



EXPLORING THE ASSOCIATION
BETWEEN FGM/C AND FISTULA:
A REVIEW OF THE EVIDENCE

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POOJA SRIPAD
POPULATION COUNCIL

CHARITY NDWIGA
POPULATION COUNCIL

KAJI TAMANNA KEYA
POPULATION COUNCIL

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The Evidence to End FGM/C programme consortium generates evidence to inform and influence investments, policies, and programs for ending female genital mutilation/cutting in different contexts. Evidence to End FGM/C is led by the Population Council in partnership with the Africa Coordination Centre for Abandonment of Female Genital Mutilation/Cutting, Kenya (ACCAF); Gender and Reproductive Health & Rights Resource Centre, Sudan (GRACE); Global Research and Advocacy Group, Senegal (GRAG); MannionDaniels, Ltd. (MD); Population Reference Bureau (PRB); University of California, San Diego (Prof. Bettina Shell-Duncan); and University of Washington (Dr. Gerry Mackie). Evidence to End FGM/C is funded by UK aid by the UK Government.



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Please address any inquiries about the Evidence to End FGM/C programme consortium to:

Dr. Jacinta Muteshi, Project Director, jmuteshi@popcouncil.org

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Acronyms

| | |
|-------|---|
| ANC | Antenatal care |
| DfID | Department for International Development (UK) |
| EmONC | Emergency obstetric and newborn care |
| FGM/C | Female genital mutilation/cutting |
| HIC | High income country |
| LMIC | Low and middle income country |
| OF | Obstetric Fistula |
| OR | Odds Ratio |
| PC | Population Council |
| PPH | Postpartum hemorrhage |
| PNC | Postnatal care |
| RR | Relative risk |
| RVF | Recto-vaginal fistula |
| SSA | Sub-Saharan Africa |
| UNFPA | United Nations Population Fund |
| VVF | Vesico-vaginal fistula |
| WHO | World Health Organisation |

Executive Summary

Female genital mutilation/cutting (FGM/C) and fistula are both conditions that have a range of health, socioeconomic and lifestyle causes and consequences for women globally. There has been sparse empirical and conceptual effort to look at how these two conditions manifest and relate to one another. This rigorous review of the literature aims to fill this research gap by assessing the state of evidence on the association of FGM/C and fistula and conceptually mapping this association within broader social, political, and health systems contexts.

We applied a systematic approach to reviewing English-language peer-reviewed and grey literature and assessed the quality of the evidence using DfID's *Assessing the Strength of Evidence* guide (DfID 2014) to capture domains such as transparency, appropriateness, reliability, validity, and cogency. We applied a non-time bound search strategy through multiple databases using a combination of terms that captured the range of names for FGM/C and fistula. In addition, we drew on key references and global reports. A four-person reviewer team identified the final list of articles from which information was extracted using a data form with open and closed-ended questions related to FGM/C, fistula, other health and social consequences, context, population sample, study methodology, and quality indicators.

The review identified 30 full text documents of mixed quality to include in our final analysis. Eleven studies formally assessed the association of FGM/C with the occurrence of fistula, out of which eight described the conditions as positively related and three described no association. Risks of fistula development increase with cut severity. Nineteen studies speculate that these two conditions are positively and indirectly related with varying degrees of evidence. Findings also covered a range of health consequences of FGM/C and fistula, including maternal and newborn, gynecological, and psychosocial consequences; quality of care and health systems capacity moderating influences; as well as underlying factors that affect both conditions. In some studies, the proposed indirect relationship was physiologically reasoned—namely, that FGM/C-induced scarring may lead to the development of fistula by the tightening of the vaginal wall, prolonged labour, or deinfibulation following delivery or perineal tears. Others describe key determinants of both conditions that lead to a speculative positive association.

Our review also found that maternal and newborn consequences during pregnancy, labour, delivery, and postpartum are the most commonly cited as factors affecting the FGM/C and fistula relationship. Beyond obstructed or prolonged labour and fetal distress, particularly in women who have undergone FGM/C Type III which predisposes women to developing fistula, maternal complications of FGM/C include postpartum hemorrhage, perineal tears and trauma and episiotomies; adverse newborn outcomes include infant resuscitation, neonatal death, and low birth weight. Gynecological consequences of FGM/C—a subset of which are also consequences of fistula—include urogenital outcomes (e.g. scarring, keloids, abscesses, fistula, damaged tissue, and cysts), infertility, and reduced sexual functioning and satisfaction. Moreover, women who undergo FGM/C and those who develop fistula independently suffer from psychosocial consequences such as trauma, fear, depression, and divorce—all of which affect their association.

Our review additionally found that quality of care and the capacity of the health system to prevent, detect, and treat consequences of FGM/C affects the likelihood of a woman developing a fistula. Access to instrumental delivery (cesarean sections, episiotomies, and deinfibulation)—particularly in severe forms of FGM, is essential to enable women to deliver safely. Studies suggest that the efficacy of longer hospital stays for delivery among women with FGM/C is debatable in preventing fistula, but the need for sufficient human and material resource capacity for emergency obstetric care and newborn resuscitation is clear. Provider knowledge of how to counsel and treat women who have undergone FGM/C—in both native and migrant populations—is variable, but overall lacking across high, middle and low income contexts. In some cases, this is due to inadequate legal and professional guidance.

Finally, all the studies describe underlying socio- economic and contextual factors, including gender and socio-cultural norms, that affect both fistula and FGM/C. Poverty, an inability to work, living in a rural area, limited access to health services, and malnutrition (stunting) increase the likelihood of women undergoing FGM/C and developing fistula. The level of education influences a family's choice to continue or abandon the FGM/C practice for a daughter and also has implications on their awareness around the importance of antenatal care and facility delivery to prevent fistula. The underlying intersectionality of gender with socio-cultural norms around controlling one's sexuality and marriageability (FGM/C) as well as early marriage and pregnancy (increases women's risk for fistula development) all affect the social praise and sanctioning of women (cut and uncut/living without and with fistula).

We have developed a conceptual mapping framework to situate the evidence of an association between FGM/C and fistula, including intermediate and underlying factors that relate to both FGM/C and fistula high burden settings. High burden settings include high-income countries (HIC) and low and middle income countries (LMICs) where migrants or underserved populations lack access to proper pregnancy and delivery care. They also include contexts in which women have a higher exposure to sexual violence. We recommend that researchers, as well as policy and program implementers, think through and utilise the framework developed in their work to consider the intersectional influences on both conditions. Qualitatively, it is critical to explore multiple perspectives around FGM/C and fistula to better develop contextually sound interventions. Quantitatively, we recommend the use of latent variables, scales and indices to investigate the influence of positive and negative social norms, health systems, and poverty on both FGM/C and fistula in high FGM/C exposure areas. Implementation research around psychosocial factors is critical to both conditions as is girl-centered holistic programming. Finally, more research is needed on the influence of laws, policies and professional guidelines for health workers on FGM/C (e.g. related to re-infibulation and deinfibulation) and how these affect the experiences of women in HIC and LMIC contexts.

Introduction

Female genital mutilation/cutting (FGM/C) is the partial or total removal of exterior female genitalia or any sort of injury in female genitalia without medical reasons (WHO 2006). WHO categorizes FGM into four different types: Type I, or clitoridectomy, refers to the removal of total or part of the clitoris and prepuce; Type II, or excision, refers to the removal of total or part of the clitoris and labia minora; Type III, also known as infibulation, is the narrowing of the external genitalia and stitching together the edges of the vulva; Type IV, known in some contexts as *Gishiri*, is any kind of non-therapeutic procedure including pricking, piercing, incising, and scraping (WHO 2006). An estimated 200 million girls and women are currently living with FGM/C with the majority residing in 30 countries in low and middle income countries (LMICs) in Africa, Asia and the Middle East (UNICEF 2016). FGM/C in high income countries (HIC) with migrant populations, such as Australia, the United States, the European Union, Norway and Switzerland, is a recognized concern as well (Reisel and Creighten 2015).

Genital fistula is a condition in which a hole between the vagina and the rectum or bladder causes a woman to continuously leak urine, feces or both. Though fistulas can be obstetric, traumatic, or iatrogenic, the most common type is obstetric fistula (OF). Of the approximate one to two million women living with fistula globally, the majority reside in South Asia and sub-Saharan Africa (Adler et al 2013). Obstetric fistula affects about 1.57 per 1,000 women in sub-Saharan Africa (SSA). Obstetric fistula results from inadequately managed prolonged obstructed labour. Error during surgical procedures such as caesarean sections and hysterectomies can lead to iatrogenic fistulas (Barageine et al 2104, Sori et al 2016, Raassen et al 2014). Injury, including through sexual violence, can lead to traumatic fistula, although this is less common than obstetric and iatrogenic fistula in most settings (Longombe 2008). Obstetric fistula is indicative of a health system that has failed to provide accessible, timely and appropriate intrapartum care (Tunçalp 2015). The condition primarily affects those of lower socioeconomic classes, who are underprivileged, un- or under-employed, and have limited access to safe delivery attended by qualified health personnel (Wall 2012, Tebeu 2012, Suzan 2013). Women living with fistula not only suffer from a physically debilitating morbidity, but also a loss of healthy life years. The disability adjusted life years weights for vesico-vaginal (VVF) and rectovaginal fistula (RVF) are 0.342 and 0.501, respectively compared to the 0.187 associated with blindness (Salomon 2015). Women with fistula often exhibit poor health indicators including other co-morbidities of obstructed labour and may experience negative social consequences including lifelong isolation, stigma, shame, and rejection by their husband, family, and community (WHO 2006, Jones 2007, Roush 2009, Mselle 2012).

FGM/C and fistula represent complex conditions that reflect socio-economic, cultural, legal, and health systems determinants of women's health (Kimani et al 2016). There is a lack of literature on these determinants as well as the factors that are associated with prevention, identification, management, and counseling for either condition. Despite the existence of independent literature on FGM/C (Almroth-Berggren 2001, WHO 2006, WHO 2013) and fistula (Bellows et al 2015, Ahmed 2015), there is limited research on the association of the two conditions. Moreover, there is little concurrence on the intensity of the health risks associated with different types of FGM/C and how cut severity relates to various types of fistula (Slanger et al 2002). It is plausible, given the proximity and relationship of the FGM/C site to genital fistula, implications of pregnancy and childbirth, informal cutting procedures and the context of FGM/C, that FGM/C could lead to the development of fistula (obstetric, traumatic, iatrogenic). To date, to the best of our knowledge, there has been no conceptual work mapping the association between FGM/C and fistula.

This rigorous review seeks to fill this research gap and aims to 1) assess the state of evidence on the association of FGM/C and fistula, 2) conceptually map this association within broader social, political, and health systems contexts, 3) identify evidence gaps and areas for further research, and 4) develop recommendations for policy and programming.

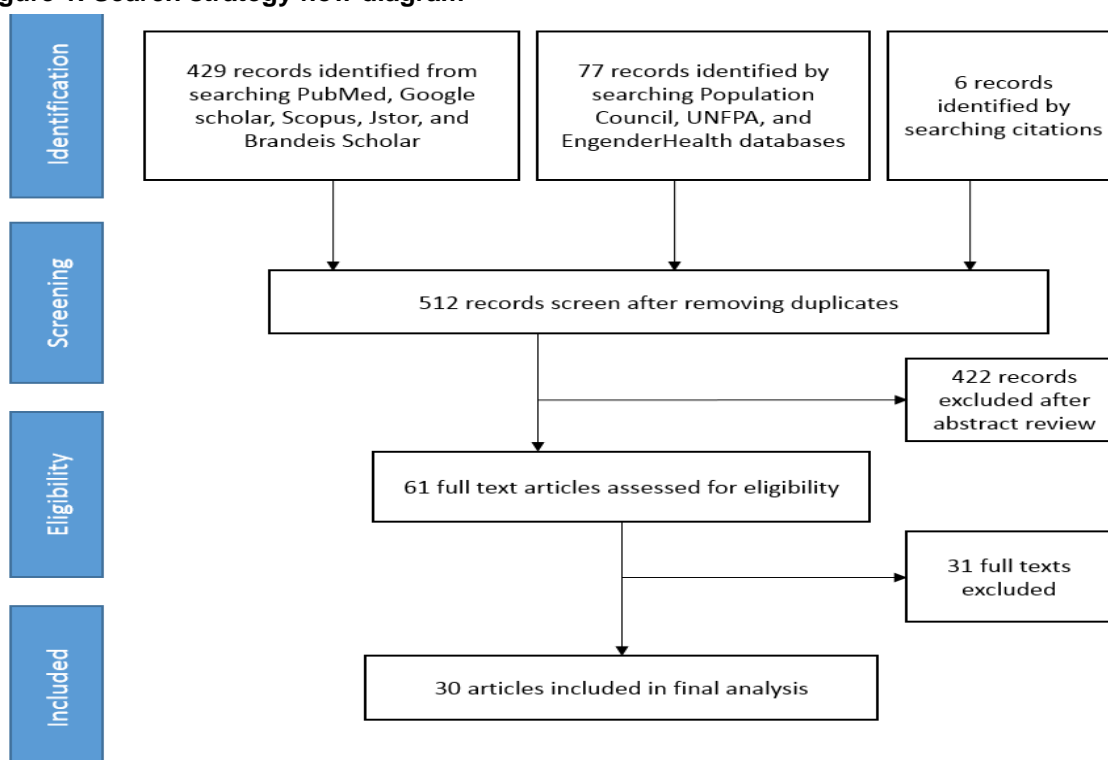
Methods

We conducted a rigorous review of English-language peer-reviewed and grey literature, drawing upon processes identified in DfID's *Assessing the Strength of Evidence* guide (2014). We applied a search strategy using the following terms in combination with each other and as Medical Subject Headings (MeSH) terms: 'female genital mutilation', 'FGM', 'female genital cutting', 'FGC', 'female circumcision', 'infibulations' AND 'fistula', 'obstetric complications', 'obstetric consequences', 'types of tears', 'excision', and 'delivery outcomes'. The databases and search engines utilised included PubMed, Google Scholar, Scopus, JSTOR, and Brandeis Scholar as well as organizational databases including Population Council, UNFPA, and EngenderHealth. Our search did not apply time nor geographic restrictions. In addition to the database searches; we drew upon key references cited in review articles and global reports.

Our search strategy (Figure 1) yielded a total of 512 records. Upon screening of titles and abstract review, 422 records were excluded and 61 articles, reports, and academic theses were identified as eligible for full text review. A four-person team read and reviewed the full text documents identified. Each full text document was reviewed by two independent readers and classified as "to include", "to exclude", "not sure". Any discrepancy between two reviewers was deliberated and jointly reviewed by the research team. Thirty full text documents were selected for inclusion in the body of evidence exploring the association between FGM/C and fistula. Articles were excluded if FGM/C and fistula were not mentioned in the articles' findings.

Data were collected through a Google survey form jointly developed by the research team after critical reflection of an initial set of peer-reviewed articles and grey literature. Descriptive measures extracted include authors' names, publication year, country or countries (region) in which the study was conducted, research type (primary, secondary), sample size and population, study design (observational, systematic review, other review), analytic methods used (qualitative, quantitative, mixed), data sources (community, facility, other), and data collection time-frame. Reviews without a specific indication of sample size (number of studies reviewed) were labeled "NI" (not indicated); similarly, facility and community data sources were classified as not applicable (N/A). Research questions, objectives, and key findings were extracted through open-ended responses.

Figure 1. Search strategy flow diagram



Both formal and speculative associations of FGM/C and fistula were abstracted from each article—formal associations refer to those that were explicitly assessed in the results’ section and speculative relationships that may be implicit or explicitly mentioned in the results and discussion with or without data and reference support. Direct, indirect, and inconclusive associations were also extracted. Formal associations were categorically deemed positive (increase in FGM/C, increase in fistula), indirect/did not describe (which included indirect associations and those that weren’t explicitly described), or none (i.e. ‘no association’). Speculative associations were classified as positive (if increase in FGM/C, increase in fistula is supported by data or reference), indirect/did not describe (indirect association through prolonged labour, but lacked explicit descriptive link), and none (i.e. ‘no association’). Negative associations between FGM/C and fistula were not seen nor extracted. Review 1 and Review 2 findings were jointly analysed and reconciled to obtain findings on the association of FGM and fistula.

Reviewers additionally assessed the health and social causes and consequences, including contextual factors that influence FGM/C and fistula using open- and close-ended questions. Obstetric and newborn complications—including postpartum hemorrhage (PPH), pre-term births, sepsis, prolonged labour, maternal death, newborn death and newborn complications – and mode of delivery (e.g. cesarean section, episiotomy, vaginal birth) were explicitly extracted. Open-ended questions about any other emerging associations with FGM/C and fistula allowed reviewers to extract data on a range of other health and social factors including physical (gynecological, sexual functioning, other) and psychological factors, health systems capacity, underlying poverty, gender dynamics, education, socio-cultural norms and legal contexts.

Finally, the quality of evidence was assessed using 19 questions, informed by DFID’s ‘Assessing strength of the evidence’ report, as listed in Table 1. Questions were evaluated as both binary and 3-point responses resulting in a total potential score of 29 points. Each article’s score was calculated and quality level classified in its relation to the maximum score as ‘high’ (>75% score), ‘moderate’ (50-75% score) or ‘low’ (<50% score).

Data were analysed using mixed methods. Closed-ended questions were tabulated and analysed quantitatively using Microsoft Excel. Textual analysis of open-ended questions around causes and consequences of FGM/C and fistula was conducted by the research team through deliberation and corroborating reviewer responses.

Table 1. Assessing quality of evidence

| Quality Area | Specific question | Responses |
|----------------------|---|--|
| Conceptual framing | Does the study acknowledge existing research? | {0: no, 1:yes} |
| | Does the study construct a conceptual framework? | {0:no, 1:yes} |
| | Does the study pose a research question or outline a hypothesis? | {0:no, 1:yes} |
| Transparency | Does the study present or link to the raw data it analyses? | {0:no, 1:yes} |
| | Did the authors describe the geography/context in which the study was conducted? | {0:no, 1:partially, 2: in a detailed way} |
| | Does the study declare sources of support/funding? | {0:no, 1:yes} |
| Appropriateness | Does the study identify a research design? | {0:no, 1:yes} |
| | Does the study identify a research method? | {0:no, 1:yes} |
| | Does the study demonstrate why the chosen design and method are well suited to the research question? | {0: not at all, 1: describes partially, 2: strongly describes} |
| Cultural sensitivity | Does the study explicitly consider any context-specific cultural factors that may bias the analysis/findings? | {0:no, 1:yes} |
| Validity | To what extent does the study demonstrate measurement validity? | {0:low, 1: moderate, 2: high} |
| | To what extent is the study internally valid? | {0:low, 1: moderate, 2: high} |
| | To what extent is the study externally valid? | {0: not, 1: somewhat, 2: high} |

| | | |
|-------------|---|---|
| Reliability | To what extent are the measures used in the study stable? | {0: <i>not very</i> , 1: <i>moderately</i> , 2: <i>very</i> } |
| | To what extent are the measures used in the study internally reliable? | {0: <i>not vary</i> , 1: <i>moderately</i> , 2: <i>very</i> } |
| | To what extent are the findings likely to be sensitive/changeable depending on the analytical technique used? | {0: <i>highly</i> , 1: <i>moderately</i> , 2: <i>not very</i> } |
| Cogency | Does the author 'signpost' the reader throughout? | {0: <i>no</i> , 1: <i>yes</i> } |
| | To what extent does the author consider the study's limitations and/or alternative interpretations of the analysis? | {0: <i>does not</i> , 1: <i>moderately</i> , 2: <i>strongly</i> } |
| | Are the conclusions clearly based on the study's results? | {0: <i>no</i> , 1: <i>somewhat</i> , 2: <i>yes</i> } |

Findings

Article descriptions

Descriptive characteristics of the 30 final documents (detailed in Appendix 1, Table 2) show that more than half (23 of 30) were published between 2000 and 2016 and comprise both grey (8) and peer-reviewed (22) literature. Documents were classified as primary research studies (n=18, 60%) and reviews (n=12) and applied qualitative (13), quantitative (13), and mixed methods (4). With the exception of one high income country investigation, the majority of studies were in Africa, in low- and medium income countries, including Nigeria (5), Kenya, (3), Ethiopia (3), Egypt (2), and Sudan (1). The 14 multi-country studies covered both LMIC and HIC settings. Six of the primary studies focused on women following their fistula repair surgeries; eight explicitly looked at women attending antenatal (ANC), maternity, family planning (FP), and postnatal (PNC) services; and four explored health service provider and other stakeholder perspectives; the latter included nurses, midwives, surgeons, and obstetricians and gynecologists as well as policymakers, opinion leaders and community members. Although the majority of data sources were collected in facilities through record review, clinical observation, and self-reporting, two qualitative studies explicitly drew on community-based sources, and one quantitative secondary analysis of household surveys. Timeframes for primary studies' data collection ranged from under six months (n=6), six months to one year (n=3), and over a year (n=6); the longest were those focused on fistula patients, typically between three and 11 years. Sample sizes ranged from eight to 35,037; eight studies had fewer than 100 respondents, three studies had between 101 and 200, and nine had over 200. Nine studies, all of which were secondary reviews, did not explicitly describe the number of articles reviewed.

An array of research questions and objectives were asked in the studies, focusing either on FGM/C, fistula, or both. Only two studies presented a conceptual framework (Berg et al 2014, Fantu 2007). With the exception of one study that explicitly set out to study the direct association of FGM/C and fistula (Browning et al 2010), rarely did the studies focus on this association. Rather, the reviewed articles sought to explore broader confounding, modifying, and mediating factors relating to both conditions. Research questions and objectives were under the following aims:

- To study the prevalence and the social and health consequences of fistula (n=5)
- To study the association between FGM/C and obstetric complications (n=7)
- To study the social and health consequence of FGM/C (n=13)
- To understand the type, context, and management of FGM (n=4)
- To study health center readiness to treat FGM/C or fistula (n=2)

Association between FGM/C and Fistula

Of the 30 articles, 11 articles formally assessed the association of FGM/C with the occurrence of fistula, out of which eight described the conditions as positively related and three described no association. Eighteen studies speculated that these two conditions were indirectly positively related with varying degrees of evidence (Table 3).

Table 3. Associations between FGM/C and Fistula

| Authors | Year Published | Type of FGM/C* | Type of association between FGM/C and fistula (formally assessed) | Type of association between FGM/C and fistula (speculative/assumed) |
|--|----------------|----------------|---|---|
| Jaldesa G, Askew I, Njue C, Wanjiru M | 2005 | NI; (e.g. III) | Positive | |
| Jones H, Diop N, Askew I, Kabore I | 1999 | I, II, III | Positive | |
| Mabeya HM | 2004 | III | Positive | |
| Saracoglu M, Zengin T, Ozturk H, Genc M | 2014 | IV (Gishiri) | Positive | |
| Sharfi AR, Elmeqboul MA, Abdella AA | 2013 | I, II, III, IV | Positive | |
| Tahzib F | 1983 | IV (Gishiri) | Positive | |
| Tukur J, Jido TA, Uzoho CC | 2006 | IV | Positive | |
| WHOa | 2000 | I, II, III, IV | Positive | |
| Browning A, Allsworth JE, Wall LL | 2010 | I, II | None | |
| Peterman A & Johnson K | 2009 | I, II, III | None | |
| Slanger TE, Snow RC, Okonofua FE | 2002 | I, II, III, IV | None | |
| Al-Hussaini TK | 2003 | I, II | | Indirect /did not describe |
| Berg RC, Underland V, Odgaard-Jensen J, Fretheim A, Vist GE | 2014 | NI, review | | Indirect /did not describe |
| Creighton SM | 2015 | NI, review | | Indirect /did not describe |
| Fantu T | 2007 | Type II | | Indirect /did not describe |
| Ibekwe PC, Onoh RC, Onyebuchi AK, Ezeonu PO, Ibekwe RO | 2012 | NI | | Indirect /did not describe |
| Kasim K, Shaaban S, Sadak AEAE, Hassan H | 2012 | NI | | Indirect /did not describe |
| Khisa M, Nyamongo IK | 2012 | NI | | Indirect /did not describe |
| Kimani S, Mutesi J, Njue C | 2016 | NI, review | | Indirect /did not describe |
| Magoha GAO, Magoha OB | 2000 | NI, review | | Positive |
| Mumtaz A, Mohammed R | 2007 | NI, review | | Indirect /did not describe |
| Onuh, SO, Igberase GO, Umeora JOU, Okogbenin SA, Otiodo VO, Gharoro EP | 2006 | NI | | Indirect /did not describe |
| Reisel D, Creighton SM | 2015 | NI, review | | Positive |
| Ruiz JI, Martinez AP, Bravo PMM, Roche PF | 2012 | NI, review | | Positive |
| Rushwan H | 2000 | NI, review | | Positive |
| Ryan F | 2012 | NI | | Indirect /did not describe |
| Teufel K, Dorfler DM | 2013 | NI, review | | Positive |
| Toubia N | 1994 | NI, review | | Positive |
| UNFPA and Engender Health | 2003 | NI | | Indirect /did not describe |
| WHOb | 2006 | I, II, III, IV | | Indirect /did not describe |

*NI: not indicated explicit

Formal assessment of association of FGM and fistula

Eleven studies, including primary and secondary designs, in LMIC settings with high FGM/C prevalence (56% to 95%), formally assessed the association of fistula and FGM/C, with mixed findings reflecting both direct and indirect relationships (Table 4). Among mostly obstetric fistula patients in Kenya, 80 percent had undergone Type III FGM/C (Mabeya 2004). In northern Nigeria, where Type IV FGM/C (*gishiri*) is common, nine to 18 percent of fistulae were attributed to trauma (i.e. cutting process or sexual violence) in addition to 60 percent attributed to obstetric situations (Tahzib 1985, Tukur et al 2006). Among women in Mali and Burkina Faso clinics who had undergone FGM/C and had developed obstetric complications, two and one percent, respectively, reported developing obstetric fistula (Jones et al 1999). Among Sudanese women attending clinics who had undergone FGM/C, 9.2 percent and 14.6 percent developed vesico-vaginal fistula (VVF) and recto-vaginal fistula (RVF), respectively. Two studies in Ethiopia and Nigeria found no association between FGM/C (Type I and II) and the development of obstetric fistula (RR/OR ~1.0).

The studies demonstrate variability and a distinct pattern of FGM/C and fistula association by FGM/C types. Positive associations are reported in studies investigating Type III (infibulation) and Type IV (*gishiri*) FGM/C only (Jaldesa et al 2005, Mabeya 2004, Saracoglu et al 2014, Tahzib 1985, Tukur et al 2006), or in studies of all FGM/C types, individually or in aggregate (Jones et al 1999, Sharf et al 2003, WHOa 2000). Studies with no evidence of association report on fistula effects in populations with greater exposure to Types I and II FGM/C and lack sufficient data to assess Type III's contribution to obstructed labour or fistula (Browning et al 2010, Peterman and Johnson 2009, Slanger et al 2002). One study (Jones et al 1999) that considered FGM/C Types I, II and III, demonstrated that the risk of developing fistula increases with the severity of FGM/C cut.

Quite a few of the formally assessed studies, including those finding no direct statistical association between FGM/C and fistula, describe indirect associations (e.g. via prolonged labour, scarring, tears) and mention contextual, personal, and health systems factors that may influence both conditions and confound the association (Mabeya 2004, Tukur et al 2006, Slanger et al 2002).

Speculative association

Out of the 30 studies, 19 suggested, either explicitly or through presentation and discussion of intermediate factors, association between FGM/C and fistula (Table 5): These studies present indirect mechanisms through which FGM/C affects fistula through related health consequences, including those that might occur at childbirth, and moderated by levels clinical management. Six multi-country reviews (Table 5) explicitly speculated that fistula is an outcome of FGM/C (mainly Type III); five indicated that fistula as sequelae to long-term obstetric complications (prolonged labour, perineal or paraurethral tears) and poor clinical management (of episiotomy or instrumental delivery), while one suggested that the transformation of an infibulation-induced infection into an abscess propels fistula development (Ruiz et al 2012). Some studies speculated that at the time of, and immediately following, cutting procedures (particularly infibulation), women are at risk for urinary and traumatic fistula, as late complications due to injury while being cut (Magoha and Magoha 2000, Muntaz and Mohammed 2007, Reisel and Creighton 2015, Ruiz et al 2012).

The other 13 studies elaborated long-term health consequences incurred during childbirth (Table 5) but implied only indirect relationships to fistula, reporting that fistula can be caused by vaginal wall tightening resulting in FGM/C-induced scarring, prolonged labour, or deinfibulation during delivery or perineal tears that may result in both RVF or VVF (Tal-Hassaini 2003, Reisel and Creighton 2015, Teufel and Dorfler, Rushwan 2000, Slanger et al 2002). Although these articles do not directly causally link FGM/C and fistula, they mention key intermediate conditions comparable to those in the articles explicitly offering speculative positive associations.

Table 4. Studies in which associations were formally assessed

| Author | Methods and sample | Prevalence of FGM/C or fistula | Study setting | Type of FGM studied | Association |
|------------------------------|--|--|---|--|--|
| POSITIVE ASSOCIATIONS | | | | | |
| Jaldesa et. al., 2005 | Mixed methods exploratory: In-depth Interview-FGDs, Facility assessments, Client exit interviews attending ANC, & service provider interviews | FGM/C: 97.5% Fistula: 2-3% (reported by nurse-midwife) | Two districts in Kenya (<i>one urban and other rural</i>) with women of the Somali ethnic community and providers | NI; (e.g. III) | Qualitative data from providers suggest FGM indirectly leads to fistula (VVF and RVF) via difficult delivery due to tightening of the vagina, prolonged labour, and perineal tears. |
| Jones et.al. 1999 | Retrospective facility and patient survey (clinical observation & self-report) 5337 women attending clinic for range of RH and maternal services | FGM/C: 93%-94% 477 (of 5337 women at clinic) women developed gynecological complications due to FGM | Mali (rural and urban) and Burkina Faso (rural) | I, II, III | Eight out of 477 women who had delivery complication had fistula. 1% of women who had undergone FGM and developed obstetric complications in Burkina Faso and Mali reported VVF; in Mali an additional 1% reported RVF. |
| Mabeya 2004 | Retrospective facility-based record review 66 women who received fistula repair surgeries | Fistula: 0.1 % | Two rural districts in Kenya | III | 80% of repaired women had undergone severe FGM/infibulation |
| Sharfi et. al., 2003 | Retrospective patient survey 2000 women (including university students and women attending outpatient clinic) | FGM/C: 73.4% Type III: 63% | A Sudanese university tertiary hospital in Khartoum | I, II, III, IV (majority: type III) | 9.2% (n=57) of women developed VVF 14.6% (n=90) developed rectal vagina fistula. VVF occurred as a result of prolonged obstructed labour. |
| Saracoglu M. et al., 2014 | Review Not Indicated | NI | Multicounty | IV (Gishiri) | <i>Gishiri</i> cut is common among Hausa's living in Northern Nigeria and Southern Niger. 5.68 to 18% of all the fistulae cases in the region result from <i>Gishiri</i> . |
| Tahzib F, 1985 | Facility record reviews 1443 fistula repair patients, out of which 80 were pediatric cases (<13 years) | NI | University hospital in, Northern Nigeria | IV (Gishiri) | 60%, 15%, and 9% of pediatric fistula cases resulted from prolonged labour, <i>Gishiri</i> cut, and trauma (sexual violence), respectively. |
| Tukur et al, 2006 | Facility record reviews 50 consecutive fistula patients | Fistula incidence: 150,000-2000,000 cases/year | Jigawa state, Northern Nigeria | IV | After prolonged obstructed labour, the most common cause of VVF was the <i>Gishiri</i> cut (18%) administered by local barbers. |
| WHOa, 2000 | Review 422 articles | NI | Multicounty | I, II, III, IV | 7 studies looked at the association, 4 of which found that Type III and IV lead to the development of fistula through obstructed labour, vulval scarring, anatomical distortion and incorrectly performed episiotomy (Damas 1972, Shandall 1967, De Villeneuve 1937 and Pieters 1972). Indirect positive association through obstructed labour was speculative (Tahzib, 1985; Harrison, 1983). |

| NO ASSOCIATION | | | | | |
|------------------------------|---|--|--|----------------|---|
| Browning. A et al 2010 | Facility record/ case notes review – comparative study 492 fistula patients who were cut (255) and uncut (237) | FGM/C: 81% | Fistula center, Amhara, Ethiopia | I, II | There is no independent association between Type I and II FGM/C the development of obstetric fistula from obstructed labour (RR = 0.93, 95% CI 0.85, 1.02). There was no association between FGC and urinary incontinence (RR = 0.92, 95% CI 0.82, 1.03). |
| Peterman A & Johnson K, 2009 | DHS secondary analysis of population-level surveys 35037 women of reproductive age (15-49 years) across Malawi (11,698), Rwanda (11,321); Uganda (8524) and Ethiopia (4,066) | NI | Community – households in Malawi, Rwanda, Uganda, and Ethiopia | I, II, III | No statistically significant evidence that FGM/C contributes to the prevalence of incontinence (OR = 0.819, 95% CI -0.299, 0.137). In this study, incontinence was used as a proxy for fistula –both traumatic and obstetric. |
| Slanger et al., 2002 | Facility-based comparative study (self-reported on first delivery) 1107 women attending FP or antenatal services (with at least one child) | FGM/C: 56% (n=621 in study); 25% (country) | Edo, Nigeria | I, II, III, IV | No difference in complications and procedures was found between those who had undergone types I and II FGM/C and those who were uncut. Both obstructed labour and perineal tears (major contributing factors to fistula) – odds were not significantly associated with FGM. |

Table 5. Speculative associations between FGM/C and fistula

| | Authors | Key findings (including proposed relationships) | Data sources |
|-----------------------------------|---|--|---|
| Speculative positive | Magoha and Magoha, 2000 | FGM/C may cause fistula in the long term through injury of urological organs such as the uterus and bladder. | Review; cited two articles as evidence (Baker et al. 1993; Dirie and Landmark 1992) |
| | Reisel and Creighton 2015 | FGM/C may lead to damage to the urethra and urethral strictures and lead to fistula formation. | Review; cited Amin et al. 2013 as evidence |
| | Toubia 1994 | Infibulated (Type III FGM/C) women: during labour, the fetal head may get obstructed and lead to VVF. | Review; cited Warsame 1989 as evidence |
| | Ruiz et.al., 2012 | Cut women may have infection that evolves into an abscess and progresses to fistula development. | Review; did not present data or refer to an article |
| | Teufel and Dorfler, 2013 | FGM/C leads to prolonged delivery resulting to VVF in some cases; in other instances, necrosis and perineal tear from FGM/C lead to fistulae. Very tight circumcision scar may lead to what is normally referred to as dribbling incontinence. | Review; did not present data or refer to an article |
| | Rushwan. H, 2000 | Infibulation could increase the risk of fistula through prolonged labour and constant pressure of the fetal head on adjacent organs. | Review; did not present data or refer to an article |
| Indirect/ did not describe | Al Hussain 2003 Berg et al. 2014 Creighton 2015 Fantu 2007 Ibekwe et al. 2012 Kasim 2012 Khisa 2012 Kimani 2016 Mumtaz & Muhammed 2007 Onuh et al. 2006 Ryan 2012 UNFPA & EngenderHealth 2003 WHO, 2006 | Described long term complication of FGM/C such as prolonged labour, perineal tear, episiotomy, postpartum hemorrhage, perineal trauma and caesarean section, difficult delivery, episiotomies. | Data includes primary data, secondary data and review |

Health and Social Consequences of FGM/C and Fistula

Nearly all the studies report that FGM/C has consequences for a woman's physical and mental health, from the moment she is cut as a child, throughout her adult life (Berg et al 2014). The most common immediate or short-term consequences described include bleeding, sepsis, urine retention, urethral injury, and urinary fistula (Sharfi et al 2013, Toubia 1994). Consequences due to fistula include serious bad odor due to continuous dripping of urine or feces and stillbirths (UNFPA and EngenderHealth 2003). Long-term health consequences of FGM/C, described in 26 of the 30 articles, include maternal, newborn, and gynecological complications. The psychosocial consequences of FGM/C overlap with those of fistula. In some cases, consequences of FGM/C are the causes of fistula.

Maternal and newborn consequences

Maternal and newborn consequences and outcomes during pregnancy, labour, delivery and postpartum are the most commonly cited intermediate factors in the FGM/C and fistula relationship. Obstructed or prolonged labour is the largest risk factor for obstetric fistula. Sixteen studies describe scenarios where FGM/C scarring, and in cases of Types III and IV, leads to a narrowing of the vaginal orifice and fetal distress. This combined with prolonged labour predisposes women to developing fistula (Mabeya et al 2004, Jaldesa et al 2005, Teufel and Dorfler 2013, Fantu 2007, Tukur et al 2006, Sharfi et al 2013, Toubia 1994, Khisa and Nyamongo 2012, Slanger et al 2002, Kasim et al 2012, Magoha and Magoha 2000). Infibulated women are 2.5 times more likely to develop any kind of obstetric complication than uncut women (Jones et al 1999). Given the nature and severity of Type III FGM/C, these women in particular risk perineal tears and need de-infibulation during ANC or delivery care to safely give birth (Toubia 1994 and Rushwan 2000).

Thirteen articles described PPH as a consequence of FGM/C or hemorrhage at the time of the FGM/C procedure (Sadak et al, Jaldesa et al 2005, Kimani et al 2016, Onuh et al 2016, Kasim et al 2012, Mumtaz and Mohammed 2007, Reisel and Creighton 2015, Ruiz et al 2012, Sharfi et al 2013, Toubia 1994, Teufel and Dorfler 2013, WHO 2006). Seven articles cite perineal tears, trauma, and episiotomy as long-term consequences of FGM/C (Creighton et al 2015, Reisel and Creighton 2015, Kimani et al 2016, Magoha and Magoha 2000, Slanger et al 2002, WHO 2006, WHOa 2000). At least six articles mention maternal death as a consequence of FGM/C (WHOa 2000, Kimani et al 2016, Tukur et al 2006, Ruiz et al 2012, Teufel et al 2013) and 11 report newborn deaths or health complications due to FGM/C (WHOa 2000, Tukur et al 2006, Jones et al 1999, Toubia 1994, Ruiz et al 2012, Teufel and Dorfler 2013, Fantu 2007). Similarly, stillbirths often co-occur with obstetric fistula, given the precursor of prolonged labour (UNFPA and EngenderHealth, 2003). According to one systematic review (WHO 2006), despite unclear explanatory mechanisms, women who have undergone FGM/C are more likely to suffer adverse newborn outcomes including infant resuscitation, neonatal death, and low infant birth weights.

Gynecological consequences

The gynecological consequences of FGM/C revealed in the studies include urogenital outcomes, infertility, and sexual functioning and satisfaction. Urogenital outcomes such as scarring, keloids, abscesses, fistula, damaged tissue (perineum, anal sphincter), disfigurement, vaginal obstruction, and cysts are mentioned in the background sections of studies but are not frequently reported in the research findings (Kimani et al 2016). Five of the 30 articles describe urogenital infection, urine retention, and other moderating factors after FGM/C that could lead to fistula development (Creighton et al 2015, Magoha and Magoha 2000, Sharfi et al 2013, Tukur et al 2006). To address these compounding factors in clinics, some studies stress the need for specialised provider training

and to involve urologists in fistula surgery teams for successful repair (Creighton et al 2015, Magoha and Magoha 2000). Although it may not have implications on obstetric fistula, infertility, reported as a late complication of FGM/C (Tahzib 1983), is more common (2.9 times increased likelihood) among cut women (Kasim et al 2012). Quite a few articles describe women who had undergone FGM/C as more likely to report painful intercourse, no sexual desire, less sexual satisfaction, and less experience of orgasm compared to uncut counterparts (Khisa and Nyamongo 2012, Kimani et al 2016, Saracoglu et al 2014, Sharfi et al 2013).

Psychosocial consequences

Women who undergo FGM/C as well as those who develop fistula independently suffer a number of psychosocial consequences. Understanding how these consequences interrelate may have implications for the associations between the two conditions. At least five articles found that women who had undergone FGM/C suffered from psychosocial consequences such as trauma, fear, depression; in some cases, these led to or were compounded by divorce (Tahzib et al 1983, Creighton et al 2015, Browning 2010, Jaldesa et al 2005, Saracoglu et al 2014, Teufel and Deufel 2013). In cases where women migrate to new contexts where FGM/C is less prevalent, the shock around its non-normative practice also has psychosocial consequences for a woman (Teufel and Deufel 2013). Psychosocial consequences suffered by women living with fistula include isolation, prejudice, depression, and anxiety, many of which are mitigated by divorce or living separately from one's husband or family (Khisa 2015, Tahzib 2015).

Quality of care and health system capacity

Quality of care and the capacity of the health system emerged as a potential mediator in the association between FGM/C and fistula. Qualitative and quantitative findings suggest that improving the capacity of the health system to prevent, detect, and treat consequences of FGM/C affects the progression to fistula (Sharfi 2013, Mabeya 2004, WHO 2006, Onuh et al 2006). Twelve out of the 30 articles describe that FGM/C and delivery type, including access to instrumental delivery (cesarean sections, episiotomies, de-infibulation), is essential to enabling women to deliver safely (Slanger et al 2002, Teufel et al 2013, Onuh 2006, Creighton 2015, Kimani et al 2016). In contexts where a needed episiotomy was not performed, women who have undergone FGM/C have 1.31 to 3.19 times higher relative risk of a perineal tear compared to uncut women (WHO 2006). This risk increases with cut severity: 1.31 greater for those with Type I, 1.92 times greater for those with Type II, and 3.19 for those with Type III. Instrumental delivery is critical, especially for women living with Types III and IV. Some studies observed that women with FGM/C require extended hospital stays (WHO 2006), while others suggest no difference (Creighton 2015, Browning 2010). Many studies advocate that hospitals need human and material resource capacity for newborn resuscitation and emergency obstetric care (EmOC) to avoid fistula, mitigate other maternal consequences (potentially intensified by FGM/C), and avoid newborn death or morbidity (Browning 2010, Kimani et al 2016, WHO 2006). The provision of timely and personalized ANC for women with FGM/C where women are counseled on the importance of skilled birth attendance and deinfibulation in preparation of a vaginal delivery also emerged as critical in HIC and LMIC contexts (Jones et al 1999, Rushwan 2000, UNFPA and Engender Health 2003).

Provider knowledge of how to counsel and treat women who have undergone FGM/C is variable by context. There was less understanding in HICs as FGM/C cases were concentrated among migrant women (Creighton 2015, Rushwan 2000, Reisel and Creighton 2015). In these settings, inadequate or a complete lack of legal and professional guidance constrains providers from managing FGM/C clients appropriately—including policies on re-infibulation and de-infibulation (Reisel and Creighton 2015). De-infibulation by traditional birth attendants (or unskilled attendants)

can cause fistula in some cases (Browning 2010, Toubia 1994, Onuh et al 2006). One study in Kenya showed five in eight women with FGM/C developed fistula during their first delivery due to the unskilled birth attendant with whom they delivered (Khisa and Nyamongo 2013). Capacity building of skilled assistants in many LMICs where women have less access is critical (Jaldesa et al 2005).

Underlying contextual factors influencing both fistula and FGM

Nearly all studies in either the results or interpretation sections, touch on one or more underlying socio-economic and contextual factors that affect both fistula and FGM/C, including poverty, place of residence, and malnutrition. For example, women living in poverty, who are unable or not allowed to work for pay, have limited access health services and have a higher likelihood of continuing FGM/C, are at greater risk of developing fistula. These women have the lesser means to deal with the health and social consequences of either condition (Ryan 2012, Jones et al 1999, Fantu 2007). Women living with fistula are further unable to work given the constant smell they emit, which plays into their cycle of poverty (UNFPA and EngenderHealth 2003). In high prevalence FGM/C areas, malnutrition, which stunts pelvic growth predisposing some women to prolonged labour and elevates their risk of fistula, may further contribute to FGM/C-fistula indirect association (Mabeya 2004). The level of education may influence a family's choice to continue or abandon the FGM/C practice for a daughter. Mothers and fathers with more schooling tend to discontinue the practice over generations (Fantu 2007, Ibekwe et al 2012). Education also affects awareness around the importance of antenatal care and facility delivery, both of which help women with FGM/C to identify prolonged labour and seek skilled care to prevent fistula.

Studies also demonstrate that gender and socio-cultural norms with respect to women's relational roles to spouses and families affect FGM/C and fistula. While early marriage and pregnancy places women at risk for fistula (Mabeya 2004, UNFPA and EngenderHealth 2003), FGM/C is often described as way to control women's sexuality (Ibekwe et al 2012). Moreover, the reduced sexual satisfaction and strained marital relationships due to FGM/C and fistula described by many studies (UNFPA and EngenderHealth 2003, Jaldesa 2005, WHO 2006) have implications for gender-dynamics in households. In both cases, the intersectionality of gender and socio-cultural preferences around women are at stake. Socio-cultural, including religious, norms around marriageability, virginity, sexuality and social status influence FGM/C (Magoha and Magoha 2000, Kimani et al 2016, Ruiz et al 2012, Saracoglu et al 2014). Social sanctioning for uncut women (un-marriageability, promiscuity, dishonor) tends to sustain the FGM/C practice (Ibekwe et al 2012, Browning et al 2010), though these normative claims are rarely assessed in quantitative assessments. Social sanctioning of women living with fistula in many cases relates to their isolation from family and community spaces as well as their inability to remain employed due to their leaking condition (Khisa and Nyamongo 2012, Fantu 2007, Onuh et al 2006). Less information exists on sexual violence and/or rape though it is described as influencing fistula formation (Peterman and Johnson 2009), there is insufficient evidence on how it affects the relationship of FGM/C's with fistula (Tahzib 1985, Fantu 2007).

Quality of evidence

The quality of evidence is mixed on the association of FGM/C and fistula (Table 2). Of the 30 final documents, 12 (40%) are of high quality, 13 (43%) of medium quality, and five (17%) are classified as low quality studies. This mixed level of evidence on a relatively under-studied issue reflects manuscript transparency, cogency, and moderate reliability and transferability of findings, as opposed to an inability to draw significant causal inferences from the limited study designs. Most were facility-based record reviews, of both primary and secondary designs, that although provide

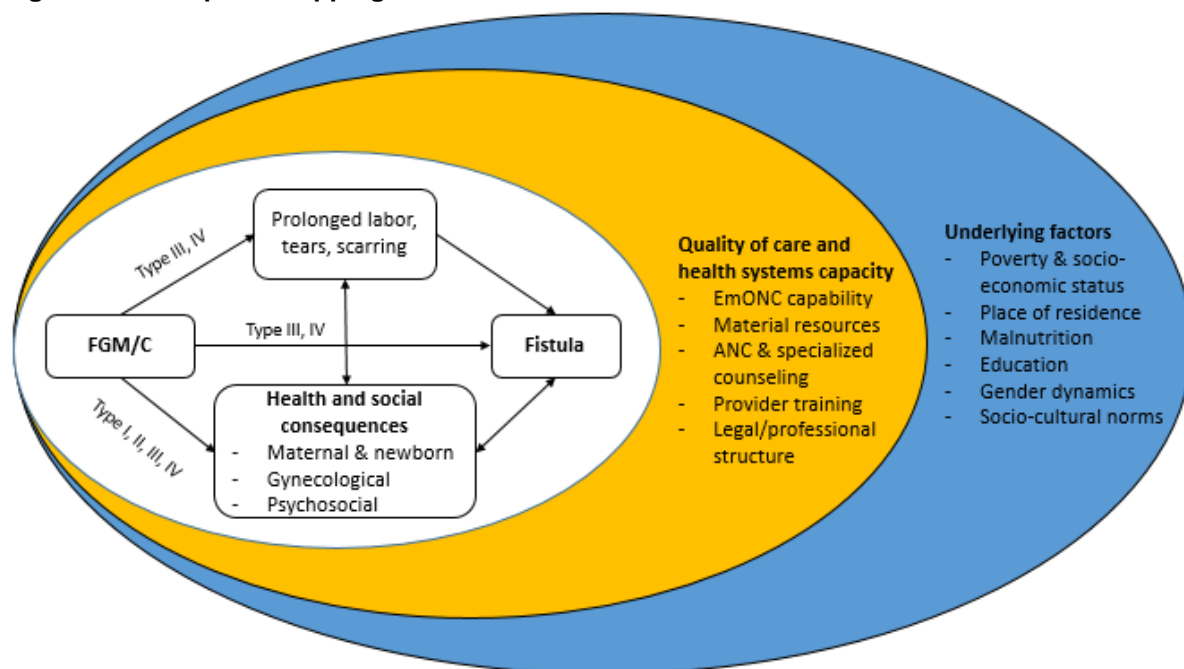
clinical evidence, do not enable us to ascribe a population-based assessment of the association of FGM/C and fistula. Comparative studies that looked at uncut and cut women were helpful, although rarely was fistula a focused outcome. Primary qualitative and quantitative studies were strongest when they insight into the broader context that influenced both FGM/C and fistula.

Discussion

Summary and conceptual mapping

This review sought to describe the existing evidence on the association of FGM/C and fistula and found mixed evidence that reflects both direct and indirect relationships. We found 30 peer-reviewed and grey literature studies that commented on both conditions; while 11 documents formally demonstrated positive and no association between FGM/C and fistula, many others speculate about positive relationships. The dissonance between speculative and evidence-based associations demonstrates a lack of formal rigorous quantitative measures (relative risks and odds ratios) investigating the FGM/C and fistula relationship. Interestingly, the three documents that confirmed no association were carried out in contexts of predominantly FGM/C Types I and II, while those that saw positive associations covered all or focused on FGM/C Types III and IV, indicating a potential dose-response of severity, i.e. the more severe the cut, the more likely a woman is to develop pregnancy and other complications that lead to fistula development. Within these higher severity types, Type III was often associated with obstetric fistula, while Type IV was additionally linked to traumatic fistula caused by cutting procedures themselves. We also found that the indirect association of FGM/C and fistula, including via intermediate health and social consequences of FGM/C, mediating health systems challenges, and underlying socio-economic and context factors, are critical to consider in conceptually framing the two conditions in a way that helps frame implications for research and practice (Figure 2).

Figure 2. Conceptual mapping of associations between FGM/C and fistula



EmONC refers to emergency obstetric and newborn care and ANC refers to antenatal care

This conceptual framework demonstrates the complexity of factors identified through our review that affect both FGM/C and fistula. In addition to the direct and mediated pathway through which FGM/C leads to the development of fistula, there are health consequences of both conditions that may influence the association. Though the data on severity is mixed and insufficiently identified in articles (Table 3), the conceptual framework maps our current understanding. Moreover, the similarities in sociodemographic, economic and cultural context of women exposed to both FGM/C and fistula is reflected in their lack of access to health care services broadly and in the specific context of pregnancy and childbirth; this moderates the association between the two conditions.

Quality of antenatal and delivery care, including the facility material and human resource mix, adequate legal and professional guidance, as well as the competency and sensitivity required of providers tasked with counseling women exposed to both conditions plays a mitigating role. Finally, FGM/C and fistula are situated in settings with shared underlying societal factors that affect our ability to capture a measurable association between the conditions. The intersectional influence of socio-economic (wealth, education) and poverty statuses—including women’s abilities to maintain employment, gender dynamics including women’s autonomy to make health care-seeking decisions, socio-cultural norms and sanctions related to marriageability, cleanliness, and women’s sexuality—determine both FGM/C and fistula. These underlying and health systems causes and consequences of both conditions demand more attention in terms of research, programming, and policy in LMICs and HICs.

Limitations

We faced several challenges in carrying out this review. First, given the complexity of both conditions, there were only a few studies that captured a diverse range of perspectives (qualitative) and reached adequate samples to comment statistically significantly on associations and their strength (quantitative). Secondary analyses were predominantly clinical cases, or other reviews, that did not adequately define replicable methodologies or provide clear evidence of assessing an association. A number of studies took place in facility settings where women received surgical repair or instrumental delivery to safely give birth. By contrast, there were relatively few studies that drew on population-based surveys or targeted community samples; our review had insufficient data from women and men living in communities with high burden of FGM/C and fistula. This selection bias likely lead to an underreporting of evidence on the association of FGM/C and fistula, given that these groups are less likely have access to medical services overall and more likely to experience a co-occurrence of both conditions. Also, a paucity of longitudinal quantitative designs limits our ability to detect causal inference; however, conducting such studies is not pragmatic in most settings given the long data collection time-frame required to collect relevant information, the relative rarity of fistula, and the high rates of loss-to-follow up (WHO 2006).

Implications for research, programming, and policy

Despite these limitations, this rigorous review of the literature points to the lack of an in-depth understanding around the contexts in which associations between various types of FGM/C and different types of fistula exist and require more deliberate programming efforts. The conceptual mapping presented in this report may serve as a particularly useful tool for those involved in programming in that it may help decision makers think through the various consequences, positive and negative, of intervening at various levels. First contexts in which FGM/C is a norm ought to more systematically take the associated risks of fistula and other childbirth outcomes into account. For example, the type of provider counseling intervention may differ between HIC and LMIC contexts based on the types of FGM/C and fistula women typically present with. A social norms targeting program strategy may require a variety of inputs including increasing male and female awareness around the risks of FGM/C on maternal and newborn outcomes and soliciting buy-in of community leaders. The quality of care and underlying factors that affect both FGM/C and fistula point to the importance of advocating for multifaceted girl-centered policy and programming.

There is a need to build the evidence base around understanding and addressing the association of FGM/C and fistula. A second phase of this study is planned and will undertake limited multivariate analyses of the DHS/MICS datasets from multiple African countries that have the relevant data to investigate the associations, causative factors and relevant correlates for FGM/C and fistula. Comparative research across high and low burden FGM/C settings, types of cutting,

socio-economic status, and country policy environments would be useful for shared learning on this ambiguous area in both LMIC and HIC contexts. We recommend that researchers focus on studying confounders, mediators, and underlying factors as related to both FGM/C and fistula at the community level and in high burden settings where both are prevalent. High burden FGM/C settings may be in HICs and LMICs, including among migrants or other underserved populations that lack access to proper pregnancy and delivery care. The association of FGM/C and traumatic fistula may be directly explored in contexts where women are exposed to sexual violence, for example in conflict and post-conflict settings. Traumatic fistula should also be further studied in contexts where unskilled persons are performing FGM/C procedures and unintended injury directly results in fistula formation.

We recommend utilising the conceptual mapping as a guide to look at different complex relationships between factors through both qualitative and quantitative means. Qualitatively, it is critical to explore multiple perspectives around FGM/C and fistula to better develop contextually sound interventions. Quantitatively, we recommend the use of latent variables, scales and indices to investigate the influence of positive and negative social norms, health systems, and poverty on both FGM/C and fistula in high FGM/C exposure areas. Implementation research around psychosocial interventions that addresses depression, anxiety, social and cultural challenges associated with both conditions is a key investigative space to pursue—especially in LMIC contexts where the double burden of FGM/C and fistula is more pronounced. Finally, it is important to conduct further nuanced research on the influence of laws, policies and professional guidelines for health workers around FGM/C (e.g. related to re-infibulation and deinfibulation) and how these affect the experience of women in HIC and LMIC contexts.

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Appendix 1

Table 2. Descriptive characteristics of final documents

| Authors | Year published | Country/ies (region) | Research type | Sample size | Study design | Methods | Data sources | Collection time-frame | Quality |
|---|----------------|------------------------|---------------|--|-------------------|--------------|---------------|-----------------------|---------|
| Al-Hussaini TK | 2003 | Egypt | Primary | 306 primigravida maternity admissions | Observational | Quantitative | Facility | 21 months | medium |
| Berg RC, Underland V, Odgaard Jensen J, Fretheim A, Vist GE | 2014 | Multi-country | Secondary | 57 studies | Systematic review | Qualitative | NA | NA | high |
| Browning A, Allsworth JE, Wall LL | 2010 | Ethiopia (Amhara) | Primary | 492 fistula patients (cut:255, uncut:237) | Observational | Quantitative | Hospital | 3 years | high |
| Creighton SM | 2015 | UK | Secondary | NI | Other review | Mixed | NA | NA | medium |
| Fantu T | 2007 | Ethiopia (Addis Ababa) | Primary | 422 fistula patients | Observational | Mixed | Health center | 2 months | high |
| Ibekwe PC, Onoh RC, Onyebuchi AK, Ezeonu PO, Ibekwe RO | 2012 | Nigeria (Southeast) | Primary | 320 antenatal and postnatal women | Observational | Quantitative | Facility | 4 months | medium |
| Jaldesa G, Askew I, Njue C, Wanjiru M | 2005 | Kenya | Primary | 29 influential persons, 14 groups with community members, 15 facility assessments, 101 antenatal clients, 44 service providers | Observational | Qualitative | Community | 3-5 months | high |
| Jones H, Diop N, Askew I, Kabore I | 1999 | Burkina Faso & Mali | Primary | 7257 women attending clinic for antenatal, FP, ObGyn services (Burkina Faso: 1920; Mali: 5337) | Observational | Quantitative | Facility | 4 months | high |
| Kasim K, Shaaban S, Sadak AEAE, Hassan H | 2012 | Egypt (Alexandria) | Primary | 200 circumcised & 200 uncircumcised women attending maternal clinics | Observational | Mixed | Facility | 7 months | high |

| | | | | | | | | | |
|---|------|----------------------------------|-----------|--|-------------------|--------------|----------------------------|---------------------------------|--------|
| Khisa M, Nyamongo IK | 2012 | Kenya (West Pokot) | Primary | 8 women with fistula, 2 FGDs: fistula survivors and persons attending awareness seminar | Observational | Qualitative | Community | 2 months | medium |
| Kimani S, Mutesi J, Njue C | 2016 | Multi-country | Secondary | 44 reference materials related to FGM/C | Systematic review | Qualitative | NA | NA | high |
| Mabeya HM | 2004 | Kenya (West Pokot) | Primary | NI (women treated over 5 year period) | Observational | Quantitative | Health center | 5 years | medium |
| Magoha GAO, Magoha OB | 2000 | Multi-country (Africa) | Secondary | NI | Other review | Qualitative | NA | NA | medium |
| Mumtaz A, Mohammed R | 2007 | Multi-country (UK and HICs) | Secondary | NI | Systematic review | Quantitative | NA | NA | low |
| Onuh, SO, Igberase GO, Umeora JOU, Okogbenin SA, Otiode VO, Gharoro EP | 2006 | Nigeria (Benin) | Primary | 193 nurses | Observational | Quantitative | Health center | 3 months | medium |
| Peterman A & Johnson K | 2009 | Malawi, Rwanda, Uganda, Ethiopia | Secondary | 35037 women of reproductive age (15-49 years) | Observational | Quantitative | Community | ~1 year (DHS household surveys) | high |
| Reisel D, Creighton SM | 2015 | Multi-country | Review | NI | Other review | Qualitative | NA | NA | medium |
| Ruiz JI, Martinez AP, Bravo PMM, Roche PF | 2012 | Multi-country | Secondary | NI | Other review | Qualitative | NA | NA | medium |
| Rushwan H | 2000 | Multi-country | Review | NI | Other review | Qualitative | NA | NA | low |
| Ryan F | 2012 | Ethiopia | Primary | 15 interviews with & participant observation of missionaries, local medical staff and fistula patients | Observational | Qualitative | NA | NA | medium |
| Saracoglu M, Zengin T, Ozturk H, Genc M | 2014 | Multi-country | Secondary | NI | Other review | Qualitative | NA | NA | low |
| Sharfi AR, Elmeqboul MA, Abdella AA | 2013 | Sudan (Khartoum focus) | Primary | 2000 women of reproductive age (attending clinic & not) | Observational | Quantitative | Health center & university | 5 years | medium |

| | | | | | | | | | |
|---|------|---|-----------|--|-------------------|--------------|---------------|-----------|--------|
| Slanger TE, Snow RC, Okonofua FE | 2002 | Nigeria (Edo) | Primary | 1107 women attending FP or antenatal services | Observational | Quantitative | Health center | 8 months | high |
| Tahzib F | 1985 | Nigeria (Northern) | Primary | 80 fistula patients (<13 years of age) | Observational | Quantitative | Health center | 11 years | medium |
| Teufel K, Dorfler DM | 2013 | Multi-country (Africa) | Primary | NI | Other review | Qualitative | NA | NA | low |
| Toubia N | 1994 | N/A | Review | NI | Other review | Qualitative | NA | NA | low |
| Tukur J, Jido TA, Uzoho CC | 2006 | Nigeria | Primary | 50 fistula patients | Observational | Quantitative | Health center | 4 years | medium |
| UNFPA and Engender Health | 2003 | Benin, Chad, Mali, Malawi, Mozambique, Niger, Nigeria, Uganda, Zambia | Primary | >35 facilities; government and UNFPA representatives & relevant stakeholders | Observational | Mixed | Health center | 6 months | high |
| WHOa | 2000 | Multi-country | Secondary | 422 articles | Systematic review | Qualitative | NA | NA | high |
| WHOb | 2006 | Burkina Faso, Ghana, Kenya, Nigeria, Senegal and Sudan | Primary | 28393 women attending maternity services | Observational | Quantitative | Health center | 17 months | high |