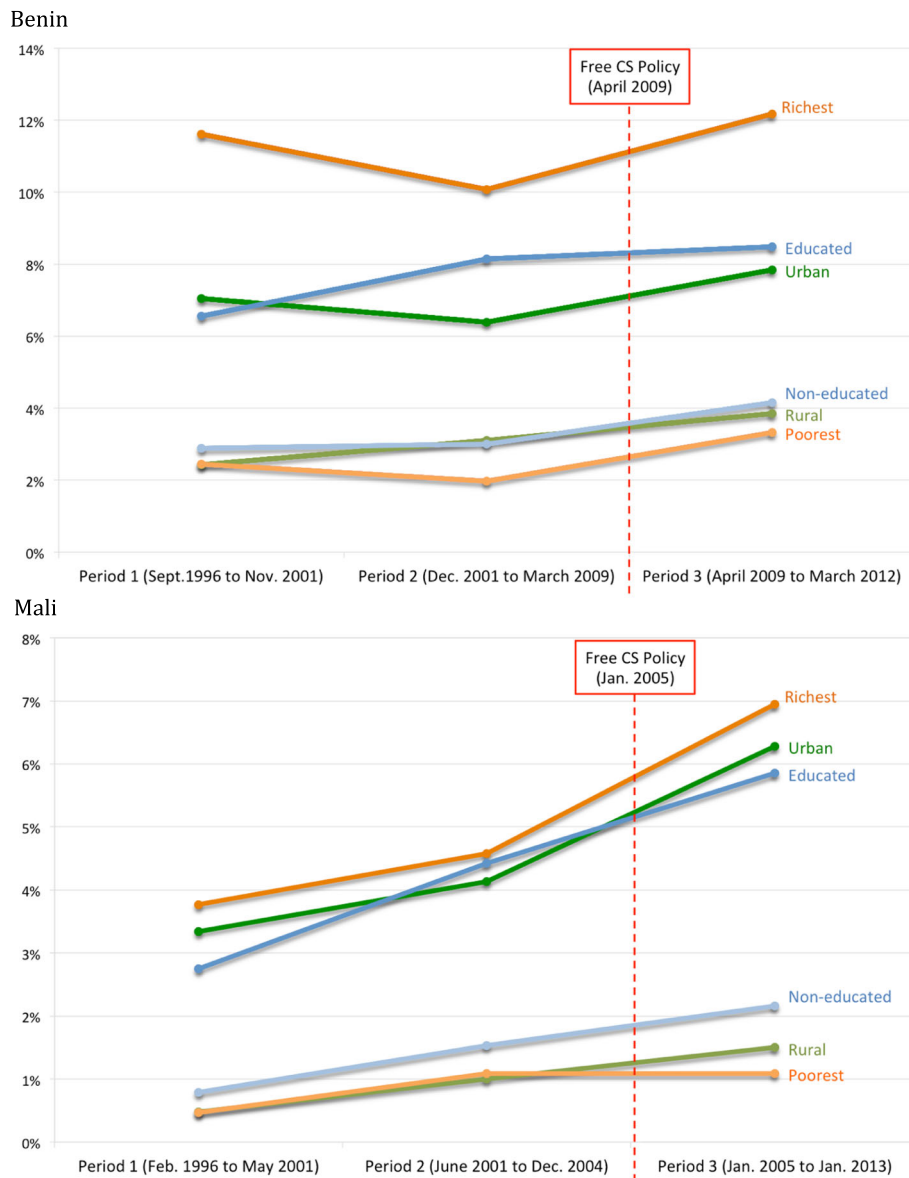


Fig. 2 Trends in C-section rates in Benin and Mali by category and period. DHS surveys (<http://dhsprogram.com/>). CS = caesarean section. In Benin: Period 1 = September 1996 to November 2001; Period 2 = December 2001 to March 2009; and Period 3 = April 2009 to March 2012. In Mali: Period 1 = February 1996 to May 2001; Period 2 = June 2001 to December 2004; and Period 3 = January 2005 to January 2013

The number of pregnant women differs greatly by period and country because the births from the study periods do not correspond to the births from the DHSs. Indeed, the policy introduction date is shifted in time: 2005 in Mali and 2009 in Benin. This difference in timing explains why there are more studied births in period 2 than in period 3 in Benin and why these results contrast with the results for Mali.

The mode of delivery was available for 99.8% of the included women (47,209 women), and the place of delivery was available for 99.8% of the included women (47,217 women).

Benin

Table 2 shows that in Benin, there is no significant inequality in access to C-section between urban and rural women or between educated and non-educated women, irrespective of the period. However, the results revealed significant inequality in access to FBD between educated and non-educated women in period 1 (adjusted OR = 1.58; 95% CI 1.21 to 2.05), and this inequality increased between periods 1 and 2 before implementation of the C-section fee exemption policy and between periods 1 and 3 (p -value = 0.001). There are significant poorest-richest inequalities in access to C-sections and FBD

Table 1 Characteristics of pregnant women^a by period in Benin and Mali

| | Benin | | | Mali | | |
|---------------------------------------|-------------------|-------------------|-------------------|-------------------|-------------------|-------------------|
| | Period 1 N (%) | Period 2 N (%) | Period 3 N (%) | Period 1 N (%) | Period 2 N (%) | Period 3 N (%) |
| Age categories | | | | | | |
| < 18 yr. | 48 (1.3) | 140 (1.1) | 105 (1.5) | 241 (2.9) | 57 (1.4) | 460 (4.0) |
| 18–34 yr. | 2634 (72.8) | 9297 (72.6) | 5359 (78.3) | 5914 (71.4) | 2799 (67.2) | 8696 (75.0) |
| 35 yr. and more | 934 (25.8) | 3372 (26.3) | 1377 (20.1) | 2125 (25.7) | 131 (31.5) | 2434 (21.0) |
| Parity | | | | | | |
| Primipara | 729 (20.1) | 2206 (17.2) | 1368 (20.0) | 1344 (16.2) | 655 (15.7) | 2029 (17.5) |
| Multipara | 2887 (79.8) | 10,603 (82.8) | 5473 (80.0) | 6936 (83.8) | 3511 (84.3) | 9561 (82.5) |
| Multiple pregnancy | | | | | | |
| Singleton | 351 (97.0) | 12,403 (96.8) | 6673 (97.5) | 8132 (98.2) | 4093 (98.3) | 11,386 (98.2) |
| Multiple | 106 (2.9) | 406 (3.2) | 168 (2.5) | 148 (1.8) | 73 (1.8) | 204 (1.8) |
| Education level | | | | | | |
| None | 2637 (72.9) | 9515 (74.3) | 4943 (72.3) | 6960 (84.1) | 3465 (83.2) | 9486 (81.9) |
| Primary or more | 979 (27.1) | 3294 (25.7) | 1898 (27.7) | 1320 (15.9) | 701 (16.8) | 2104 (18.2) |
| Zone of residence | | | | | | |
| Urban | 1138 (31.5) | 4744 (37.0) | 2577 (37.7) | 1801 (21.8) | 1357 (32.6) | 3216 (27.8) |
| Rural | 2478 (68.5) | 8065 (63.0) | 4264 (62.3) | 6479 (78.3) | 2809 (67.4) | 8374 (72.3) |
| Wealth quintiles of households | | | | | | |
| Q1 Poorest | 816 (22.6) | 2850 (22.2) | 1533 (22.4) | 1706 (20.6) | 731 (17.6) | 2211 (19.1) |
| Q2 Poorer | 782 (21.6) | 2623 (20.5) | 1473 (21.5) | 1641 (19.8) | 813 (19.5) | 2353 (20.3) |
| Q3 Middle | 752 (20.0) | 2675 (20.9) | 1448 (21.2) | 1854 (22.4) | 903 (21.7) | 2341 (20.2) |
| Q4 Richer | 697 (19.3) | 2584 (20.2) | 1326 (19.4) | 1720 (20.8) | 887 (21.3) | 2324 (20.1) |
| Q5 Richest | 569 (15.7) | 2077 (16.2) | 1061 (15.5) | 1359 (16.4) | 832 (20.0) | 2361 (20.4) |
| Place of delivery | | | | | | |
| Home or other | 814 (22.5) | 2432 (19.0) | 870 (12.7) | 5193 (62.7) | 2202 (52.9) | 5367 (46.3) |
| Public facility | 2347 (64.9) | 8676 (67.7) | 5210 (76.2) | 2979 (36.0) | 1890 (45.4) | 5941 (51.3) |
| Private facility | 447 (12.4) | 1689 (13.2) | 743 (10.9) | 76 (0.9) | 66 (1.6) | 275 (2.4) |
| Missing | 8 (0.2) | 12 (0.1) | 18 (0.3) | 32 (0.4) | 8 (0.2) | 7 (0.1) |
| Delivery by C-section | | | | | | |
| No | 3474 (96.1) | 12,208 (95.3) | 6471 (94.6) | 8156 (98.5) | 4079 (97.9) | 1126 (97.2) |
| Yes | 140 (3.9) | 552 (4.3) | 366 (5.4) | 91 (1.1) | 84 (2.0) | 328 (2.8) |
| Missing | 2 (0.1) | 49 (0.4) | 4 (0.1) | 33 (0.4) | 3 (0.1) | 2 (0.0) |

Source: DHS surveys (<http://dhsprogram.com/>)

C-section = caesarean section

In Benin: Period 1 = September 1996 to November 2001; Period 2 = December 2001 to March 2009; and Period 3 = April 2009 to March 2012

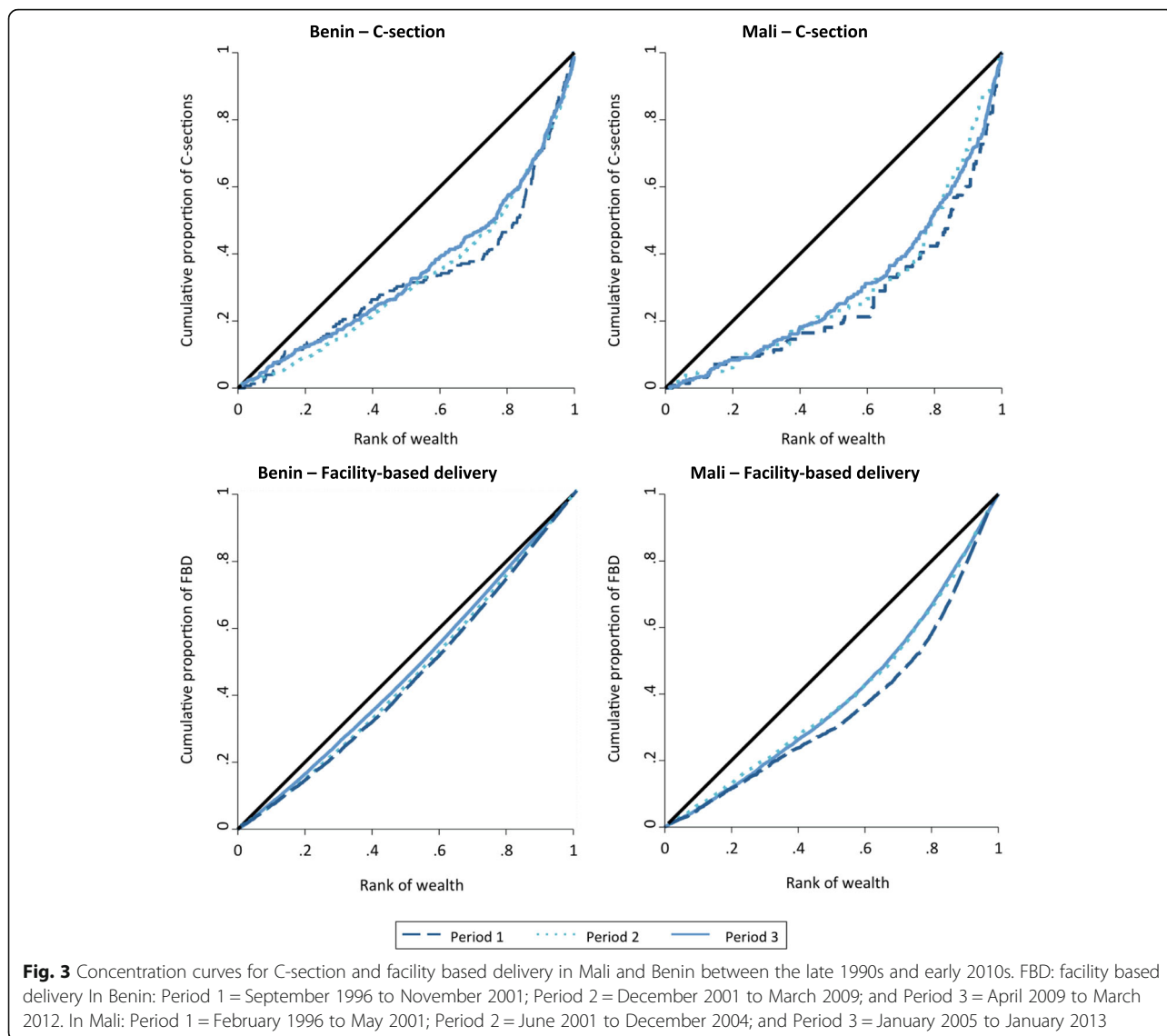
In Mali: Period 1 = February 1996 to May 2001; Period 2 = June 2001 to December 2004; and Period 3 = January 2005 to January 2013

^aWomen who delivered a live-born child within 5 years prior to the interview

but no significant change after the introduction of the policy.

Figure 3 shows that the CCs are below the line of equity irrespective of the outcome or period. Thus, our results confirm that the richest women have greater access to C-sections and FBD than the poorest women

do. The results indicated no significant change between periods in wealth-related inequalities in access to C-section or FBD. In particular, the concentration index for C-sections in period 1 was not significantly different from the concentration index in period 3 (p -value of the Z -test = 0.322). A significant reduction in



inequalities in FBD access was found between periods 1 and 3 (p -value = 0.000), but this decrease had already started prior to implementation of the policy.

Mali

In period 1, the probability of having a C-section was four times higher for urban women than for rural women in Mali (Table 3). Inequalities in favour of urban women were still significant in period 3 but were halved after implementation of the policy (p -value = 0.032). Notably, these disparities started to decline in the early 2000s, prior to implementation of the policy. Urban-rural inequalities in access to FBD were also significant, but no significant change was found between periods 1 and 3.

The risk of C-section delivery was higher among educated women than among non-educated women

(aOR > 1) irrespective of the period, but these inequalities decreased after implementation of the policy (p -value = 0.043). Inequalities linked to maternal education were also significant for FBD, but no significant change was found between periods 1 and 3 (p -value = 0.702, Table 2).

We observe inequalities in favour of the richest compared to the poorest in access to C-sections from period 2 and in access to FBD for all periods. No significant change was found between periods 1 and 3. Finally, confirmation of these results is shown in Fig. 3. In Benin, the richer women were, the better access they had to C-sections and FBDs, irrespective of the period.

Although no significant change in wealth-related inequalities related to C-section access was found between periods, we observed a significant decrease in

Table 2 Rate of delivery by C-section and in a facility for the most recent birth of each woman within 5 years prior to their interview in Benin

| | Before implementation of the policy | | After implementation of the policy Period 3 | Trends in inequalities between period 1 and period 3 <i>p</i> -value ^c |
|--|-------------------------------------|-------------------------|--|--|
| | Period 1 | Period 2 | | |
| Total | | | | |
| All pregnant women | 3614 | 12,760 | 6837 | |
| % delivery by C-section | 3.87 | 4.33 | 5.35 | |
| % FBD | 77.44 | 81.00 | 87.25 | |
| Women by zone of residence | | | | |
| Urban | 1137 | 4723 | 2574 | |
| % delivery by C-section | 7.04 | 6.39 | 7.85 | |
| % FBD | 85.44 | 86.97 | 92.02 | |
| Rural | 2477 | 8037 | 4263 | |
| % delivery by C-section | 2.42 | 3.11 | 3.85 | |
| % FBD | 73.78 | 77.48 | 84.37 | |
| Urban/rural inequalities | | | | |
| aOR (95% CI) for C-section ^a | 1.44 (0.88; 2.35) | 1.18 (0.96; 1.45) | 1.24 (0.92; 1.66) | 0.135 |
| aOR (95% CI) for FBD ^b | 0.75 (0.47; 1.21) | 0.85 (0.65; 1.12) | 0.97 (0.70; 1.35) | 0.244 |
| Women by level of education | | | | |
| Educated | 977 | 3280 | 1896 | |
| % delivery by C-section | 6.55 | 8.14 | 8.49 | |
| % FBD | 90.79 | 95.53 | 97.36 | |
| Non-educated | 2637 | 9480 | 4941 | |
| % delivery by C-section | 2.88 | 3.01 | 4.15 | |
| % FBD | 72.48 | 75.97 | 83.36 | |
| Educated/non-educated inequalities | | | | |
| aOR (95% CI) for C-section ^a | 1.02 (0.71; 1.48) | 1.87 (1.50; 2.33)*** | 1.21 (0.92; 1.59) | 0.665 |
| aOR (95% CI) for FBD ^b | 1.58 (1.21; 2.05)** | 3.31 (2.71; 4.06)*** | 3.13 (2.27; 4.33)*** | 0.001 |
| Women by wealth quintiles of households (Q1 and Q5) | | | | |
| Q1 - Poorest | 816 | 2842 | 1532 | |
| % delivery by C-section | 2.45 | 1.97 | 3.33 | |
| % FBD | 56.81 | 60.57 | 72.17 | |
| Q5 - Richest | 568 | 2073 | 1060 | |
| % delivery by C-section | 11.62 | 10.08 | 12.17 | |
| % FBD | 98.24 | 98.41 | 99.24 | |
| Poorest/richest inequalities | | | | |
| aOR (95% CI) for C-section ^a | 4.08 (1.96; 8.43)*** | 3.01 (1.96; 4.64)*** | 2.86 (1.69; 4.83)*** | 0.487 |
| aOR (95% CI) for FBD ^b | 58.63 (23.09; 148.89)*** | 35.02 (21.52; 57.01)*** | 37.57 (16.55; 85.28)*** | 0.525 |

aOR adjusted odds ratio; C-section caesarean section, and FBD facility based delivery

Period 1 = September 1996 to November 2001; Period 2 = December 2001 to March 2009; and Period 3 = April 2009 to March 2012

p* ≤ 0.05; *p* ≤ 0.01; and ****p* ≤ 0.001

^aThe aORs for C-section access were estimated with the use of multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, zone of residence, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

^bThe aORs for FBD access were estimated with the use of multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, zone of residence, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

^c*p*-values of the interaction between categories (urban vs rural or educated vs non-educated) and period (period 1 vs period 3) were estimated with the use of the multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

Table 3 Rate of delivery by C-section and in a facility for the most recent birth of each woman within 5 years prior to their interview in Mali

| | Before implementation of the policy | | After implementation of the policy | Trends in inequalities between period 1 and period 3 <i>p</i> -value ^c |
|--|-------------------------------------|-----------------------|------------------------------------|--|
| | Period 1 | Period 2 | Period 3 | |
| Total | | | | |
| All pregnant women | 8247 | 4163 | 11,588 | |
| % delivery by C-section | 1.10 | 2.02 | 2.83 | |
| % FBD | 37.04 | 47.04 | 53.66 | |
| Women by zone of residence | | | | |
| Urban | 1794 | 1355 | 3215 | |
| % delivery by C-section | 3.34 | 4.13 | 6.28 | |
| % FBD | 80.42 | 79.57 | 87.15 | |
| Rural | 6453 | 2808 | 8373 | |
| % delivery by C-section | 0.48 | 1.00 | 1.50 | |
| % FBD | 24.99 | 31.30 | 40.80 | |
| Urban/rural inequalities | | | | |
| aOR (95% CI) for C-section ^a | 4.68 (2.09; 10.46)*** | 1.96 (0.58; 6.61) | 2.05 (1.40; 3.00)*** | 0.032 |
| aOR (95% CI) for FBD ^b | 4.37 (2.93; 6.53)*** | 3.22 (2.14; 4.86)*** | 2.60 (1.91; 3.55)*** | 0.121 |
| Women by level of education | | | | |
| Educated | 1310 | 701 | 2103 | |
| % delivery by C-section | 2.75 | 4.42 | 5.85 | |
| % FBD | 64.74 | 80.54 | 81.53 | |
| Non-educated | 6937 | 3462 | 9485 | |
| % delivery by C-section | 0.79 | 1.53 | 2.16 | |
| % FBD | 31.80 | 40.27 | 47.49 | |
| Educated/non-educated inequalities | | | | |
| aOR (95% CI) for C-section ^a | 2.01 (1.23; 3.30)** | 2.02 (0.99; 4.14)*m | 1.39 (1.02; 1.89)* | 0.043 |
| aOR (95% CI) for FBD ^b | 2.18 (1.74; 2.72)*** | 3.68 (2.57; 5.29)*** | 2.06 (1.74; 2.43)*** | 0.702 |
| Women by wealth quintiles of households (Q1 and Q5) | | | | |
| Q1 - Poorest | 1702 | 731 | 2211 | |
| % delivery by C-section | 0.47 | 1.09 | 1.09 | |
| % FBD | 21.16 | 29.26 | 30.76 | |
| Q5 - Richest | 1353 | 830 | 2360 | |
| % delivery by C-section | 3.77 | 4.58 | 6.95 | |
| % FBD | 86.03 | 86.64 | 92.37 | |
| Poorest/Richest inequalities | | | | |
| aOR (95% CI) for C-section ^a | 0.91 (0.13; 6.27) | 13.86 (3.36; 7.30)*** | 2.45 (1.04; 5.78)* | 0.743 |
| aOR (95% CI) for FBD ^b | 5.54 (3.32; 9.26)*** | 2.21 (1.17; 4.16)* | 8.58 (5.42; 13.56)*** | 0.449 |

aOR adjusted odds ratio C-section caesarean section, and FBD facility based delivery

Period 1 = February 1996 to May 2001; Period 2 = June 2001 to December 2004; and Period 3 = January 2005 to January 2013

* $p \leq 0.05$; ** $p \leq 0.01$; *** $p \leq 0.001$; *m marginal level of significance ($0.05 < m < 0.10$)

^aThe aORs for C-section access were estimated with the use of the multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, zone of residence, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

^bThe aORs for FBD access were estimated with the use of the multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, zone of residence, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

^c*p*-values of the interaction between categories (urban vs rural or educated vs non-educated) and period (period 1 vs period 3) were estimated with the use of the multivariate logistic regression models (adjusted based on age, education, wealth quintile of household, parity, and multiple pregnancy, as well as sampling weight, clustering and strata)

wealth-related inequalities in access to FBD between periods 1 and 2 (p -value of the Z-test = 0.000).

For all the regressions performed in Tables 2 and 3, the F-adjusted mean residual goodness-of-fit test was applied, and the results suggested no evidence of lack of fit. No collinearity between the variables was detected.

Discussion

In both countries, we found no significant change in socioeconomic inequalities in access to C-section and FBD after the introduction of the free C-section policy, except for a reduction in education-related inequalities in access to C-section in Mali.

Otherwise, all significant changes that we observed began prior to the introduction of the policy.

Our results challenge ideas about free healthcare policies for all, which mainly benefit the most favoured social groups, such as the richest people or those who live in urban areas [36]. This study provides evidence that a user fee exemption policy does not necessarily lead to an increase in existing inequalities and can benefit the least advantaged population categories. Similar results have already been shown not only in Africa but also in Asia and Latin America [7, 37]. However, even though a free C-section policy does not increase inequalities, these inequalities are still present, and increased health care access is insufficient for the poorest, non-educated and rural women. Some prior studies have already reached the same conclusions [24, 38, 39]. Similar to other reports [40–42], we recommend switching to mechanisms that combine both a universal approach (health care for all) and targeted action for vulnerable populations to address this issue. The goal is to ensure equal health care access across individuals.

Our results on wealth-related inequalities are consistent with the results found by McKinnon et al. [22] concerning user fee removal policies focused on pregnant women (not specifically focused on C-sections) in other sub-Saharan African countries. They did not find robust evidence that this reform was associated with a reduction in wealth-related inequality in access to FBD. We found no difference in the evolution of wealth-related inequalities in access to C-section after implementation of the policy. The free policy benefitted the richest and poorest women in the same way. This conclusion is very similar to findings from another study, which concluded that the user fee policy in Burkina Faso benefitted all categories of women, including the poorest women [43].

We found that education-related inequalities related to maternal education in access to C-section decreased in Mali after implementation of the free policy. Education allows women to evaluate whether they require treatment [44]. Prior to implementation of the free policy (when C-sections were more expensive), we hypothesize

that educated women tended to pay for this intervention because they were aware of its benefits for themselves and their newborn infant when faced with obstetric complications. In contrast, non-educated women (often the poorest) could not afford to pay for an expensive intervention, regardless of its necessity. After implementation of the reform, even if non-educated women were not sensitive to the role of C-sections, they were more likely to follow their doctor's decision when the fear of having a financial burden disappeared.

Furthermore, user fee exemptions can contribute to improving the decision-making power of women in health matters [45]. This policy might thus encourage pregnant women to request a C-section.

In Benin, unlike in Mali, the results revealed no significant differences in access to C-sections between urban and rural women or between educated and non-educated women, before or after the implementation of the policy. Benin and its population of 11 million is approximately 10 times smaller in terms of surface area than Mali (population 18 million) (World Bank data). This difference implies that women have a better access to health care during pregnancy, even if they live in rural areas, as the distance to a health centre is less important.

Furthermore, according to the DHS 2011–12 in Benin, women who gave birth within the last 5 years prior to the interview were more urban (40% vs 19% in Mali) and more educated (28% received a primary education or more vs 16% in Mali) [46, 47]. The only related inequality was that the richest women had greater access to C-sections than the poorest women. Policies must therefore focus on measures that truly eliminate this inequality.

We did not find any studies that confirmed our findings on urban-rural access to C-sections in Africa. This result is an original finding showing that a user fee exemption at the point of service is not enough to reduce urban-rural disparities in access to C-section, even if rural women are often the poorest. Among the approximately 32,469 women living in rural areas who were included in our analysis, 27% belonged to the poorest quintile of wealth, while 49% of urban women were in the richest quintile.

Urban-rural inequalities in Mali decreased between period 1 (the end of the 1990s) and period 2 (the early 2000s). The Reference Evacuation System (RES) launched in 2002 in Mali can explain this decrease. The RES relies on improvements in communication, transportation, community cost-sharing, training and equipment in referral hospitals. A study showed that the RES had a positive impact on C-section rates among rural populations [48].

While the free C-section policy in Mali helped reduce education-related inequalities, some measures taken before the exemption had an effect on the

inequalities between urban and rural areas. Mali still needs to focus more on the poorest women to achieve wealth-related equity.

This study has limitations. First, we used DHS data to investigate access to C-sections and FBDs. The reliability of this kind of survey can be questioned as every variable is self-reported and thus potentially subject to possible misclassification and recall bias. Moreover, this kind of survey collects information only on live births, leading to the omission of all stillbirths in these analyses. Stillbirth rates are high in western African countries [49], and there is evidence that C-section rates and intrapartum stillbirth rates are correlated [50]. However, we could not study these cases with our data. Second, this study did not consider the level of implementation of the free C-section policy in both countries. Previous studies showed that the cost of C-sections was still high in many hospitals in Benin after the policy was introduced [15] and that 91% of women still paid for their C-sections in a rural area of Mali during the period 2008–2011 [51]. The impact of this policy on socioeconomic inequalities might have been more important than the observed impact if C-sections were truly free for every woman. Third, DHSs provide information only on whether a child was born by C-section, but we could not verify whether the procedure was required. A recent study showed that C-section rates in Mali and Benin are very high for low-risk women and for women with a previous C-section [52]. These findings suggest that some of these C-sections would not be medically justified. The World Health Organization (WHO) stated in 2015 that “C-sections should be undertaken when medically necessary” [53], and there is no evidence showing a benefit of C-sections for women or infants who do not require this procedure. Fourth, we have only 3 years of data in Benin after the implementation of the policy. This limitation might partly explain why we found no significant evolution of inequalities in Benin after introduction of the policy. The use of the next DHS is necessary to study the sustainability of these impacts.

Conclusion

This study provides evidence that urban/rural and socioeconomic inequalities in C-section access did not change substantially in Benin or Mali after the countries implemented free C-section policies.

To achieve universal health coverage (UHC) and allow every woman to have access to C-sections without suffering from financial issues, the removal of financial barriers such as user fees must be a priority.

However, to improve equity in access to C-sections, user fee exemptions should not be enacted alone, and a voluntary governmental policy must be established to target vulnerable women without leaving anyone behind.

Abbreviations

aOR: Adjusted odds ratio; BI: Bamako Initiative; CC: Concentration curve; C-sections: Caesarean sections; DHS: Demographic and Health Survey; FBD: Facility based delivery; RES: Reference Evacuation System; UHC: Universal health coverage; WHO: World Health Organization

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Availability of data and materials

The datasets generated and analysed during the current study are available on the DHS program website, <http://dhsprogramme.com/>.

Authors' contributions

MR and AD designed the study and prepared the first draft of the manuscript. MR performed the statistical analyses. All authors contributed to data interpretation and revisions to the manuscript and approved the final submission.

Ethics approval and consent to participate

Not applicable.

Competing interests

VR has served as a consultant on the issue of user fee abolition to non-governmental organizations in West Africa.

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References

- Kassebaum NJ, Barber RM, Bhutta ZA, Dandona L, Gething PW, Hay SI, et al. Global, regional, and national levels of maternal mortality, 1990–2015: a systematic analysis for the Global Burden of Disease Study 2015. *Lancet*. 2016;388:1775–812.
- Say L, Raine R. A systematic review of inequalities in the use of maternal health care in developing countries: examining the scale of the problem and the importance of context. *Bull World Health Organ*. 2007;85:812–9.
- Dumont A, De Bernis L, Bouvier-colle M-H, Bréart G. MOMA study group. Caesarean section rate for maternal indication in sub-Saharan Africa: a systematic review. *Lancet*. 2001;358:1328–33.
- Boatin AA, Schlottheuber A, Betran AP, Moller A-B, Barros AJD, Boerma T, et al. Within country inequalities in caesarean section rates: observational study of 72 low and middle income countries. *BMJ*. 2018;360 Available from: <http://www.bmj.com/content/360/bmj.k55.abstract>.
- Cavallaro FL, Cresswell JA, França GP, Victora CG, Barros AJ, Ronsmans C. Trends in caesarean delivery by country and wealth quintile: cross-sectional surveys in southern Asia and sub-Saharan Africa. *Bull World Health Organ*. 2013;91:914–922D.
- Ridde V. Is the Bamako initiative still relevant for west African health systems? *Int J Health Serv*. 2011;41:175–84.
- Lagarde M, Palmer N. The impact of user fees on access to health services in low-and middle-income countries. *Cochrane Libr*. 2011; <https://doi.org/10.1002/14651858.CD009094>.

8. Lagarde M, Palmer N. The impact of user fees on health service utilization in low- and middle-income countries: how strong is the evidence? *Bull World Health Organ.* 2008;86:839–48.
9. Koblinsky M, Moyer CA, Calvert C, Campbell J, Campbell OM, Feigl AB, et al. Quality maternity care for every woman, everywhere: a call to action. *Lancet.* 2016;388:2307–20.
10. Borghi J, Hanson K, Acquah CA, Ekanmian G, Filippi V, Ronsmans C, et al. Costs of near-miss obstetric complications for women and their families in Benin and Ghana. *Health Policy Plan.* 2003;18:383–90.
11. Thiede M, Koltermann K. Access to health services—analyzing non-financial barriers in Ghana, Rwanda, Bangladesh and Vietnam using household survey data: a review of the literature, vol. 61. New York: UNICEF; 2013.
12. Arsenault C, Fournier P, Philibert A, Sissoko K, Coulibaly A, Tourigny C, et al. Emergency obstetric care in Mali: catastrophic spending and its impoverishing effects on households. *Bull World Health Organ.* 2013;91:207–16.
13. Richard F, Witter S, De Brouwere V. Innovative approaches to reducing financial barriers to obstetric care in low-income countries. *Am J Public Health.* 2010;100:1845–52.
14. UNDP. Human development for everyone. New York: United Nations Development Programme; 2016.
15. Mongbo V, Godin I, Mahieu C, Ouendo E-M, Ouedraogo L. La césarienne dans le contexte de gratuité au Bénin. *Santé Publique.* 2016;28:399–407.
16. Witter S, Boukhalfa C, Cresswell JA, Daou Z, Filippi V, Ganaba R, et al. Cost and impact of policies to remove and reduce fees for obstetric care in Benin, Burkina Faso, Mali and Morocco. *Int J Equity Health.* 2016;15 [cited 2017 Mar 2]. Available from: <http://equityhealth.biomedcentral.com/articles/10.1186/s12939-016-0412-y>.
17. El-Khoury M, Gandaho T, Arur A, Keita B, Nichols L. Improving access to life saving maternal health services: the effects of removing user fees for caesareans in Mali. *Health Syst.* 2011;20:20.
18. Ministère de la Santé du Mali. Décret n° 05–350/P-RM du 04 août 2005 - Relatif à la prise en charge gratuite de la césarienne. [Internet]. 2005. Available from: http://mail.cnom.sante.gov.ml/index.php?option=com_content&task=view&id=406&Itemid=87.
19. Ravit M, Audibert M, Ridde V, de Loenzien M, Schantz C, Dumont A. Removing user fees to improve access to caesarean delivery: a quasi-experimental evaluation in western Africa. *BMJ Glob Health.* 2018;3:e000558.
20. Dzakupasu S, Powell-Jackson T, Campbell OMR. Impact of user fees on maternal health service utilization and related health outcomes: a systematic review. *Health Policy Plan.* 2014;29:137–50.
21. El-Khoury M, Hatt L, Gandaho T. User fee exemptions and equity in access to caesarean sections: an analysis of patient survey data in Mali. *Int J Equity Health.* 2012;11:49.
22. McKinnon B, Harper S, Kaufman JS. Who benefits from removing user fees for facility-based delivery services? Evidence on socioeconomic differences from Ghana, Senegal and Sierra Leone. *Soc Sci Med.* 2015;135:117–23.
23. Ganle JK, Parker M, Fitzpatrick R, Otuipiri E. Inequities in accessibility to and utilisation of maternal health services in Ghana after user-fee exemption: a descriptive study. *Int J Equity Health.* 2014;13:89.
24. De Allegri M, Ridde V, Louis VR, Sarker M, Tiendrebéogo J, Yé M, et al. Determinants of utilisation of maternal care services after the reduction of user fees: a case study from rural Burkina Faso. *Health Policy.* 2011;99:210–8.
25. Mladovsky P, Ba M. Removing user fees for health services: a multi-epistemological perspective on access inequities in Senegal. *Soc Sci Med.* 2017;188:91–9.
26. Dossou J-P, Cresswell JA, Makoutodé P, De Brouwere V, Witter S, Filippi V, et al. 'Rowing against the current': the policy process and effects of removing user fees for caesarean sections in Benin. *BMJ Glob Health.* 2018;3:e000537.
27. Leone T, Cetorelli V, Neal S, Matthews Z. Financial accessibility and user fee reforms for maternal healthcare in five sub-Saharan countries: a quasi-experimental analysis. *BMJ Open.* 2016;6:e009692.
28. Holtz SA, Stanton CK. Assessing the quality of cesarean birth data in the demographic and health surveys. *Stud Fam Plan.* 2007;38:47–54.
29. Rutstein SO, Johnson K. The DHS wealth index. DHS comparative reports no. 6. Calverton: ORC Macro; 2004.
30. Sahn DE, Stifel DC. Poverty comparisons over time and across countries in Africa. *World Dev.* 2000;28:2123–55.
31. Archer KJ, Lemeshow S. Goodness-of-fit test for a logistic regression model fitted using survey sample data. *Stata J.* 2006;6:97–105.
32. Spinakis A, Anastasiou G, Panousis V, Spiliopoulos K, Palaiologou S, Yfantopoulos J. Expert review and proposals for measurement of health inequalities in the European Union—full report. Luxemb: Eur Comm Dir Gen Health Consum; 2011.
33. Mackenbach JP, Kunst AE. Measuring the magnitude of socio-economic inequalities in health: an overview of available measures illustrated with two examples from Europe. *Soc Sci Med.* 1997;44:757–71.
34. O'Donnell OA, Wagstaff A. Analyzing health equity using household survey data: a guide to techniques and their implementation. Washington: World Bank Publications; 2008.
35. Kakwani N, Wagstaff A, Van Doorslaer E. Socioeconomic inequalities in health: measurement, computation, and statistical inference. *J Econom.* 1997;77:87–103.
36. Victora CG, Vaughan JP, Barros FC, Silva AC, Tomasi E. Explaining trends in inequities: evidence from Brazilian child health studies. *Lancet.* 2000;356:1093–8.
37. Li Z, Li M, Fink G, Bourne P, Bärnighausen T, Atun R. User-fee-removal improves equity of children's health care utilization and reduces families' financial burden: evidence from Jamaica. *J Glob Health.* 2017;7. <https://doi.org/10.7189/jogh.07.010502>.
38. Atchessi N, Ridde V, Zunzunegui M-V. User fees exemptions alone are not enough to increase indigent use of healthcare services. *Health Policy Plan.* 2016;31:674–81.
39. Ajayi AI, Akpan W. Who benefits from free institutional delivery? Evidence from a cross sectional survey of north central and southwestern Nigeria. *BMC Health Serv Res.* 2017;17:620.
40. Thomsen S, Hoa DTP, Målqvist M, Sanneving L, Saxena D, Tana S, et al. Promoting equity to achieve maternal and child health. *Reprod Health Matters.* 2011;19:176–82.
41. Marmot M, Bell R. Fair society, healthy lives. *Public Health.* 2012;126:S4–10.
42. Carey G, Crammond B, De Leeuw E. Towards health equity: a framework for the application of proportionate universalism. *Int J Equity Health.* 2015;14:81.
43. Ridde V, Kouanda S, Bado A, Bado N, Haddad S. Reducing the medical cost of deliveries in Burkina Faso is good for everyone, including the poor. *PLoS One.* 2012;7:e33082.
44. Ensor T, Cooper S. Overcoming barriers to health service access: influencing the demand side. *Health Policy Plan.* 2004;19:69–79.
45. Samb OM, Ridde V. The impact of free healthcare on women's capability: a qualitative study in rural Burkina Faso. *Soc Sci Med.* 2018;197:9–16.
46. EDSM V. Enquête démographique et de Santé au Mali 2012–2013. 2013.
47. Ministère du Développement de l'Analyse économique et de la Prospective Institut National de la Statistique et de l'Analyse économique - INSAE/Bénin, ICF International. République du Bénin Enquête Démographique et de Santé (EDSM-IV) 2011–2012 [Internet]. Calverton: INSAE/Bénin and ICF International; 2013. Available from: <http://dhsprogram.com/pubs/pdf/FR270/FR270.pdf>.
48. Fournier P, Dumont A, Tourigny C, Philibert A, Coulibaly A, Traore M. The free caesareans policy in low-income settings: an interrupted time series analysis in Mali (2003–2012). *PLoS One.* 2014;9:e105130.
49. Blencowe H, Cousens S, Jassir FB, Say L, Chou D, Mathers C, et al. National, regional, and worldwide estimates of stillbirth rates in 2015, with trends from 2000: a systematic analysis. *Lancet Glob Health.* 2016;4:e98–108.
50. Goldenberg RL, McClure EM, Bann CM. The relationship of intrapartum and antepartum stillbirth rates to measures of obstetric care in developed and developing countries. *Acta Obstet Gynecol Scand.* 2007;86:1303–9.
51. Ravit M, Philibert A, Tourigny C, Traore M, Coulibaly A, Dumont A, et al. The hidden costs of a free caesarean section policy in West Africa (Kayes region, Mali). *Matern Child Health J.* 2015;19:1734–43.
52. Schantz C, Ravit M, Traoré AB, Aboubakar M, Goyet S, de Loenzien M, et al. Why are caesarean section rates so high in facilities in Mali and Benin? *Sex Reprod Healthc.* 2018;16:10–4.
53. Betran AP, Torloni MR, Zhang JJ, Gülmezoglu AM. WHO Statement on caesarean section rates. *BJOG Int J Obstet Gynaecol.* 2015; [cited 2016 Nov 6]; Available from: <http://onlineibrary.wiley.com/doi/10.1111/1471-0528.13526/pdf>.