# Articles

# Obstetric fistula in Bangladesh: estimates from a national survey with clinical validation correction

Saifuddin Ahmed, Sian Louise Curtis, Kanta Jamil, Quamrun Nahar, Mizanur Rahman, SK Nazmul Huda, Imteaz Ibne Mannan, Shusmita Khan, Anadil Alam, Emily H Weaver, Shams El Arifeen

# Summary

Background Obstetric fistula, which develops after a prolonged or obstructed labour, is preventable and treatable. However, many women are still afflicted with the condition and remain untreated in low-income and middle-income countries. Concerns have also been raised that an increasing trend of caesarean sections is increasing the risk and share of iatrogenic obstetric fistula in these countries. The true prevalence of this condition is not known, which makes it difficult for health planners and policy makers to develop appropriate national health strategies to address the problem. The estimation of obstetric fistula with surveys is difficult because self-reporting of incontinence symptoms is subject to misclassification bias. In this study, we aimed to estimate the prevalence and burden of obstetric fistula in Bangladesh.

Methods For a valid estimation addressing misclassification bias, we implemented the study in two steps. First, we did the Maternal Morbidity Validation Study (MMVS) among a population of 65740 women in Sylhet, Bangladesh, to assess the sensitivity, specificity, positive predictive values (PPVs), and negative predictive values of the survey questions. This was done through confirmation of the diagnosis with clinical examinations of suspected cases by female physicians; a sample of women who screened positive for pelvic organ prolapse and other urinary incontinence symptoms were also examined and used as controls for clinical diagnosis confirmation. Second, we used the estimated diagnostic test values, after correcting for verification bias, to adjust the reported prevalence in the nationally representative Bangladesh Maternal Mortality and Health Care Survey 2016 for the unbiased estimation of obstetric fistula prevalence in Bangladesh.

Findings The MMVS, done from Aug 3 to Dec 9, 2016, identified 67 potential cases of obstetric fistula; of them, 57 (85%) women completed the clinical examination, and 19 were confirmed as obstetric fistula cases. The adjusted sensitivity of the self-reports of obstetric fistula was 100% (95% uncertainty interval [UI] 99·8–100) and the observed specificity was 99·9% (95% UI 99·9–100) among women aged 15–49 years. However, the PPV was low, at 31·6% (95% UI 19·2–46·2), suggesting that almost two thirds of the self-reported cases were not true obstetric fistula cases. We estimated an adjusted obstetric fistula prevalence rate of 38 (90% UI 25–58) per 100000 women aged 15–49 years in Bangladesh. Nationally, we estimated about 13 376 (90% UI 8686–20112) women of reproductive age living with obstetric fistula. Additionally, we estimated 4081 (1773–8790) women aged 50–64 years to be living with obstetric fistula in Bangladesh; overall, we estimated that there are 17 457 (10 459–28 902) women aged 15–64 years in Bangladesh with obstetric fistula.

Interpretation The burden of obstetric fistula is still high in Bangladesh. Prevention and provision of surgical treatment to so many women will need coordinated efforts, planning, allocation of resources, and training of surgeons.

Funding US Agency for International Development, Government of Bangladesh, and UKAid.

**Copyright** © 2022 The Author(s). Published by Elsevier Ltd. This is an Open Access article under the CC BY-NC-ND 4.0 license.

#### Introduction

Bangladesh achieved remarkable success in reducing the maternal mortality ratio by 4.7% per year between 1990 and 2015, compared with a 2.3% per year reduction globally.<sup>1</sup> However, for every 100 000 livebirths in the country, about 195 women still die each year from pregnancy-related complications.<sup>2</sup> For each maternal death, an additional 20 to 30 women are estimated to have acute or chronic maternal morbidities.<sup>3</sup> The information on the incidence or prevalence of maternal morbidities, however, is grossly scarce in low-income



Only in the past decade has interest emerged for the estimation of maternal morbidities in LMICs, recognising that many morbidities are also the causes or precursors of maternal mortality, and their prevention will not only reduce maternal mortality, but also improve the quality of life of women.<sup>45</sup> Among all maternal morbidities, obstetric fistula is the most devastating and debilitating,<sup>6</sup> and it is considered a public health problem.<sup>7</sup>





#### Lancet Glob Health 2022; 10: e1347–54

#### See Comment page e1228

Department of Population, Family and Reproductive Health, Bloomberg School of Public Health, Johns Hopkins University, Baltimore, MD, USA (Prof S Ahmed PhD); Data for Impact, University of North Carolina at Chapel Hill, Chapel Hill, NC, USA

(Prof S L Curtis PhD, M Rahman PhD, S Khan MSc, E H Weaver PhD); **IAP World** Services, Cape Canaveral, FL, USA (K Jamil PhD); **International** Centre for Diarrhoeal Disease Research, Bangladesh, Dhaka, Bangladesh (Q Nahar PhD, A Alam MBBS, S El Arifeen DrPH); EngenderHealth, Dhaka, Bangladesh (SK N Huda PhD\*); Save the Children International, Dhaka, Bangladesh (I I Mannan MBA\*) \*Former employee

#### Correspondence to: Prof Saifuddin Ahmed, Department of Population, Family and Reproductive Health, Bloomberg School of Public Health. Johns Honkins University.

Baltimore, MD 21205, USA

sahmed@jhu.edu

#### **Research in context**

#### Evidence before this study

We searched PubMed on Dec 03, 2021, for published studies in English using the terms "obstetric fistula" and ("validation" or "sensitivity" or "specificity" or "misclassification"). We also examined the population-based studies that reported prevalence. Several studies in the past few years suggest low validity of self-reporting of obstetric fistula symptoms. The only population study that was done in Bangladesh in 2003 estimated the number of women living with obstetric fistula as 169 per 100 000 ever married women. With substantial reduction of maternal mortality and increase in facility deliveries in recent surveys in Bangladesh, obstetric fistula was expected to have substantially declined in Bangladesh.

#### Added value of this study

To our knowledge, this is the first published study done to estimate the burden of obstetric fistula at national and subnational levels with clinical diagnosis validation. Our study applied a novel approach in combining the national-level survey data with diagnostic test results of the survey instruments from a validation study. Our study results suggest that self-reported obstetric fistula prevalence rates are about three times higher than the clinically confirmed true prevalence. These estimates of prevalence and burden of obstetric fistula can help Bangladesh to develop data driven strategies for addressing the problems of obstetric fistula prevention and treatment.

#### Implications of all the available evidence

Findings from our study suggest that obstetric fistula is still prevalent in Bangladesh: about 17 500 women aged 15–64 years are living with obstetric fistula. The UNFPA calls for eliminating fistula globally by 2030, and the Ministry of Health of Bangladesh aims to reach that goal. Our study results can serve as a baseline for tracking progress in achieving this goal. We also provide valuable insight about the problems of measuring obstetric fistula in LMIC settings and developed a novel method to address it. The study methods might be applied in other countries to validly estimate obstetric fistula prevalence.

Obstetric fistula develops after prolonged and obstructed labour. In this condition, an abnormal opening develops between the vagina and urinary bladder, rectum, or both, leading to continuous urinary or faecal incontinence. When progression of labour becomes obstructed, the soft tissues of a woman's bladder, vagina, and rectum are compressed between the foetal head and maternal pelvic bones because of the sustained contractions of the uterus. Consequently, the blood supply to the woman's genitourinary organs is progressively reduced, resulting in tissue necrosis. Almost 90% of such deliveries result in foetal deaths.8 In a few cases, vaginal fistula can occur from accidental injury during pelvic surgery (iatrogenic fistula), including caesarean section, or by violence (traumatic fistula). There are growing concerns that an increasing trend of caesarean section in LMICs is increasing the risk and proportion of iatrogenic fistula.9-11 Women who have obstetric fistula are highly stigmatised, and they often become social outcasts.12

Obstetric fistula is preventable and treatable by surgical repairs. This condition has been eliminated in highincome countries about a century ago through improved access to obstetric care, but still many women develop obstetric fistula in LMICs.<sup>7</sup> 1–3 million women are estimated to have obstetric fistula globally.<sup>713</sup> However, the true prevalence is not known.<sup>14,15</sup> The Demographic and Health Surveys (DHS) collected data on obstetric fistula in selected countries, especially in sub-Saharan Africa, but there were significant concerns about the validity of self-reporting.<sup>16</sup>

Several studies suggested over-estimation of obstetric fistula rates on the basis of self-reporting, which has

hindered its reliable estimation in the affected countries. A large study in Ethiopia found 35 women who reported fistula symptoms, but clinical examinations confirmed only 13 women as true obstetric fistula cases (37%).17 Another study among women who had recently given birth in Nepal found only two confirmed cases of fistula among 55 patients with symptoms.18 A study in two states of Nigeria reported a positive predictive value (PPV) of 47% for self-reporting of fistula-like symptoms among women who perceived fistula symptoms.19 In a study in rural India, 23.7% of women of reproductive age with a history of pregnancy reported obstetric fistula symptoms, but clinical diagnosis confirmed an obstetric fistula prevalence of 0.3%.<sup>20</sup> These study findings suggest that the obstetric fistula prevalence estimates from self-reported symptoms are overtly biased and not reliable.

The USAID-Bangladesh and the Bangladesh Maternal Mortality and Health Care Survey (BMMS)<sup>2</sup> Technical Working Group recognised the need to collect information on chronic maternal morbidities, particularly about obstetric fistula and pelvic organ prolapse (POP). The only population study that was done in 2003 estimated an obstetric fistula prevalence rate of 169 per 100000 evermarried women in Bangladesh.<sup>21</sup> The nationally representative survey BMMS 2016 aimed to estimate the current prevalence of obstetric fistula and POP in Bangladesh. To address the problem of self-reporting bias, we applied a novel approach in combining the nationallevel survey data with diagnostic test results of the survey instruments from a validation study to unbiasedly estimate obstetric fistula prevalence in Bangladesh at the national and subnational levels.

# Methods

# Study design

For a valid estimation of the prevalence of obstetric fistula, this study was implemented in two steps: first, a validation study—Maternal Morbidity Validation Study (MMVS) 2016—was done among a population of 65740 women, for estimating sensitivity, specificity, PPV, and negative predictive value (NPV) of self-reporting through confirmation of the diagnosis by clinical examinations with female physicians; second, we used the estimated diagnostic values, after correcting for verification bias, to adjust the reported prevalence in the BMMS 2016 for unbiased estimation of the obstetric fistula prevalence rate.

# **Description of the MMVS**

The MMVS was implemented in a random sample of unions in two *upazilas* (administrative regions) in Sylhet, Bangladesh. Any enumeration areas included in the BMMS 2016 sample in these two *upazilas* were excluded from the MMVS. The study was done in three phases: household census, community sensitisation, and clinical examination.

For the household census, all households in the selected unions were interviewed by use of a household questionnaire to identify members of the household. An individual morbidity screening questionnaire was then administered to all consenting ever-married women aged 13–64 years in the households. Women who had ever given birth were asked a series of questions on obstetric fistula, POP, and urinary incontinence symptoms. To identify suspected obstetric fistula cases, women were asked a series of questions on whether they had any problem in controlling urine or faeces. The questions were finalised following pre-testing in a pilot survey. Any women who responded positively to either or both of the questions "does your urine leak continuously, even when you are not urinating or trying to urinate?" and "do you currently experience faeces passing through the birth canal that you cannot stop, even when you are not defecating?" were considered suspected cases with obstetric fistula symptoms. The survey instrument is available publicly.<sup>2</sup>

For community sensitisation, all women who screened positive for obstetric fistula on the screening questions were visited at home by field workers, who provided them with information about obstetric fistula and gave them a referral card with a date and location for a clinical examination. A sample of women who screened positive for POP and other urinary incontinence symptoms on the screening questionnaire were also visited at home and invited for clinical examination. These women were used as controls for clinical diagnosis confirmation. For ethical reasons, women with no gynaecological symptoms were not invited for clinical examination that involved invasive clinical examination of pelvic areas. The referral cards were double blinded to hide the woman's response to the screening questions.



Figure: Flow diagram of Maternal Morbidity Validation Study participants POP=pelvic organ prolapse.

At the designated health facility, women who were selected for the clinical examination were examined by a qualified female physician to obtain a clinical diagnosis that could be linked to their responses to the screening questions for the validation of self-reporting. Clinically positive cases of obstetric fistula and POP were then referred to an appropriate clinical facility for treatment.

The flowchart of the sampling procedure and patient selections is shown in the figure. Selected sample characteristics of the women in the MMVS, including suspected and control cases, are shown in the appendix (p 1).

See Online for appendix

#### Data analysis

The analysis involved three steps: the estimation of sensitivity, specificity, PPV, and NPV by cross-tabulating the self-reported survey responses to the obstetric fistula screening questions and the clinical examination diagnosis; the adjustment of the estimate of sensitivity and specificity for verification bias<sup>22</sup> to account for the fact that not all women had a clinical examination; and the use of the adjusted estimates of the diagnostic test values to adjust population-based estimates of obstetric fistula prevalence rates in the nationally representative survey, BMMS 2016, which interviewed 314687 evermarried women aged 15–49 years. The details of the

survey design of the BMMS have been previously described.  $^{\rm 2}$ 

The obstetric fistula and POP module, which contains the same screening questions as the MMVS questionnaire, was administered to a subsample of 203 339 women who were randomly selected for the BMMS women's short questionnaire; 183 544 (90%) women who had a history of giving birth responded to the obstetric fistula-related questions. The reported prevalence of obstetric fistula in BMMS and MMVS among women aged 15–49 years were almost the same, and under an assumption that a similar finding is expected for women aged 50–64 years, we applied the MMVS estimates to extrapolate the national estimates for the older age group who were not interviewed in the BMMS to estimate the overall disease burden of obstetric fistula in the country among women aged 15–64 years.

#### Statistical analysis

Following the epidemiological method proposed by Rogan and Gladen,<sup>23</sup> we can use the estimates of sensitivity (Se) and specificity (Sp) of a diagnostic or screening test to correct a (self-reported) prevalence level ( $P_{obs}$ ) to obtain an unbiased estimate ( $P_{unb}$ )

$$P_{\rm unb} = \frac{P_{\rm obs} + Sp - 1}{Se + Sp - 1}$$

and variance

$$\operatorname{var}(P_{\text{unb}}) = P_{\text{obs}} \frac{1 - P_{\text{obs}}}{N(\text{Se} + \text{Sp} - 1)^2}$$

For ethical and logistical reasons, it is a common practice to do the confirmatory tests on individuals with positive symptoms. A major problem of this practice is verification bias22 that overestimates sensitivity and underestimates specificity, which consequently biases prevalence estimates. Another major problem of using sensitivity and specificity for correcting prevalence when the prevalence level is very low is that, if the observed prevalence rate is lower than the false positive rate (1-specificity) of the diagnostic test, the estimated unbiased prevalence rate based on the described formula becomes negative, which is unrealistic.24 However, it is possible to estimate an unbiased prevalence level from the PPV and NPV of the diagnostic test under the assumption of conditional independence of a missing confirmed diagnosis of unverified cases, which is often referred to as missing at random for the unobserved values of the test results. In this case,

$$P_{unb}=PPV*P(T=1) + (1-NPV)*P(T=0)$$

where P(T=1) is the probability of self-reporting positive, and P(T=0) is the probability of self-reporting negative, with *T* being test (self-reporting).

In this study, we have estimated the diagnostic indicators NPV and PPV using a logistic regression model specification for capturing uncertainty in estimates:

$$\log(\frac{P[D=1]}{1-P[D=1]}) = logit(\theta_i) = \beta_0 + \beta_1 T_i$$

where P(D=1) is the probability of (confirmed) disease diagnosis. The PPV and NPV are estimated by

$$PPV = \frac{exp(\beta_0 + \beta_1)}{1 + exp(\beta_0 + \beta_1)}$$
$$NPV = 1 - \frac{exp(\beta_0)}{1 + exp(\beta_0)}$$

Note that reversing the model specification with T as the outcome and disease diagnosis as the covariate leads to the PPV formula becoming the formula for sensitivity and the NPV formula becoming the formula for specificity.

Because the Bayesian method considers that the estimated parameters ( $\beta_0$ ,  $\beta_1$ ) are random, it is convenient to estimate uncertainty of the estimates from their posterior distributions. The Bayesian method is also shown to perform better for prevalence estimation when correcting for misclassification.<sup>25</sup> We have incorporated uncertainty in both self-reported point estimates of fistula symptoms and diagnostic tests in our Bayesian analytical method. The model specification details are provided in the appendix (p 2).

The diagnostic test indicators were estimated from the medians (50th percentile) of posterior distribution with a 95% uncertainty interval (UI) range. Considering a higher level of uncertainty in the estimates of prevalence rates of obstetric fistula, we present the 50th percentiles as the point estimates with 90% UIs (5th to 95th percentiles). The country-level number of obstetric fistula cases was estimated by multiplying the estimated prevalence rates with the UN estimated population size. We fitted the Bayesian models in R statistical software using the Stan<sup>26</sup> program.

#### Role of the funding source

The funders of the study had no role in study design, data collection, data analysis, data interpretation, or writing of the report.

#### Results

MMVS field work was done from Aug 3 to Dec 9, 2016, and BMMS 2016 was done over a period of 6 months from Aug 1, 2016, to Feb 27, 2017. The MMVS household census identified 51642 households, of which 48 816 households were successfully interviewed (94 · 5%). In these households, 65740 women between the ages

of 13 and 64 years were identified, and 61930 (94.2%) were interviewed. Of these, 56140 reported they had ever given birth and were asked the screening questions for obstetric fistula and POP. The analysis was limited to 56098 women aged 15-64 years. The screening identified 67 suspected obstetric fistula cases. Of them, 57 women (85.1%) completed the clinical examination with a female physician, and 19 were confirmed as obstetric fistula cases (14 cases among women aged 15-49 years and five among women aged 50-64 years). For the clinical diagnosis controls, 181 women who reported POP symptoms were randomly selected and invited for clinical examination, of whom 150 attended the examination and 149 completed it, and 244 women who reported other urinary incontinence symptoms (but not obstetric fistula or POP symptoms) were randomly selected and invited for clinical examination, of whom 200 attended the examination and 191 completed it.

The estimated sensitivity, specificity, PPV, and NPV from the MMVS data are shown in table 1. Among women of reproductive age (15–49 years), the observed sensitivity of the self-reports of obstetric fistula was 100% (95% UI 99·8–100) and the observed specificity was 89.7% (95% UI  $85\cdot8-92\cdot9$ ). The verification bias correction increased the specificity to 99.9% (99·9–100). However, PPV was low (31·6%, 95% UI 19·2–46·2), suggesting that almost two thirds of the self-reported cases were not true fistula cases. The NPV was 100% (95% UI approximately 100–100), suggesting that any non-suspected cases were highly unlikely to be a true obstetric fistula case.

We observed almost no difference in sensitivity and specificity estimates between women of reproductive age (15–49 years) and postmenopausal age (50–64 years). The PPV was slightly higher among older women than younger women (37.8% vs 31.6%), but this difference was not significant. The prevalence rates—both self-reported (115 per 100000 women) and adjusted with correction for reporting errors using the diagnostic PPVs and NPVs of the survey instrument (37 per 100000 women)—were lower among women of reproductive age than among women of postmenopausal age (table 1).

In the national BMMS 2016, the reported prevalence rate of obstetric fistula was 120 per 100 000 women (90% UI 108–134) among women aged 15–49 years (table 2). The adjusted prevalence rate—with correction for reporting errors—was 38 per 100 000 women (25–58).

34.8 million women aged 15–49 years with at least one child were estimated to live in Bangladesh in 2016 (based on the UN Population Prospects database). On the basis of the adjusted prevalence rate of obstetric fistula, we estimated that there were 13 376 (90% UI 8686–20112) women aged 15–49 years with obstetric fistula in Bangladesh at the time of the study. Although obstetric fistula occurs during the reproductive age, high prevalence of obstetric fistula among older women is also

	Age 15-49 years	Age 50-64 years	
Concitivity	(95% 01, 11=44 404)	(95% 01; 11=11 094)	
Obconved	100 (00 8 100)	100 (00 4 100)	
Observed Adjusted*	100 (99.8-100)	100 (99.4-100)	
Adjustea	100 (99-8-100)	100 (99-4-100)	
Specificity			
Observed	89.7 (85.8–92.9)	91.2 (84.2–95.9)	
Adjusted*	99.9 (99.9–100)	99·9 (99·9–100)	
PPV	31.6 (19.2–46.2)	37.8 (15.3–64.8)	
NPV	100 (approximately 100–100)	100 (approximately 100–100)	
Prevalence rate, per 2	100 000 women		
Self-Reported	115 (86–149)	134 (80–211)	
Adjusted†	37 (21–71)	51 (19–165)	
n=397 for the observed diagnostic tests. NPV=negative predictive value.			

PPV=positive predictive value. UI=uncertainty interval. \*Adjusted for verification bias. †Adjusted for reporting errors using diagnostic PPVs and NPVs.

*Table* 1: Sensitivity, specificity, PPVs, NPVs, and prevalence of obstetric fistula by age group, based on the Maternal Morbidity Validation Study 2016

Age			Total (90% UI)
	15–49 years (90% UI; n=183 544)	50-64 years (90% UI)	
Self-reported prevalence, per 100 000 women	120 (108–134)	NA	
Adjusted prevalence*, per 100 000 women	38 (25–58)	NA	
Female population with at least one birth†	34 840 027	8014556	42 854 583
Total estimated number of cases	13 376 (8686-20 112)	4081 (1773-8790)‡	17 457 (10 459–28 902)§
BMMS=Bangladesh Ma	aternal Mortality and Health Car	e Survey. NA=not available in	the BMMS 2016 that interviewed

women aged 15–49 years. UI=uncertainty interval. \*Adjusted for reporting errors using the diagnostic positive and negative predictive values. †UN data, estimated from the Revisions of World Population Prospects 2017. ‡Estimate based on Maternal Morbidity Validation Study prevalence data. §Summative total based on two age groups for estimating the total at a national level for women aged 15–64 years.

Table 2: Estimated prevalence and numbers of obstetric fistula cases in Bangladesh, based on BMMS 2016

expected because many women do not receive or have no access to surgical care after developing obstetric fistula. Because BMMS 2016 did not collect data among older women, we used the reported prevalence of obstetric fistula in MMVS among women aged 50–64 years to estimate the national burden of obstetric fistula. We estimated that an additionally 4081 (1773–8790) women aged 50–64 years have obstetric fistula in Bangladesh. Overall, we estimated that there are 17457 (10459–28902) women aged 15–64 years living with obstetric fistula in Bangladesh.

We assessed obstetric fistula prevalence rates from the BMMS 2016 data for selected socioeconomic and geographical variables (table 3). The adjusted prevalence rates of obstetric fistula for women aged 15–49 years were For the **UN Population Prospects database** see https://population.un.org/wpp/

	Self-reported prevalence rate of obstetric fistula, per 100 000 women (n=183 544)	Adjusted prevalence rate of obstetric fistula*, per 100 000 women (n=183 544)
Residence		
Urban	115 (92–141)	36 (23–54)
Rural	122 (106–138)	38 (25–55)
Division		
Barisal	202 (138–283)	63 (36–103)
Chittagong	91 (67–120)	29 (17-44)
Dhaka	206 (173–241)	65 (42-93)
Khulna	83 (55–120)	26 (15-43)
Rajshahi	120 (87–161)	38 (23–59)
Rangpur	44 (24–72)	14 (7–25)
Sylhet	78 (41–132)	24 (12-46)
Mymensingh	21 (7-48)	7 (2–16)
Women's Educatio	on	
No education	176 (143–211)	55 (35-81)
Primary	162 (136–191)	50 (33-74)
Secondary	73 (57–92)	23 (14–35)
Higher	25 (10–52)	8 (3–18)
Wealth quintile		
Lowest	125 (96–158)	39 (24–59)
Second	157 (124–194)	49 (31–72)
Middle	116 (89–148)	36 (25–55)
Fourth	107 (81–137)	33 (21–51)
Highest	94 (70–122)	29 (18–49)
Data are prevalence (9 Using the diagnostic p	00% uncertainty interval). *Adj ositive and negative predictive	usted for reporting errors values.

socioeconomic and geographical variables, based on Bangladesh Maternal Mortality and Health Care Survey 2016

similar in urban and rural areas (36 *vs* 38 per 100 000 women). Obstetric fistula was most prevalent among women with no education (self-reported rate 176 [90% UI 143–211] per 100 000 women; adjusted rate 55 [35–81] per 100 000 women), and possibly among poorer women (lower 40% in wealth quintile). Regarding geographical location, obstetric fistula prevalence was highest in the Barisal and Dhaka divisions.

# Discussion

To our knowledge, this is the first survey that has empirically estimated unbiased national obstetric fistula prevalence rates and the number of women living with obstetric fistula in a country. Our study addressed the problem of misclassification errors in self-reporting of obstetric fistula in surveys with clinical examinations of suspected obstetric fistula cases by trained female physicians in a validation sample. We then used the diagnostic test validity results to correct the overreporting errors of fistula symptoms in the nationally representative sample of women to unbiasedly estimate obstetric fistula prevalence rates in Bangladesh. The 100% observed sensitivity of the self-reports of obstetric fistula suggests that true obstetric fistula cases were correctly identified by the survey questions in the field, and the very high specificity (99.9%) suggests that true non-cases will be extremely unlikely to be characterised falsely as an obstetric fistula case (ie, false positive).

We recognise some limitations of the study. First, the number of suspected cases for the validation study was low. Although the MMVS was implemented with a large sample size of 65740 women, only 67 women were identified as suspected obstetric fistula cases, which has implications for the precision of estimates. A rule of thumb for the sample size calculation is to keep the relative margin of error within 50% when the prevalence rate is expected to be low;<sup>27,28</sup> our post-hoc analysis estimated this relative margin of error to be 23.9%within the recommended limit. However, low numbers of the suspected and confirmed cases are likely to increase uncertainty estimates of the diagnostic statistics. Second, the MMVS was implemented in two areas in one division, but the diagnostic results were applied to all eight divisions of the country for adjusting self-reporting errors at the national level. Logistically, it was not feasible to do clinical examinations on all suspected cases in the national survey with a trained experienced gynaecologist. Our comparison between the MMVS and BMMS shows that the prevalence of self-reported obstetric fistula symptoms was similar in the two surveys; we assumed that similar results would also be expected for the clinical diagnosis at the national level for conducting the analysis. Third, the BMMS was limited to women of reproductive age (15-49 years). We considered that many older women remained untreated, and they should be considered for estimating the burden of obstetric fistula at a national level. As mentioned earlier, the reported prevalence rate of obstetric fistula in BMMS and MMVS among women aged 15-49 years was similar and, under an assumption of a similar finding expected for the older women, we applied the MMVS estimates to extrapolate the national estimates for older women aged 50-64 years. We presented the stratified results so that the estimates for older women can be interpreted cautiously. Because no data were collected for women older than 65 years in the study, these study results are limited to ages 15-64 years, which might slightly underestimate the true prevalence among all women. Fourth, for ethical reasons, the MMVS excluded healthy non-symptomatic negative cases from gynaecological clinical examinations. The exclusion of healthy individuals from the control group is likely to reduce the specificity of the diagnostic test.<sup>29</sup> Our adjustment for unbiased estimation of the prevalence rate was not based on the specificity rate, recognising that when the reported prevalence is less than 1-specificity-which is especially the case with very low prevalence diseases-the adjusted rate becomes negative.<sup>24</sup> Therefore, the exclusion of healthy

cases from clinical examinations is unlikely to affect our prevalence estimations.

The Demographic and Health Surveys (DHS) began to collect data on incontinence symptoms from 2005 onwards in selected countries with high risk of obstetric fistula. However, the DHS survey questionnaire on fistula was never validated for the true diagnosis of obstetric fistula. The reported incontinence rates ranged from a high of about 5% to a low of less than 1%. Substantial concerns have been raised that the tool had low sensitivity and specificity and was thus likely to grossly over-report the prevalence of obstetric fistula. Maheu-Giroux and colleagues re-estimated the prevalence of obstetric fistula in several sub-Saharan African countries after correcting for misclassification in self-reported symptoms in DHS, using priors of a set of ranges in sensitivity (95-100%) and specificity (0-99.95%) values through a Bayesian analytical method.<sup>16</sup> Our study results provide an empirical basis of sensitivity, specificity, PPVs, and NPVs that might be used for correcting self-reported obstetric fistula symptoms. We also discussed the problems of using sensitivity and specificity for correcting prevalence values when the prevalence is extremely low. Our PPV estimates among women aged 15-49 years are similar to those found in Ethiopia (37.1%)<sup>17</sup> but lower than those found in Nigeria (47%)<sup>19</sup> and much higher than those in Nepal (3.6%).18 The study in Ethiopia was based on a community-level population sample, which was similar to the MMVS. However, the Nigeria study was based on women who presented for fistula screenings at health facilities. The Nepal study was based on women who had recently given birth. Studies suggest high prevalence of incontinence among women with a recent delivery,30 which might substantially increase self-reporting of fistula-like symptoms among women during postpartum.

Our results support that the prevalence of obstetric fistula is much lower than the estimates produced from self-reports of symptoms in household surveys. We recommend household surveys with adjustment as the most viable way of obtaining representative national and subnational estimates of obstetric fistula prevalence rates in LMIC settings.

Despite improvement of maternity care and substantial maternal mortality reduction in Bangladesh in the past decades, our estimates suggest that there are about 17500 women aged 15–64 years living with obstetric fistula. Only a few hundred obstetric fistula repair operations are done every year in Bangladesh.<sup>31</sup> Providing surgical treatment to so many women will need coordinated efforts, planning, allocation of resources, and training of surgeons. Two divisions in Bangladesh, Barisal and Dhaka, have high prevalence of obstetric fistula, which might need a geographically targeted approach for reaching this population. Higher reported prevalence among older women and women with low

education might indicate their limited access to fistula care and challenges to care seeking. Additionally, there are concerns of a perceived increase in iatrogenic fistula associated with a rapid increase in caesarean sections in Bangladesh in the past few years, from 9% in 2007 to 33% in 2017. Our validation study did not attempt to identify cases of iatrogenic fistula. A record review of fistula cases reported to the Fistula Care Plus project in Bangladesh showed that the percentage of cases that were iatrogenic fistula cases increased from 27% in 2012–14 to  $35 \cdot 8\%$  in 2016 and  $42 \cdot 7\%$  in 2018.<sup>11</sup>

Bangladesh recognises the problems of obstetric fistula and aims to eliminate obstetric fistula by 2030, aligning with the UN Population Fund's call for eliminating fistula globally by 2030.8 The second national strategy for obstetric fistula, developed in 2017, states that "all the girls born henceforth in Bangladesh will enjoy a life free from the risk of obstetric fistula and all the cases of fistula will receive the highest quality of reproductive health care enabling a high quality of life".<sup>32</sup> This strategy adopted a theme of zero incidence of obstetric fistula and to treat all genital fistula on a road map to a fistula-free Bangladesh by 2030. The adjusted estimates of the prevalence of obstetric fistula in Bangladesh from this study, including estimates for different regions and subpopulations, provide strengthened evidence on the scale of the problem in the country and will assist in planning to achieve a fistula-free Bangladesh by 2030.

#### Contributors

SA, SLC, SEA, MR, QN, and KJ conceptualised the study design. AA, SKNH, IIM, and SK provided fieldwork supports. SA and EHW accessed and verified the data and did the analysis, and SA developed the initial draft of the manuscript. SLC, QN, KJ, MR, and SEA edited the draft. All authors reviewed and contributed to finalisation of the manuscript. All authors approved the final version of the Article.

#### **Declaration of interests**

We declare no competing interests.

#### Data sharing

Deidentified BMMS 2016 data are publicly available at the UNC Dataverse data repository (https://dataverse.unc.edu/dataverse/d4i). Deidentified MMVS data will be made available to others upon request to the Data for Impact project (www.data4impactproject.org).

#### Acknowledgments

Funding for MMVS 2016 was provided by the US Agency for International Development (USAID) through the MEASURE Evaluation Project, Data for Impact Project and Research for Decision-Makers Project, and by UKAid through the International Centre for Diarrhoeal Disease Research, Bangladesh. Funding to support the clinical examination component of the validation study was provided by USAID through the Fistula Care Plus and MaMoni Maternal and Newborn Care Strengthening Project activity. Funding for BMMS 2016 was provided by the Government of Bangladesh, USAID, and UKAid. Additional infrastructure support for SLC, MR, and SK was provided by the National Institute of Child Health and Development of the US National Institutes of Health under award number P2C HD050924. Views expressed in this article are those of the authors, and do not necessarily reflect those of the US Government, USAID, Government of Bangladesh, and UKAid.

#### References

WHO, UNICEF, UNFPA, World Bank Group, UN Population Division. Trends in maternal mortality: 1990 to 2015. Estimates by WHO, UNICEF, UNFPA. Washington, DC: World Bank Group and the United Nations Population Division, 2015.

- 2 National Institute of Population Research and Training, International Centre for Diarrhoeal Disease Research, Bangladesh. MEASURE Evaluation. Bangladesh Maternal Mortality and Health Care Survey 2016: Final report. 2019. https://www.measureevaluation.org/ resources/publications/tr-18-297/html (accessed July 5, 2022).
- 3 Firoz T, Chou D, von Dadelszen P, et al. Measuring maternal health: focus on maternal morbidity. *Bull World Health Organ* 2013; 91: 794–96.
- 4 Chou D, Tunçalp Ö, Firoz T, et al. Constructing maternal morbidity towards a standard tool to measure and monitor maternal health beyond mortality. *BMC Pregnancy Childbirth* 2016; 16: 45.
- 5 Hardee K, Gay J, Blanc AK. Maternal morbidity: neglected dimension of safe motherhood in the developing world. *Glob Public Health* 2012; 7: 603–17.
- 6 Ahmed S, Genadry R, Stanton C, Lalonde AB. Dead women walking: neglected millions with obstetric fistula. Int J Gynaccol Obstet 2007; 99 (suppl 1): S1–3.
- 7 Wall LL. Obstetric vesicovaginal fistula as an international publichealth problem. *Lancet* 2006; 368: 1201–09.
- 8 Ahmed S, Anastasi E, Laski L. Double burden of tragedy: stillbirth and obstetric fistula. *Lancet Glob Health* 2016; **4**: e80–82.
- 9 Huda N, Richards J, Rahman L, Syed S. Why has the caesarean rate increased dramatically in Bangladesh? Int Nurs Rev 2020; 67: 231–38.
- 10 Hilton P. Trends in the aetiology of urogenital fistula: a case of 'retrogressive evolution'? Int Urogynecol J Pelvic Floor Dysfunct 2016; 27: 831–37.
- 11 Tasnim N, Bangash K, Amin O, Luqman S, Hina H. Rising trends in iatrogenic urogenital fistula: a new challenge. *Int J Gynaecol Obstet* 2020; **148** (suppl 1): 33–36.
- Ahmed S, Holtz SA. Social and economic consequences of obstetric fistula: life changed forever? *Int J Gynaecol Obstet* 2007; 99 (suppl 1): S10–15.
- 13 Adler AJ, Ronsmans C, Calvert C, Filippi V. Estimating the prevalence of obstetric fistula: a systematic review and meta-analysis. BMC Pregnancy Childbirth 2013; 13: 246.
- 14 Ahmed S, Tunçalp Ö. Burden of obstetric fistula: from measurement to action. Lancet Glob Health 2015; 3: e243–44.
- 15 Tunçalp Ö, Tripathi V, Landry E, Stanton CK, Ahmed S. Measuring the incidence and prevalence of obstetric fistula: approaches, needs and recommendations. *Bull World Health Organ* 2015; **93**: 60–62.
- 16 Maheu-Giroux M, Filippi V, Samadoulougou S, et al. Prevalence of symptoms of vaginal fistula in 19 sub-Saharan Africa countries: a meta-analysis of national household survey data. *Lancet Glob Health* 2015; 3: e271–78.
- 17 Ballard K, Ayenachew F, Wright J, Atnafu H. Prevalence of obstetric fistula and symptomatic pelvic organ prolapse in rural Ethiopia. Int Urogynecol J Pelvic Floor Dysfunct 2016; 27: 1063–67.

- 18 Chen C, Barry D, Khatry SK, et al. Validation of an obstetric fistula screening questionnaire in rural Nepal: a community-based crosssectional and nested case-control study with clinical examination. *BJOG* 2017; 124: 955–64.
- 19 Tunçalp Ö, Isah A, Landry E, Stanton CK. Community-based screening for obstetric fistula in Nigeria: a novel approach. BMC Pregnancy Childbirth 2014; 14: 44.
- 20 Swain D, Parida SP, Jena SK, Das M, Das H. Prevalence and risk factors of obstetric fistula: implementation of a need-based preventive action plan in a south-eastern rural community of India. BMC Womens Health 2020; 20: 40.
- 21 Waiz NK, Faruqui N, Begum A, Sarker S, Faisel AJ. Situational analysis of obstetric fistula in Bangladesh. 2003. https://fistulacare. org/archive/files/8/8.3/bangladesh-fistula-report.pdf (accessed July 5, 2022).
- 22 Kosinski AS, Barnhart HX. Accounting for nonignorable verification bias in assessment of diagnostic tests. *Biometrics* 2003; 59: 163–71.
- 23 Rogan WJ, Gladen B. Estimating prevalence from the results of a screening test. Am J Epidemiol 1978; 107: 71–76.
- 24 Hilden J. A further comment on "Estimating prevalence from the results of a screening test". Am J Epidemiol 1979; 109: 721–23.
- 25 Flor M, Weiß M, Selhorst T, Müller-Graf C, Greiner M. Comparison of Bayesian and frequentist methods for prevalence estimation under misclassification. *BMC Public Health* 2020; 20: 1135.
- 26 McElreath R. Statistical rethinking: a Bayesian course with examples in R and Stan. New York, NY: CRC press, 2020.
- 27 Arya R, Antonisamy B, Kumar S. Sample size estimation in prevalence studies. *Indian J Pediatr* 2012; **79:** 1482–88.
- 28 Naing L, Winn T, Rusli BN. Practical issues in calculating the sample size for prevalence studies. Arch Orofac Sci 2006; 1: 9–14.
- 29 Hall MK, Kea B, Wang R. Recognising bias in studies of diagnostic tests part 1: patient selection. *Emerg Med J* 2019; 36: 431–34.
- 30 Thom DH, Rortveit G. Prevalence of postpartum urinary incontinence: a systematic review. Acta Obstet Gynecol Scand 2010; 89: 1511–22.
- 31 Beattie K, Arrowsmith S, Delamou A, Sultana N. US Agency for International Development. Strategic Assessment of USAID Bangladesh Fistula Program. 2019. https://pdf.usaid.gov/pdf\_docs/ PA00TPZ9.pdf (accessed July 5, 2022).
- 32 Directorate General of Health Services. National strategy for obstetric fistula: 2017–2022. Dhaka: Ministry of Health and Family Welfare, 2018.