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Towards developing a guideline to inform policy on foetal alcohol spectrum disorder: A study protocol

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ABSTRACT

Introduction: Maternal alcohol consumption during pregnancy results in mental and physical birth defects in individuals. These birth defects are usually described as Foetal Alcohol Spectrum Disorder (FASD). With an estimated of 183- 259 per 1000 children born with FASD, South Africa is identified to have the highest prevalence of FASD in the world. Nevertheless, there is a lack of relevant policies, guidelines, and interventions addressing the issues around FASD. This protocol outlines a proposed process for developing a guideline to inform policies on FASD.

Methods and Analysis: This process will have three phases. In phase one, we plan to conduct a document review of available policies on the prevention, treatment, and management of FASD and update the existing systematic review on FASD interventions. The aim of the two reviews is to explore the availability and content of existing policies and interventions on FASD. In addition, we will conduct an exploratory qualitative research to obtain the perspectives of various stakeholders including the Department of Education, the Department of Health and Social Development of the existing or possible guidelines and policies for the management of FASD and available interventions and services. In phase two, we will aggregate the findings of the previous phase to develop a prototype guideline. In phase three, using the developed prototype, we will apply the Delphi approach with experts on FASD, soliciting their opinions on the nature and content of the proposed guidelines for policies. The information gathered will be used to modify the prototype to formulate a policy guideline on FASD. Data will be analysed using thematic analysis and narrative synthesis.

Ethics and Dissemination: Ethical clearance has been obtained from the ethics committee of the University and governmental departments. The findings will be

disseminated through publications and the guideline will be submitted to relevant departments.

Strengths and limitations of this study

- It proposed multiple sources for data collection.
- It proposed proven methodology such as Delphi technique to develop the guideline.
- This study will not include people with FASD.
- This study is expected to influence policy nationally, however, it will be conducted in only one province out of nine provinces.

Introduction

Foetal alcohol spectrum disorders (FASDs) refer to an array of birth disorders produced by foetal exposure to alcohol during pregnancy. The word “foetal alcohol syndrome” was not used until 1973. This was when the negative effects were medically recognized [1, 2]. The word “fetal alcohol spectrum disorder” not was used until 2002 at a conference organized by the United States of America (USA) National Organization on Fetal Alcohol Syndrome (NOFAS). The new term was used to describe the range of outcomes that may occur as a result of prenatal alcohol exposure [3]. (FASDs) include foetal alcohol syndrome (FAS), partial FAS, alcohol-related neurodevelopment disorders (ARND) and alcohol-related birth defects (ARBD). FAS is identified as the most serious anomalies among FASDs [4].

FASD leads to primary and secondary disabilities. Primary disabilities are those that the child is born with [5], resulting in damages to the brain domains that are responsible for physical motor skills, sensory processing skills, cognition, communication skills, academic achievement, memory skills, executive functioning and abstract reasoning, attention deficit/ hyperactivity, and adaptive skills [6, 7]. It can also result in physical abnormalities such as ante- and post-natal growth retardation, eye and ear malformations, mouth and jaw deformation, skeletal defects, organ pathology, sensory deficits, and impaired immune system [4]. Secondary disabilities relate to those disabilities that developed because of lack of timely and appropriate interventions to primary disabilities. These include: fatigue, frustration, anxiety, fearfulness, rigid, resistant, argumentative behavior, becoming overwhelmed, shut down (withdrawn), a poor self-concept, feelings of failure, low self-esteem, isolation, acting out, aggression, family and/or school problems, depression and other mental health problems,

trouble with the law, drug and alcohol problems, problems with employment, and homelessness [4, 8].

Studies conducted in United States of America (USA), South Africa, Canada and Australia have identified various maternal risk factor for FASD. These include demographic factors (socioeconomic status, employment status, educational status, marital status, religion, living area, income and age), psychiatric and neuropsychological factors (psychiatric comorbidity, identified stressful, physical aggression and sexual abuse), family and socials (family lifestyle, drinking habit, alcohol use, illegal drugs use and tobacco use) and pattern of alcohol consumption [9].

The prevalence reported in the literature for FAS/FASD were conducted in certain regions of various countries. Therefore, there is no national prevalence of FAS /FASD anywhere in the world [10].The prevalence of foetal alcohol spectrum disorders vary from one country to another, in the USA and Western Europe, it is assumed to be between 2% to 5% and FAS ranges from 0.2 to 9 per 1000 live births [11]. Zelner and Koren [12] estimated the prevalence of FAS and FASD at 1 to 3 and 9 per 1000 live births respectively in Canada. In Italy, a study estimated the prevalence rate of 3.7 to 7.4 and 20.3 to 40.5 per 1000 births for FAS and FASD respectively [13]. In South Africa, a study conducted in four rural communities reported highest prevalence rate ever recorded in any part of the world. It was estimated to be 93-128 per 1000 children for FAS and 183 to 259 per 1000 children for FASD [14].

Prevention, care and support for persons affected by FAS and other alcohol-related disorders are not being implemented, particular in developing countries. Evidence shows that only in developed countries like Sweden and Canada that provide universal health care (health care

for all), that are making effort to recognise and accommodate the needs of women with an alcohol problem and their children. Active and coordinated efforts towards addressing alcoholism in women and their children were not evident in other parts of the world including South Africa [15]. Furthermore, only the USA and France have been successful to have Alcohol Labelling Act as part of their federal law [15]. Nevertheless, no country has successfully implemented restrictions on public advertising of alcohol beverages in print media, radio and television, and no medical school has introduced courses on FASD in any part of the world [15].

Background

South Africa is reported to have escalating levels of alcohol consumption [16], particularly in the informal settlements in the Northern and Western Cape Provinces [17–21]. The prevalence of alcohol use among pregnant women in the Cape Metropole, Western Cape Province of South Africa is estimated at 19.6% and in poor communities of the Western and Northern Cape provinces of South Africa, FASD (FAS/PFAS) is endemic [22–24]. Maternal alcohol drinking during pregnancy, especially in rural and informal settlements, has made FASD a huge problem in South Africa.

Generally, FASD is relatively unknown among health professionals and has not been given adequate consideration in policies, programmes, and interventions [25]. This is probably because FASD is not yet formalized as a medical diagnosis, even though it is now over 40 years that it was first diagnosed [25]. In spite of this, efforts have been made to recognize FASD as a public health problem. Because FASD is preventable, there have been efforts to advocate for the implementation of various prevention programme and strategies for its prevention. These strategies advocated for include adequately resourced alcohol control

services, support for pregnant women, and a sustainable commitment from communities, service providers and the government in addressing alcohol-related health and psychological problems [26, 27]. For these strategies to be effective and sustainable, a governing policy and coordinated efforts are required.

Globally, there is inadequate policy and guidelines for the management and prevention of FASD. A review on FASD in Africa reported gaps in policy and service implementation [28]. In South Africa, only two of the national policy documents namely, the National Human Genetics Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disabilities (2001) and the National Drug Master Plan (2007) scanned during a situation analysis by Rendall-Mkosi et al. used the term FAS and none of them mentioned FASD [29]. In addition, the South African Guidelines for Maternity Care (2002) only specifies that maternal alcohol, as well as tobacco and other substances, use behaviours should be taken into consideration when taking a medical history of a woman [29]. This reflects the extent to which issues related to FASD are being considered in policy.

Furthermore, current national hospital-based birth defects surveillance system developed by the National Department of Health excludes FASD [29]. This surveillance system mainly considers birth defects that can be clinically diagnosed at birth or few days after birth, which can only be identified if a woman is hospitalised after birth. The exclusion of FASD in the birth defects surveillance system is believed to be as a result of the difficulty in diagnosing FASD at birth as most of its features are rarely manifested at birth and it requires a multidisciplinary team to arrive at a definitive diagnosis [29]. The above gap speaks to the need to develop relevant policies and guidelines to identify FASD and in order to guide strategic implementation of appropriate interventions to address FASD in South Africa.

In 2001, the Western Cape Provincial executive council acknowledged FAS as a provincial health priority for the years 2001 and 2002 [30, 31]. This led to the establishment of the Western Cape Provincial FAS reference and working group (now called FASD task team). The FASD Task Team of South Africa comprises of members from the Department of health, the Department of Social Services, the Department of Education, the Medical Research Council, the University of Stellenbosch, the University of Cape Town, NGOs such as Foundation for Alcohol Related Research (FARR), Dopstop and Pebbles project. It was headed by the provincial Maternal, Child and Women's Health Deputy Director [30, 31]. The aims of the FASD task team is to raise awareness on FASD, create strategies for prevention and, share skills and information [30, 31]. The FASD task team has been successful in the development of FAS training manual for professional, designed posters for awareness on FASD, organizing public awareness on International FASD Days, organizing FASD training workshop and, development of website and Facebook page on FASD [30, 31]. The above development is an important and commendable step in developing the capacity to support management of FASD at the provincial level. However, a multi-sectoral national FAS Task Team is needed for a coordinated response is expected to address FASD in the South African population. Strengthening the newly established National FAS Task Team by the Department of Health, and the establishment of provincial level Task Teams nationally has been advocated to ensure a pragmatic coordinated national response for FASD [29]. To this end, we are proposing a national FASD guideline that will inform policy on coordinated and multi-sectoral response to FASD in South Africa. In this protocol, we propose systematic steps to developing a guideline to inform policy for FASD prevention and management.

Research aim

The aim of this protocol is to propose a process for developing a guideline to inform the development and improvement of policy on FASD.

Research objectives

To achieve the aim, we developed the following objectives:

- To review policies and their contents on FASD internationally and in South Africa;
- To explore and describe the content of FASD policies from the perspective of policy makers;
- To provide update on the interventions on FASD internationally and in South Africa;
- To explore and describe the perspectives of service providers involved in the implementation of identified FASD intervention activities.
- To apply the Delphi technique to guide the development of guidelines for an integrated policy on FASD for the South African context.

Understanding policy guidelines

According to the World Health Organization, a guideline is a document that contains recommendations on health interventions for clinical or public health [32, 33]. It is also described as a document that makes evidence-based recommendations for averting and treating certain conditions, improving health, managing medicines, planning of services for the community, development of an intervention for enhancing population health and to deliver social care for people [34]. Guidelines promote individualised and integrated care,

help service providers and recipients, and other stakeholders to make informed decisions and serve as a response to a condition that needs urgent attention [32–35]. In addition, a guideline is considered as a statement, recommendation or best practice created to give a framework for policy implementation [35]. As applies to this protocol, we considered a guideline as a document that contains recommendations that will inform the development of integrated policy on FASD.

Proposed methodology

The development of the guideline will follow three phases: (1) Information gleaning, (2) development of prototype and (3) Delphi techniques and developing of guidelines (Figure 1).

The importance of involving proven methods, which are both inclusive and participatory, was considered as essential for developing a socially acceptable FASD guideline for South Africa.

Figure 1: A proposed methodological approach for the development of guidelines for FASD policy.

Phase 1: Information gleaning

In the first phase, we aim to obtain information from various sources to enable us to develop a prototype guideline. To this end, we plan to conduct a document review on FASD policies. The documents review will consider policies developed internationally (with focus on South Africa) and the systematic review will focus on interventions globally and with focus on South Africa. In addition, we will identify relevant policy makers and service providers in the Departments of Health, Education and Social Development and engage with them through in-depth interviews and focus group discussions to obtain information on existing policies and interventions on FASD in South Africa. This phase will be conducted in the following steps.

Step 1: Document review of policy on FASD globally and in South Africa

The researchers will review available relevant documents such as policy documents and guidelines on FASD. The aim of this review is to explore the availability and content of existing guideline and polices on FASD. Government websites of low income, middle income and high-income countries, websites of various non-governmental organisations and databases will be searched for these documents. All the policies or guidelines that focused on any aspect of FASD will be included. The narrative analysis will be used to analyse the relevant documents identified. The information that will be obtained will inform the development of a prototype guideline. Following the document review, a systematic review will be undertaken.

STEP 2: Systematic review of interventions for FASD globally and in South Africa.

The aim of the systematic review will be to systematically locate, appraise and synthesize intervention on FASD. The systematic review will consider the following review question: "What are the interventions that are available on FASD globally and in South Africa?" In conducting this review, the researchers will follow nine steps of a systematic review [36]. These steps include formulating review question, defining search strategy, establishing inclusion criteria and exclusion criteria, choosing a method for the review, conducting methodological quality and critical appraisal, extracting data, analyzing and synthesizing data, and writing a report.

What are the interventions that are available on FASD globally and in South Africa? To answer this question, databases will be searched for articles discussing or evaluating

interventions for the management of FASD. These databases include, Ebsco Host (Academic Search Complete, ERIC, SoINDEX, Health Source: Nursing/Academic Edition, CINAHL and PsycARTICLES), Science Direct, Springer Links, Wiley Online Library, JSTOR, and SAGE Journals, LANCET, Pubmed, Cochrane Library, Sabinet, NEXUS and BioMed Central Journal will be searched and retrieved for the period covering 1973 to 2016 using intervention and FASD as search terms. Furthermore, the references of the retrieved article will be used to search for more articles. The titles and abstracts of the articles will be screened using inclusion/exclusion criteria and duplicates will be removed. After screening, the remaining articles will be read and their methodological quality evaluated to establish their inclusion in the systematic review. In addition, the included articles will be appraised using a critical appraisal sheet. Narrative synthesis will be used for data analysis. Narrative synthesis can be described as a descriptive written summary of included studies and their findings [36]. The data extraction tool will consist of four distinctive sections namely: the general description, a methodological appraisal, the content intervention, and the analysis of the results. The information that will be obtained will inform the development of a prototype guideline.

STEP 3: Exploratory qualitative research

An explorative qualitative study will then be conducted. This is meant to allow the perspectives of policy makers and service providers on policies and interventions for FASD to be explored and described. Glenton, Lewin and Norris [37] reported that evidence from the qualitative research can be used to develop the guideline. Policy makers and service providers from the Department of Education, Department of Health and Department of Social Development will be interviewed as part of the processes of developing guidelines that will inform policy on FASD. A policy marker is defined as an administrator involved or supposed

to be involved in the area of policy formulation and programme monitoring on FASD and related disabilities. A service provider is defined as a staff involved or supposed to be involved in providing services and interventions on FASD and related disabilities. These groups of individuals are deemed knowledgeable to provide relevant information for developing the guidelines. Purposive sampling will be employed in selecting participants based on the following criteria: policy makers with about five years' experience in policy on FASD or related disabilities and service providers in primary health care, hospital, schools, social development and NPOs with about five years of experience in providing interventions and services to individuals with FASD. The researchers intend to sample three policy makers from each department and conduct three focus group discussions in each department with an average of 6-8 participants. In addition, for the focus group discussions, the researchers will use service providers who are available in one facility as it may be difficult to combine two facilities together. This is due to nature of their jobs and this was discovered during the feasibility study. In addition, data saturation may also define the sample.

The first set of data will be collected through in-depth interviews with policy makers using an interview guide to explore the available policies and their contents on FASD. This could provide information based on the perspectives of the policy makers. The second set of data will be collected through focus group discussions with selected service providers using a discussion schedule to explore available interventions, services and policies in practice. Focus group discussions can give rich information and contributions from participants as they contribute in moderated discussions. An audio recorder will be used to capture information from the participants, notes will also be taken during the interview and the interview recordings will be transcribed for data analysis. The rigour in the qualitative study which is

trustworthiness will be established through credibility, transferability, dependability, conformability and reflexivity [38].

The data will be analysed using thematic content analysis technique [39]. The researchers will use Creswell's data analysis spiral [40]. This process involves reading and memoing, then, describe, classify, and interpret the data according to themes and the categories, and finally visualize and represent the themes and categories. An independent coder will also be used to ensure trustworthiness.

Phase 2: Development of prototype

In the second phase, we will aggregate the information from data collected in phase one and two to develop a prototype guideline. In this phase, information obtained from document review, systematic review, and in-depth and focus group interviews will be used to develop a prototype guideline for FASD policy. This will involve conflating the information to draft a prototype. The researchers will engage and agree on consensus interpretations of the findings of the previous phases and these will then be used to develop the prototype guideline. The researchers adopted this approach because it has been applicable in the guideline development process as applied by other researchers in developing guidelines for various issues [41–43].

Phase 3: Delphi techniques and developing of guidelines

In the third phase, we will identify the relevant policy makers and health service providers and key health policy advocates and engaged with them through a Delphi technique to modify the prototype to develop obtain a refined guideline. These individuals will be mixed. That is, part of them may be those that have been participated in the qualitative research. This is because anecdotal information has it that these policy makers are not many and it may be

impossible to get a different set of participants. The process of guideline development is expected to be evolving, and the researchers have included plans to follow through a thorough process and procedures as resource and time would permit to get a consensus on aspects of the policy. The use of the Delphi approach is recommended by WHO Handbook for guideline development [32, 33, 37].

The Delphi technique is a structured communication technique initially developed as a systematic, interactive forecasting method, which relies on a panel of experts [44]. There is no agreement on sample size. It is common for these experts to answer questionnaires in two or more rounds [45]. The researcher gives an anonymous summary of the experts' predictions from the previous round, and the reasons they provided for their judgments. Experts are encouraged to revise, subsequently, their earlier answers in the light of the replies of other members of their panels. This process narrows the wide range of answers and converges the group answers towards correct or an agreed answer [46]. However, the process is stopped after a predefined stop criterion and the mean or median scores of the final rounds determine the results [44]. In this study, the developed prototype will be used to engage in Delphi technique with experts on FASD in order to develop the proposed guidelines. The researchers will start by identifying the experts and contacting them to explain the research. The researchers will then purposefully select 10 experts using the following criteria: Top management decision makers; policy makers; FASD service providers and FASD researchers with about 5years of experience relevant to FASD policies, services, intervention or research [47]. These experts will be contacted through email, as well as a telephone for them to contribute to the development of the guideline.

The researchers will use the prototype with these experts soliciting their opinions on what should be included or excluded in the proposed guidelines for policies. The data generated will be analysed and these will be used to modify the prototype, which will be used as an instrument for the second round. The experts will be asked to review the items summarised by the researchers based on the information provided in round one and they will be asked to rate or rank- order items to establish priorities among items and to reach consensus. The outcome will be presented to the experts. The information generated in the second round will then be analysed and the information gathered will be used to refine the prototype to obtain the guidelines.

Discussion

The essence of the proposed guideline is to inform policy development on foetal alcohol spectrum disorder. A guideline as earlier described in this protocol is a document that contains recommendations on health interventions for clinical or public health [32, 33]. It makes evidence-based recommendations for averting and treating certain conditions, improving health, managing medicines, planning of services for the community, development of an intervention for enhancing population health and to deliver social care for people [34]. In addition, it promotes individualised and integrated care, help service providers and recipients, and other stakeholders to make informed decisions. Furthermore, it serves as a response to a condition that needs urgent attention [32–35], and a statement, recommendation or best practice created to give a framework for policy implementation [35].

During the feasibility study of this research, it was gathered by the researcher from a non-governmental organisation (foetal alcohol related research) a leading organisation in conducting prevalence studies and rolling out prevention programme on FASD. The Chief

Executive Officer said that there are no guidelines and policies are limited to facilitate effective interventions on FASD. It was also gathered from the administrators of various departments of government in the Western Cape Province that alcohol drinking is a huge problem and FASD is one of the negative outcomes. These administrators pointed out that various efforts have been made, ranging from awareness programme to funding of NGOs to response to FASD, but these efforts are not being coordinated because there are no specific guidelines or polices on FASD. Service providers were also consulted, they highlighted that there are no specific services offered to people living with FASD. They mentioned that there are no specific guidelines to guide services and interventions though they sometimes refer an individual for proper diagnosis if they present signs of FASD. They also provide health education on alcohol to pregnant women. However, this is not specifically targeting at FASD. These gaps necessitate the conduct of this proposed research for the development of guideline that will enhance the development of coordinated policy on FASD.

In developing the guideline protocol, the researchers adapted WHO Handbook for guideline development [32, 33, 37]. The reason for adaptation is that some parts of the process are not feasible and some are beyond the scope of this project. The process includes WHO department decides to produce a guideline; discussion of required elements with Guideline Review Committee (GRC) secretariat; planning, scoping, needs assessment; Guideline Development Group (GDG) formation; key question formulation (PICO questions); planning clearance submission; Guideline Review Committee (GRC) approval for development; evidence retrieval / systematic review; evidence quality assessment / GRADE; development of recommendations; Writing, external review, editing; director's executive clearance; final GRC approval; ADG executive clearance; layout, proofread, publish; dissemination, implementation, evaluation; and updating [32, 33, 37].

The strength of using this process is that it has been used to develop various guidelines for interventions to combat diseases and improve services. The recommendations from the guidelines are rooted in a comprehensive and objective assessment of the available evidence and a clear pathway on how recommendations are generated by whom and on what. The demerit of this process is that it requires a lot of resources, time and experts. The limitation of this study might be that the researcher will not include the individuals with FASD and their families directly in the study. Another limitation is that the study is restricted to the Western Cape region of South Africa, which might affect the generalizability of the study findings.

It has been reported that there are no guidelines for determining consensus, sample size and sampling techniques when using the Delphi approach. This because these aspects of the Delphi technique require a lot of time and participants' commitment, consequently, dropout is likely to happen, which leads to low response rate and a delay in data analysis between rounds [48, 49]. To mitigate the impact of the potential challenges, the researchers decided to adapt the sample size of to a manageable number to favour in-depth engagement and discursive approach with fewer experts, rather than having a large sample of experts with less engagement with the guideline development process.

Conclusion

This protocol proposes a systematic procedure to develop a guideline that will inform the development and improvement of policy on FASD. This study will, enhance coordinated management of alcohol-related disorders, improve services to individuals with FASD, facilitates the development or improvement of guidelines, and policies.

Declarations

Ethics approval and consent to participate

Ethical research requires the protection of all participants in the study. This study will be in accordance with the ethical guidelines and principals prescribed by the University of the Western Cape. Ethical clearance has been obtained from the University of the Western Cape Senate Higher Degree Committee and permission has been being requested by the stakeholders from different organizations and governmental departments. Consent will be obtained from all the participants.

Consent for publication

Not applicable

Availability of data and material

Not applicable

Competing interests

The authors declare that there is no competing interest.

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Authors' contributions

BOA and AMB conceived and conceptualised the study and the paper. BOA designed and wrote the first draft of the manuscript. FCM redesigned the manuscript, contributed to the development of the paper and provided comments to improve the manuscript. KJO contributed to the development of the paper and provided comments to improve the manuscript. All authors read and approved the final manuscript

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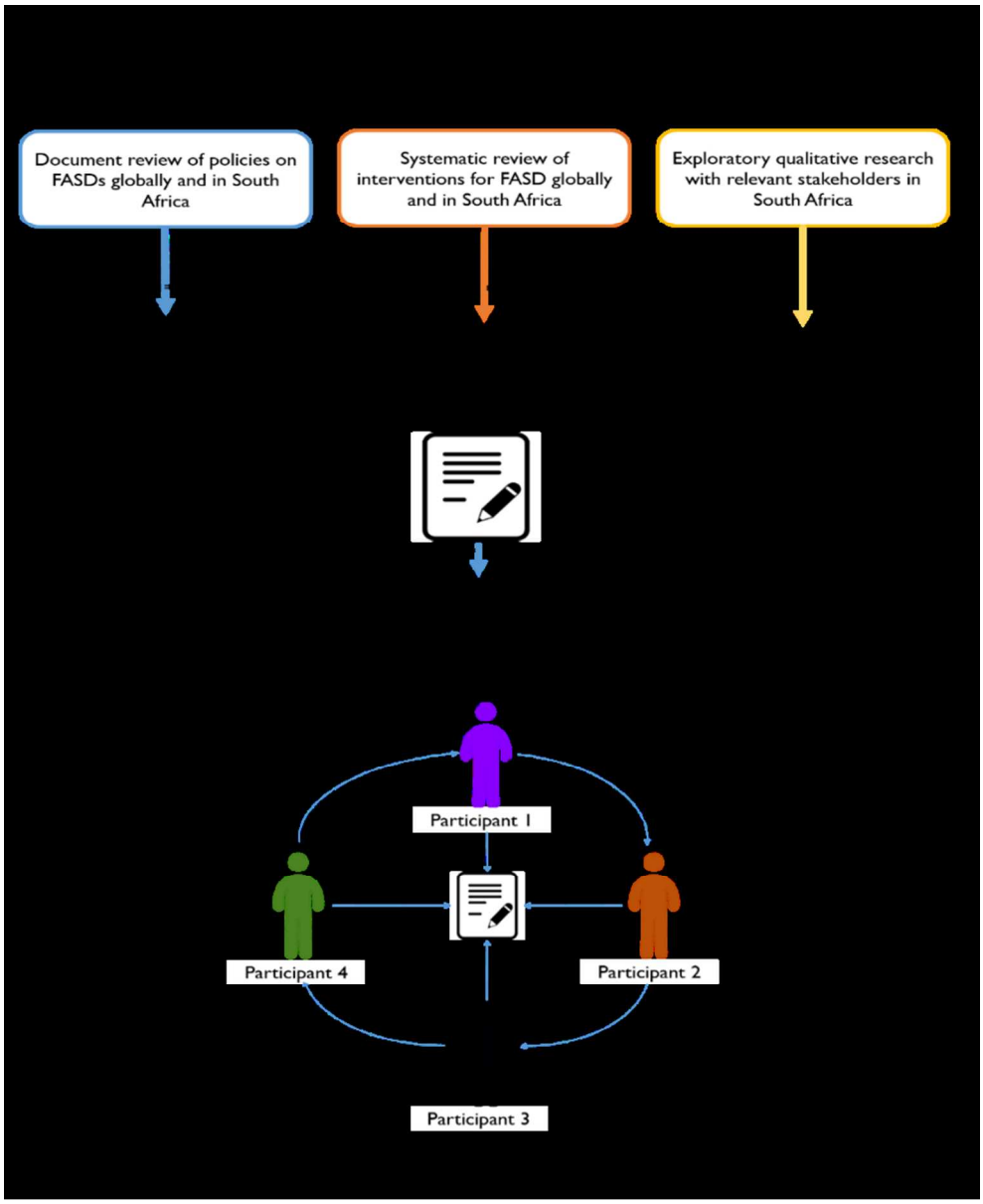
Figures

Figure 1: A proposed methodological approach for the development of guidelines for FASD policy.



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A Modified Delphi study towards developing a guideline to inform policy on Foetal Alcohol Spectrum Disorder in South Africa: A study protocol

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A Modified Delphi study towards developing a guideline to inform policy on Foetal Alcohol Spectrum Disorder in South Africa: A study protocol

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ABSTRACT

Introduction: Maternal alcohol consumption during pregnancy can result in mental and physical birth defects in individuals. These birth defects are usually described as Foetal Alcohol Spectrum Disorder (FASD). With an estimated of 183- 259 per 1000 children born with FASD, South Africa is identified to have the highest prevalence of FASD in the world. Nevertheless, there is a lack of relevant policies, guidelines, and interventions addressing the issues around FASD. This protocol outlines a proposed process for developing a guideline to inform policies on FASD.

Methods and Analysis: This process will have three phases. Phase one will be carried out in three steps; we plan to conduct a document review of available policies on the prevention, and management of FASD and update the existing systematic review on FASD interventions. The aim of the two reviews is to explore the availability and content of existing policies and global interventions on FASD. In addition, we will conduct two exploratory qualitative studies to obtain the perspectives of various stakeholders on the existing or possible guidelines and policies for the management of FASD and available interventions and services. In phase two, we will aggregate the findings of the previous phase to develop a prototype guideline. In phase three, using the developed prototype, we will apply the Delphi approach with experts on FASD, soliciting their opinions on the nature and content of the proposed guidelines for policies. The information gathered will be used to modify the prototype to formulate a policy guideline on FASD. Data will be analysed using thematic analysis and narrative synthesis.

Ethics and Dissemination: Ethical clearance has been obtained from the ethics committee of the University and governmental departments. The findings will be disseminated through publications and the guideline will be submitted to relevant departments.

Strengths and limitations of this study

- The study proposes the use of multiple sources for data collection toward developing a guideline to inform policy on Foetal Alcohol Syndrome Disorders.
- It also proposes a proven methodology – The Delphi technique to develop the guideline.
- A potential limitation to the study is that the study will not include people with FASD;
- This study is expected to inform policy on FASD nationally; however, it will be conducted in only one province out of nine provinces of South Africa.

Introduction

Foetal alcohol spectrum disorders (FASDs) refer to an array of birth disorders produced by foetal exposure to alcohol during pregnancy. FASDs include foetal alcohol syndrome (FAS), partial FAS, alcohol-related neurodevelopment disorders (ARND) and alcohol-related birth defects (ARBD). FAS is identified as the most serious anomalies among FASDs [1]. The word "foetal alcohol syndrome" was first used in 1973. This was when the negative effects were medically recognized [2, 3]. The word "fetal alcohol spectrum disorder" was not used until 2002 at a conference organized by the United States of America (USA) National Organization on Fetal Alcohol Syndrome (NOFAS). The new term was used to describe the range of outcomes that may occur as a result of prenatal alcohol exposure [4].

FASD may lead to primary and secondary disabilities. Primary disabilities are those that the child is born with [5], resulting in damages to the brain domains that are responsible for physical motor skills, sensory processing skills, cognition, communication skills, academic achievement, memory skills, executive functioning and abstract reasoning, attention, and adaptive skills [6, 7]. It can also result in physical abnormalities such as ante- and post-natal

growth retardation, eye and ear malformations, mouth and jaw deformation, skeletal defects, organ pathology, sensory deficits, and impaired immune system [1].

Secondary disabilities relate to those disabilities that developed because of lack of timely and appropriate interventions to primary disabilities. These include: fatigue, frustration, anxiety, fearfulness, rigid, resistant, argumentative behavior, becoming overwhelmed, shut down (withdrawn), a poor self-concept, feelings of failure, low self-esteem, isolation, acting out, aggression, family and/or school problems, depression and other mental health problems, trouble with the law, drug and alcohol problems, problems with employment, and homelessness [1, 8].

A systematic review conducted to identify maternal risk factor for FASD that showed the demographic (socioeconomic status, employment status, educational status, marital status, religion, living area, income and age), psychiatric and neuropsychological (psychiatric comorbidity, identified stressful, physical aggression and sexual abuse) played significant roles [9]. The review also showed that family, social (family lifestyle, drinking habit, alcohol use, illegal drugs use and tobacco use) and pattern of alcohol consumption could predispose of woman to having babies with FASDs [9].

In South Africa (SA), the national prevalence of FASD ranges from 29 to 290 per 1 000 live births [10]. The prevalence of FASD has also been reported in various regions such as in the Northern and Western Cape Provinces. The focus on these two regions is related to the high prevalence of FASD. In the Northern Cape Province, an estimated 88 per 1,000 of first grade pupils were reported to have FASD in 2008 [11]. In 2015, although the prevalence had dropped at 63.9 per 1,000 in grade one pupils, the prevalence was still relatively high [12]. In

the Western Cape Province, the prevalence of FASD among primary school pupils was estimated at 89.2/1,000 in 2007[13]. By 2013, the prevalence of FASD among first grade pupils had doubled (135.1 to 207.5 per 1,000) [14]. In 2015, the prevalence of FASD among first grade pupils recorded another increase (170 to 233 per 1,000) in the Western Cape [15].

Prevention, care and support for persons affected by FAS and other alcohol-related disorders are not being implemented, particular in developing countries [16]. Peadon et al. and Reid et al. [17, 18] reported the lack of good quality study studies and limited strong evidence for specific interventions in managing FASD. The authors, therefore, advocated for interventions targeting the specific clinical and neuropsychological deficits usually seen in individuals with FASD.

Background

South Africa is reported to have escalating levels of alcohol consumption [19], particularly in the informal settlements in the Northern and Western Cape Provinces [20–24]. The prevalence of alcohol use among pregnant women in the Cape Metropole, Western Cape Province of South Africa is estimated at 19.6% and in poor communities of the Western and Northern Cape provinces of South Africa, FASD (FAS/PFAS) is endemic [13, 25]. Maternal alcohol drinking during pregnancy, especially in rural and informal settlements, has led FASD becoming an increasing concern in South Africa.

Generally, FASD is relatively unknown among health professionals and has not been given adequate consideration in policies, programmes, and interventions [26]. This is probably because FASD is not yet formalised as a medical diagnosis, even though it is now over 40 years that it was first diagnosed [26]. In spite of this, efforts toward developing guidelines for

the diagnoses [27, 28] and management [29] of FASD have been taken toward formalising FASD as a public health problem. Because FASD is preventable, there have been efforts to advocate for the implementation of various prevention programme and strategies for its prevention. These strategies advocated for include adequately resourced alcohol control services, support for pregnant women, and a sustainable commitment from communities, service providers and the government in addressing alcohol-related health and psychological problems [30, 31]. For these strategies to be effective and sustainable, a governing policy and coordinated efforts are required.

Globally, there is inadequate policy and guidelines for the management and prevention of FASD. A review on FASD in Africa reported gaps in policy and service implementation [32]. In South Africa, only two of the national policy documents namely, the National Human Genetics Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disabilities (2001) and the National Drug Master Plan (2007) scanned during a situation analysis by Rendall-Mkosi et al. [33] used the term FAS and none of them mentioned FASD . In addition, the South African Guidelines for Maternity Care (2002) only specifies that maternal alcohol, as well as tobacco and other substances use behaviours should be taken into consideration when taking a medical history of a woman [33]. This reflects the extent to which issues related to FASD are being considered in policy.

Furthermore, current national hospital-based birth defects surveillance system developed by the National Department of Health excludes FASD [33]. This surveillance system mainly considers birth defects that can be clinically diagnosed at birth or few days after birth; which can only be identified if a woman is hospitalised after birth. The exclusion of FASD in the birth defects surveillance system is believed to be as a result of the difficulty in diagnosing

FASD at birth as most of its features are rarely manifested at birth and it requires a multidisciplinary team to arrive at a definitive diagnosis [33]. The above gap speaks to the need to develop relevant policies and guidelines to identify FASD and in order to guide strategic implementation of appropriate interventions to address FASD in South Africa.

In 2001, the Western Cape Provincial executive council acknowledged FAS as a provincial health priority for the years 2001 and 2002 [34, 35]. This led to the establishment of the Western Cape Provincial FAS reference and working group (now called FASD task team). The FASD Task Team of South Africa comprises of members from the Department of health, the Department of Social Services, the Department of Education, the Medical Research Council, the University of Stellenbosch, the University of Cape Town, NGOs such as Foundation for Alcohol Related Research (FARR), Dopstop and Pebbles project. It was headed by the provincial Maternal, Child and Women's Health Deputy Director [34, 35]. The aims of the FASD task team is to raise awareness on FASD, create strategies for prevention and, share skills and information [34, 35].

The FASD task team has been successful in the development of FAS training manual for health care workers, educators, school psychologists, social workers and other professionals working with individuals with FASD, their families and caregivers. In addition, the FASD task team has designed posters for creating awareness on FASD, organising special events to improve public awareness on International FASD Days, organising FASD training workshop and, the development of a website and Facebook page on FASD [34, 35].

The above development is an important and commendable step in developing the capacity to support the prevention and management of FASD at the provincial level. Nevertheless, a

coordinated effort to facilitate the prevention and management of FASD at the national level is required. To this end, we are proposing a national FASD guideline that will inform policy on coordinated and multi-sectoral response to FASD in South Africa. In this protocol, we propose systematic steps toward developing a guideline to inform policy for FASD prevention and management.

Research objectives

To achieve the aim, we developed the following objectives:

- To review policies and their contents on FASD internationally and in South Africa;
- To explore and describe the content of FASD policies from the perspective of policy makers;
- To provide update on the interventions on FASD internationally and in South Africa;
- To explore and describe the perspectives of service providers involved in the implementation of identified FASD intervention activities.
- To apply the Delphi technique to guide the development of guidelines for an integrated policy on FASD for the South African context.

Understanding policy guidelines

According to the World Health Organization, a guideline is a document that contains recommendations on health interventions for clinical or public health [36, 37]. It is also described as a document that makes evidence-based recommendations for averting and treating certain conditions, improving health, managing medicines, planning of services for the community, development of an intervention for enhancing population health and to

deliver social care for people [38]. Guidelines promote individualised and integrated care, help service providers and recipients, and other stakeholders to make informed decisions and serve as a response to a condition that needs urgent attention [36–39]. Therefore, a guideline is considered as a set of systematically developed statements, recommendation or best practice to give a framework for policy implementation [39]. As applies to this protocol, we considered a guideline as a document that contains recommendations that will inform the development of integrated policy on FASD.

Proposed methodology

The development of the guideline will follow three phases: (1) Information gleaning, (2) development of prototype and (3) Delphi techniques and developing of guidelines (Figure 1).

The importance of involving proven methods, which are both inclusive and participatory, was considered as essential for developing a socially acceptable FASD guideline for South Africa.

Figure 1: A proposed methodological approach for the development of guidelines for FASD policy.

Phase 1: Information gleaning

In the first phase, we aim to obtain information from various sources to enable us to develop a prototype guideline. To this end, we plan to conduct a document review on FASD policies. The documents review will consider policies developed internationally (with focus on South Africa) and the systematic review will focus on interventions globally and with focus on South Africa. In addition, we will identify relevant policy makers and service providers in the Departments of Health, Education and Social Development and engage with them through in-

depth interviews and focus group discussions to obtain information on existing policies and interventions on FASD in South Africa. This phase will be conducted in the following steps.

Step 1: Document review of policy on FASD globally and in South Africa

The researchers will review available relevant documents such as policy documents and guidelines on FASD. The aim of this review is to explore the availability and content of existing guideline and polices on FASD. Government websites of low income, middle income and high-income countries, websites of various non-governmental organisations and databases will be searched for these documents. All the policies or guidelines that focused on any aspect of FASD will be included. The narrative analysis will be used to analyse the relevant documents identified. The information that will be obtained will inform the development of a prototype guideline. Following the document review, a systematic review will be undertaken.

STEP 2: Systematic review of interventions for FASD globally and in South Africa.

The purpose of the systematic review will be to update the current most recent systematic review on FASD "Fetal Alcohol Spectrum Disorder Interventions across the Life Span" [18] to include interventions for prevention. The aim of the systematic review will be to systematically locate, appraise and synthesize intervention on FASD. The systematic review will consider the following review question: "What are the prevention and management interventions that are available on FASD globally and in South Africa?" In conducting this review, the researchers will follow nine steps of a systematic review [36]. These steps include formulating review question, defining search strategy, establishing inclusion criteria and

exclusion criteria, choosing a method for the review, conducting methodological quality and critical appraisal, extracting data, analysing and synthesizing data, and writing a report.

What are the interventions that are available on FASD globally and in South Africa? To answer this question, databases will be searched for articles discussing or evaluating interventions for the management of FASD. These databases include, Ebsco Host (Academic Search Complete, ERIC, SoINDEX, Health Source: Nursing/Academic Edition, CINAHL and PsycARTICLES), Science Direct, Springer Links, Wiley Online Library, JSTOR, and SAGE Journals, LANCET, Pubmed, Cochrane Library, Sabinet, NEXUS and BioMed Central Journal will be searched and retrieved for the period covering 1973 to 2016 using intervention and FASD as search terms. Furthermore, the references of the retrieved article will be used to search for more articles. The titles and abstracts of the articles will be screened using inclusion/exclusion criteria and duplicates will be removed. After screening, the remaining articles will be read and their methodological quality evaluated to establish their inclusion in the systematic review. In addition, the included articles will be appraised using a critical appraisal sheet. Narrative synthesis will be used for data analysis. Narrative synthesis can be described as a descriptive written summary of included studies and their findings [40]. The data extraction tool will consist of four distinctive sections namely: the general description, a methodological appraisal, the content intervention, and the analysis of the results. The information that will be obtained will inform the development of a prototype guideline.

STEP 3: Exploratory qualitative research

Two exploratory qualitative studies will be conducted. The first qualitative study will be aimed at exploring the availability of guidelines/policies for the prevention and management

of FASD within the South African health system and the need to develop a policy for the prevention and management of FASD. This study will target the policy makers from three relevant departments: Department of Education, Department of Health and Department of Social Development. A policy marker is defined as an administrator involved or supposed to be involved in the area of policy formulation and programme monitoring on FASD and related disabilities.

The second qualitative study will be aimed at identifying existing interventions and guideline/policy statements that are being used by the service providers for the prevention and management of FASD. A service provider is defined as a staff involved or supposed to be involved in providing services and interventions on FASD and related disabilities. These include paediatricians, nurses, social workers, and occupational therapist. Policy makers and service providers from the Department of Education, Department of Health and Department of Social Development will be interviewed as part of the processes of developing guidelines that will inform policy on FASD. These groups of individuals are deemed knowledgeable to provide relevant information toward developing FASD guidelines.

Purposive sampling will be employed in selecting participants based on the following criteria: policy makers with about five years' experience in policy on FASD or related disabilities and service providers in primary health care, hospital, schools, social development and NPOs with about five years of experience in providing interventions and services to individuals with FASD. The researchers intend to sample three policy makers from each department and conduct three focus group discussions in each department with an average of 6-8 participants. Data saturation will be used to determine the sample sizes in the two studies.

The first set of data will be collected through in-depth interviews with policy makers using an interview guide to explore the available policies and their contents on FASD. This could provide information based on the perspectives of the policy makers. The second set of data will be collected through focus group discussions with selected service providers using a discussion schedule to explore available interventions, services and policies in practice. Focus group discussions can give rich information and contributions from participants as they contribute in moderated discussions. An audio recorder will be used to capture information from the participants. Notes will also be taken during the interview and the interview recordings will be transcribed for data analysis. The rigour in the qualitative study, which is trustworthiness will be established through credibility, transferability, dependability, conformability and reflexivity [41].

The data will be analysed using thematic content analysis technique [42]. Both inductive – making interpretations from the raw data – and deductive – using a framework – analytic approaches will be used to analyses the data [43]. The framework will be developed from the prevention and management aspects of FASD. The researchers will use Creswell's data analysis spiral [44]. This process involves reading and memoing, then, describe, classify, and interpret the data according to themes and the categories, and finally visualize and represent the themes and categories. An independent coder will also be used to ensure trustworthiness.

Phase 2: Development of prototype

In the second phase, we will aggregate the information from data collected in phase one and two to develop a prototype guideline. In this phase, information obtained from document review, systematic review, and in-depth and focus group interviews will be used to develop a prototype guideline for FASD policy. This will involve conflating the information to draft a

prototype. The researchers will engage and agree on consensus interpretations of the findings of the previous phases and these will then be used to develop the prototype guideline. The researchers adopted this approach because it has been applicable in the guideline development process as applied by other researchers in developing guidelines for various issues [45–47].

Phase 3: Delphi techniques and developing of guidelines

In the third phase, we will identify the relevant policy makers and health service providers and key health policy advocates and engaged with them through a Delphi technique to modify the prototype to develop obtain a refined guideline. These individuals will be mixed. That is, part of them may be those that have been participated in the qualitative research. This is because anecdotal information has it that these policy makers are not many and it may be impossible to get a different set of participants. The process of guideline development is expected to be evolving, and the researchers have included plans to follow through a thorough process and procedures as resource and time would permit to get a consensus on aspects of the policy. The use of the Delphi approach is recommended by the World Health Organization (WHO) Handbook for guideline development [36, 37, 48].

The Delphi technique is a structured communication technique initially developed as a systematic, interactive forecasting method, which relies on a panel of experts [49]. There is no agreement on sample size. It is common for these experts to answer questionnaires in two or more rounds [50]. The researcher gives an anonymous summary of the experts' predictions from the previous round, and the reasons they provided for their judgments. Experts are encouraged to revise, subsequently, their earlier answers in the light of the replies of other members of their panels. This process narrows the wide range of answers and converges the

group answers towards correct or an agreed answer [51]. However, the process is stopped after a predefined stop criterion and the mean or median scores of the final rounds determine the results [49].

In this study, the developed prototype will be used to engage in Delphi technique with experts on FASD in order to develop the proposed guidelines. The researchers will start by identifying the experts and contacting them to explain the research. The researchers will then purposefully select 30 experts using the following criteria: Top management decision makers; policy makers; FASD service providers and FASD researchers with about five years of experience relevant to FASD policies, services, intervention or research [52]. These experts will be contacted through email, as well as a telephone for them to contribute to the development of the guideline.

The researchers will use the prototype with these experts soliciting their opinions on what should be included or excluded in the proposed guidelines for policies. Descriptions of prevention and management interventions identified in phase 1 will used to design Likert statements to evaluate the perceptions of the experts on prevention modalities, screening components, and management methods. Participants will be asked to rate their agreement with each statement on a 5-point Likert scale, which will range from 'strongly agree' to 'strongly disagree', and a response option of 'no comment' will be provided to enable participants to indicate that a statement was outside their area of expertise [53].

The data generated will be analysed and these will be used to modify the prototype, which will be used as an instrument for the second round. The experts will be asked to review the items summarised by the researchers based on the information provided in round one and

they will be asked to rate or rank- order items to establish priorities among items and to reach consensus. The outcome will be presented to the experts. The information generated in the second round will then be analysed and the information gathered will be used to refine the prototype to obtain the guidelines.

Discussion

The essence of the proposed guideline is to inform policy development on foetal alcohol spectrum disorder. While having informal discussions with the head of a non-governmental organisation, a leading organisation in conducting prevalence studies and rolling out prevention programme on FASD, we understood that guidelines and policies are limited to facilitate effective interventions on FASD. The administrators of the departments of Education, Health, and Social Development in the Western Cape Province also revealed that alcohol drinking is a huge problem and FASD is one of the negative outcomes. These administrators pointed out that various efforts have been made, ranging from awareness programme to funding of NGOs to response to FASD, but these efforts are not being coordinated because there are no specific guidelines or polices on FASD. Service providers were also consulted, they highlighted that there are no specific services offered to people living with FASD. They mentioned that there are no specific guidelines to guide services and interventions though they sometimes refer an individual for proper diagnosis if they present signs of FASD. They also provide health education on alcohol to pregnant women. However, this is not specifically targeting at FASD. These gaps necessitate the conduct of this proposed research for the development of guideline that will enhance the development of coordinated policy on FASD.

In developing the guideline protocol, the researchers will adapt the WHO Handbook for guideline development [36, 37, 48]. The reason for adaptation is that some parts of the process are not feasible and some are beyond the scope of this project.

- The research team decides to produce a guideline for the prevention and management of FASD based-on informal discussions with relevant stakeholders on the need of an FASD policy;
- Discussion of required elements among the researchers planning, scoping, needs assessment;
- Key question formulation (PICO questions);
- Planning clearance submission; University Ethics review committee approval for the development; evidence retrieval / systematic review;
- Conducting the various studies, evidence quality assessment; development of recommendations;
- Writing, external review, editing; final approval; proofread, publish; dissemination, [36, 37, 48].

The strength of using this process is that it has been used to develop various guidelines for interventions to combat diseases and improve services. The recommendations from the guidelines are rooted in a comprehensive and objective assessment of the available evidence and a clear pathway on how recommendations are generated by whom and on what. The demerit of this process is that it requires a lot of resources, time and experts. The limitation of this study might be that the researcher will not include the individuals with FASD and their families directly in the study. Another limitation is that the study is restricted to the Western Cape region of South Africa, which might affect the generalizability of the study findings.

It has been reported that there are no guidelines for determining consensus, sample size and sampling techniques when using the Delphi approach. This because these aspects of the Delphi technique require a lot of time and participants' commitment, consequently, dropout is likely to happen, which leads to low response rate and a delay in data analysis between rounds [54, 55]. To mitigate the impact of the potential challenges, the researchers decided to adapt the sample size to a manageable number to favour in-depth engagement and discursive approach with fewer experts, rather than having a large sample of experts with less engagement with the guideline development process.

The finalised guideline will be distributed to all the participants in the study and to the departments of Health, Education and Social Development as well as to the relevant NGOs working on FASD. The guideline will also be published in a peer review journal to add to the literature of FASD.

Ethics and Dissemination

The approval for the study was obtained from the research ethics committee of the University of the Western Cape (BM/16/4/4) and further approvals were obtained from the Western Cape Department of Education (20161212-6937), Department of Health (WC_2016RP29_862), and Social Development (12/1/2/4). Before the interviews and the FGDs, the study aims and objectives we will explain to the potential participants and they will be provided with an information sheet written in English explaining their roles. The potential participants will be requested to sign a consent form if they agreed to participant in the study. All participants for the interviews and FGDs will be asked to sign a consent form All information obtained during the study will be kept strictly confidential in a computer with password known only to the researchers in this study.

Declarations

Consent for publication

Not applicable

Availability of data and material

Not applicable

Competing interests

The authors declare that there is no competing interest.

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Authors' contributions

BOA and AMB conceived and conceptualised the study and the paper. BOA designed and wrote the first draft of the manuscript. FCM redesigned the manuscript, contributed to the development of the paper and provided comments to improve the manuscript. KJO contributed to the development of the paper and provided comments to improve the manuscript. All authors read and approved the final manuscript

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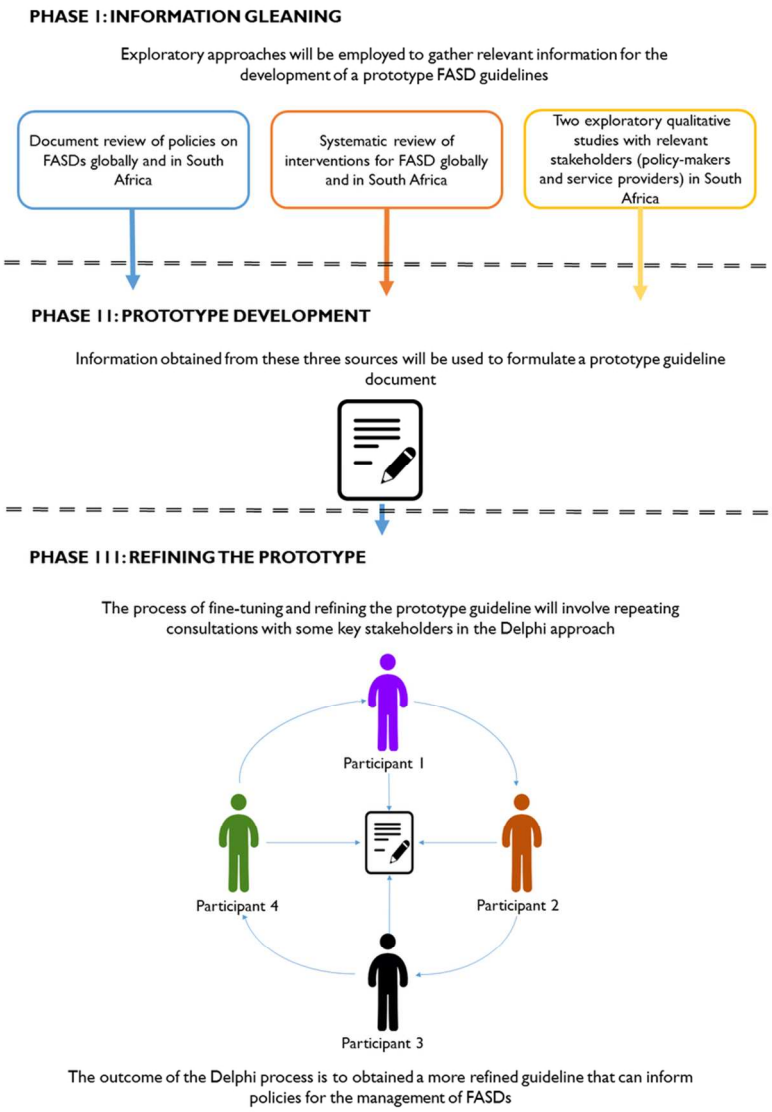
Figures

Figure 1: A proposed methodological approach for the development of guidelines for FASD policy.



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A Modified Delphi study towards developing a guideline to inform policy on Foetal Alcohol Spectrum Disorders in South Africa: A study protocol

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ABSTRACT

Introduction: Maternal alcohol consumption during pregnancy can result in mental and physical birth defects in individuals. These birth defects are usually described as Foetal Alcohol Spectrum Disorders (FASDs). With an estimated 183- 259 per 1000 children born with FASDs, South Africa is identified to have the highest prevalence of FASDs in the world. Nevertheless, there is a lack of appropriate policies, guidelines, and interventions addressing the issues around FASDs. This protocol outlines a proposed process for developing a guideline to inform policies on FASDs.

Methods and Analysis: This process will have three phases. Phase one will be carried out in three steps; we plan to conduct a document review of available policies on the prevention, and management of FASDs and update the existing systematic review on FASDs interventions. The aim of the two reviews is to explore the availability and content of existing policies and global interventions on FASDs. In addition, we will conduct two exploratory qualitative studies to obtain the perspectives of various stakeholders on the existing or possible guidelines and policies for the management of FASDs and available interventions and services. In phase two, we will aggregate the findings of the previous phase to develop a prototype guideline. In phase three, using the developed prototype, we will apply the Delphi approach with experts on FASDs, soliciting their opinions on the nature and content of the proposed guidelines for policies. The information gathered will be used to modify the prototype to formulate a policy guideline on FASDs. Data will be analysed using thematic analysis and narrative synthesis.

Ethics and Dissemination: Ethical clearance has been obtained from the ethics committee of the University and governmental departments. The findings will be disseminated through publications and the guideline will be submitted to relevant departments.

Strengths and limitations of this study

- The study proposes the use of multiple sources for data collection toward developing a guideline to inform policy on FASDs.
- It also proposes a proven methodology – The Delphi technique to develop the guideline.
- A potential limitation of the study is that the study will not include individuals with FASDs.
- This study is expected to inform policy on FASDs nationally; however, it will be conducted in only one Province out of nine Provinces of South Africa.

Introduction

Foetal alcohol spectrum disorders (FASDs) refer to an array of birth disorders related to foetal exposure to alcohol during pregnancy. FASDs are classified under four broad groups; foetal alcohol syndrome (FAS), partial FAS, alcohol-related neurodevelopment disorders (ARND) and alcohol-related birth defects (ARBD) [1]. Of these four groups, FAS is identified as having the most serious anomalies [2, 3].

FASDs may lead to primary and secondary disabilities [4]. Primary disabilities are those that the child is born with [5], usually associated with damages to the brain domains that are responsible for physical motor skills, sensory processing skills, cognition, communication skills, academic achievement, memory skills, executive functioning and abstract reasoning, attention and adaptive skills [6, 7]. The brain damages could also result in physical abnormalities such as ante- and post-natal growth retardation, eye and ear malformations, mouth and jaw deformation, skeletal defects, organ pathology, sensory deficits and impaired immune system [1].

Secondary disabilities relate to those disabilities that develop because of untimely and inappropriate interventions to primary disabilities – consequences of unaddressed primary disabilities. These include fatigue, frustration, anxiety, fearfulness, rigid, resistant, argumentative behaviour, becoming overwhelmed, shut down (withdrawn), a poor self-

concept, feelings of failure, low self-esteem, isolation, acting out, aggression, family and/or school problems, depression and other mental health problems, trouble with the law, drug and alcohol problems, problems with employment, and homelessness [8].

A systematic review conducted to identify maternal risk factors for FASDs showed that the maternal demographic (socioeconomic status, employment status, educational status, marital status, religion, living area, income, and age) and psychiatric - including neuropsychological (psychiatric comorbidity, identified stressful, physical aggression, and sexual abuse) factors play significant roles [9]. The review also showed that family, social (family lifestyle, drinking habit, alcohol use, illegal drugs use, and tobacco use) and pattern of alcohol consumption could predispose women to have babies with FASDs [9].

In South Africa, the national prevalence of FASD ranges from 29 to 290 per 1 000 live births [10]. Some Provinces such as the Northern and Western Cape Provinces of South Africa have particularly registered high FASDs prevalence. In the Northern Cape Province, an estimated 88 per 1,000 of first-grade pupils were reported to have FASDs in 2008 [11]. In 2015, although the prevalence had dropped to 63.9 per 1,000, the prevalence was still relatively high [12]. In the Western Cape Province, the prevalence of FASDs among primary school pupils was estimated at 89.2 per 1,000 in 2007 [13]. By 2013, the prevalence of FASDs among first-grade pupils had doubled (135.1 to 207.5 per 1,000) [14], and another increase (170 to 233 per 1,000) was recorded in 2015 [15].

There is evidence that strategies designed to prevent FASDs and to care and support persons affected by FASDs are not effectively implemented, particularly in developing countries [16]. Peadon et al. [17] and Reid et al. [18] also reported the lack of good quality studies and

limited strong evidence for specific interventions in managing FASDs. The authors, therefore, advocated for interventions targeting the specific clinical and neuropsychological deficits usually seen in individuals with FASDs.

Background

South Africa is reportedly having escalating levels of alcohol consumption [19], particularly in the informal settlements of the Northern and Western Cape Provinces [20–23]. The prevalence of alcohol use among pregnant women in the Cape Metropole of the Western Cape Province of South Africa is estimated at 19.6% and in poor communities of the Western and Northern Cape provinces of South Africa, FASDs are endemic [13, 24]. Maternal alcohol drinking during pregnancy, especially in rural and informal settlements, has led FASDs becoming an increasing concern in South Africa.

Generally, FASDs are relatively unknown among health professionals and have not been given adequate consideration in policies, programmes, and interventions [25]. This is probably because FASDs remain unformalised medical diagnoses since they were first identified and described 40 years earlier [25]. Despite the apparent lack of interest in FASDs, efforts toward developing guidelines for the diagnoses [26, 27] and management [28] of FASDs have been made, especially toward recognising FASDs as a public health problem. Because FASDs are preventable, there have also been advocacy efforts for the implementation of various prevention programmes. These advocacy strategies call for the availability of adequately resourced alcohol control services, support for pregnant women, and a sustainable commitment from communities, service providers, and the government in addressing alcohol-related health and psychological problems [29, 30]. Nevertheless, for

these strategies to be effective and sustainable, a governing policy and coordinated efforts are required.

Globally, there is inadequate policy and guidelines for the management and prevention of FASDs. A review on FASDs in Africa reported gaps in policy and service implementation [31]. In South Africa, only two of the national policy documents namely, the National Human Genetics Policy Guidelines for the Management and Prevention of Genetic Disorders, Birth Defects and Disabilities (2001) and the National Drug Master Plan (2007) mentioned the term FAS and none mentioned FASD [32]. In addition, the South African Guidelines for Maternity Care (2002) only suggests that maternal alcohol, tobacco, and other substances use behaviours should be taken into consideration when taking a medical history of a pregnant woman [32]. The limited occurrence of issues related to FASDs in policy documents offers a reflection on the extent to which FASDs are (not) being considered in policy.

Current national hospital-based birth defects surveillance system developed by the National Department of Health also excludes some FASDs [32]. This surveillance system mainly considers birth defects that can be clinically diagnosed at birth or few days after birth, which can only be identified during post-natal hospitalisation. The exclusion of some FASDs in the birth defects surveillance system is because of the difficulty of diagnosing certain FASDs at birth as most of their features are rarely manifested at birth. In addition, it requires a multidisciplinary team to arrive at a definitive diagnosis of FASDs [32]. The above gaps speak to the need to develop relevant policies and guidelines to identify FASDs and to guide strategic implementation of appropriate interventions to address FASDs in South Africa.

In 2001, the Western Cape Provincial executive council acknowledged FASDs as a provincial health priority for the years 2001 and 2002 [33, 34]. This led to the establishment of the Western Cape Provincial FAS reference and working group (now called FASD task team). The FASD Task Team of South Africa comprises of members from the Department of Health, the Department of Social Services, the Department of Education, the Medical Research Council, the University of Stellenbosch, the University of Cape Town, and Non-profit organisations (NPOs) [Foundation for Alcohol Related Research (FARR), Dopstop, and Pebbles project]. The task team is headed by the Provincial Maternal, Child and Women's Health Deputy Director [33, 34]. The aims of the FASD task team is to raise awareness on FASDs, create strategies for the prevention of FASDs, and share skills and information [33, 34].

The FASD task team has been successful in developing FASDs training manuals for health care workers, educators, school psychologists, social workers, and other professionals working with individuals with FASDs, their families, and caregivers. In addition, the FASD task team has designed posters for creating awareness on FASDs, organising special events to improve public awareness on International FASDs days, organising FASDs training workshops and, developing a website and Facebook page on FASD [33, 34].

The above development is an important and commendable step in developing the capacity to support the prevention and management of FASDs at the provincial level. Nevertheless, a coordinated effort to facilitate the prevention and management of FASDs at the national level is required. To this end, we are proposing a national FASDs guideline that will inform policy on coordinated and multi-sectoral response to FASDs in South Africa. In this protocol, we

propose a systematic approach toward developing a guideline to inform policy for the prevention and management FASDs in South Africa.

Research objectives

To develop a guideline for the prevention and management of FASDs in South Africa, we aimed to achieve the following objectives:

- To review policies and their contents on FASDs in South Africa;
- To explore and describe the content of FASDs policies from the perspective of policy makers;
- To provide an update on the interventions on FASDs internationally and in South Africa;
- To explore and describe the perspectives of service providers involved in the implementation of identified FASDs intervention activities;
- To apply the Delphi technique to guide the development of guidelines for an integrated policy on FASDs for the South African context.

Understanding policy guidelines

According to the World Health Organization, a guideline is a document that contains recommendations for clinical or public health interventions [35, 36]. It is also described as a document that makes evidence-based recommendations for averting and treating certain conditions, improving health, managing medicines, planning of services for the community, development of an intervention for enhancing population health, and to deliver social care for people [37]. Thus, guidelines promote individualised and integrated care, help service providers and recipients, and other stakeholders to make informed decisions, and serve as a response to a condition that needs urgent attention [35–38]. Therefore, a guideline is

considered as a set of systematically developed statements, recommendation or best practice to give a framework for policy implementation [38]. As applies to this protocol, we considered a guideline as a document that contains recommendations that will inform the development of integrated policy on FASDs.

Adopted Approach

In developing the guideline for FASDs, we adopted the WHO's approach (steps) as stipulated in the WHO's Handbook for guideline development [35, 36].

1. We agreed to design a guideline for the prevention and management of FASDs based-on informal discussions with relevant stakeholders on the need for a policy governing the prevention and management efforts of FASDs;
2. We planned on scoping of the literature and conducting a needs assessment;
3. After gathering preliminary information, the team formulated key PICO questions [39] Population – individuals with FASDs,
   Intervention – a guideline to inform policy,
   Comparison – (Not applicable) and
   Outcomes – a guideline for the prevention and management of FASDs;
4. After agreeing on the need to design the guideline to inform policy on the prevention and management of FASDs, the next step involved designing the project protocol (which is reported in this paper);
5. The team applied for Ethics clearance for the project from the University of the Western Cape – this has been obtained;
6. The next step will entail conducting the various studies and exercises outlined in this protocol;

7. The team will develop and prepare the guideline to inform policy on FASDs based on the findings of the various studies and the outcomes of the various exercises;
8. The team will disseminate the developed guideline through various channels including having feedback meetings with the various stakeholders and sharing our findings, publish the guideline in a peer-reviewed journal, present the findings in journal club meeting organised at the provincial department of Health and at other national and international conferences [35, 36].

Some of the steps outlined in the WHO's handbook were adapted to our context. The reason for the adaptation is that some parts of the process are not applicable to this study and other parts are beyond the scope of this project.

Proposed methodology

The development of the guideline will follow three phases: (1) Information gleaning, (2) development of prototype and (3) Delphi techniques and developing of guidelines (Figure 1). The importance of involving proven methods, which are both inclusive and participatory, was considered as essential for developing a socially acceptable FASDs guideline for South Africa.

Figure 1: A proposed methodological approach for the development of guidelines for FASDs policy.

Phase 1: Information gleaning

In phase 1, three steps will be undertaken with the purpose of obtaining diverse information to develop a prototype guideline. To this end, we plan to conduct a document review on

FASDs policies and a systematic review on FASDs interventions. The document review will consider policies developed South Africa and the systematic review will focus on interventions globally with a focus on South Africa. In addition, we will identify relevant policy makers and service providers from the Departments of Health, Education, and Social Development and engage with them through in-depth interviews and focus group discussions to obtain information on existing policies and interventions on FASDs in South Africa. This information-gleaning phase will be conducted in the following steps.

Step 1: Document review of policy on FASD in South Africa

The researchers will review available relevant documents such as policy documents and guidelines on FASDs. The aim of this review is to explore the availability and content of existing guidelines and policies on FASDs. Websites (government and NPOs) and databases will be searched for these documents. All the policies or guidelines that focused on any aspect of FASDs will be included. A narrative analysis will be used to analyse the relevant documents identified. The information that will be obtained will inform the development of a prototype guideline. Following the document review, a systematic review will be undertaken.

STEP 2: Systematic review of interventions for FASDs globally and in South Africa.

The purpose of the systematic review will be to update the current most recent systematic review on FASDs "Foetal Alcohol Spectrum Disorder Interventions across the Life Span" [18] to include interventions for prevention. The aim of the systematic review will be to systematically locate, appraise, and synthesise intervention on FASDs. The systematic review will consider the following review question: "What are the prevention and management interventions that are available on FASDs globally and in South Africa?" In conducting this

review, the researchers will follow nine steps of a systematic review [40]. These steps include formulating review question, defining search strategy, establishing inclusion criteria and exclusion criteria, choosing a method for the review, conducting methodological quality and critical appraisal, extracting data, analysing and synthesizing data, and writing a report.

What are the interventions that are available on FASDs globally and in South Africa? To answer this question, databases will be searched for articles discussing or evaluating interventions for the prevention and management of FASDs. These databases include, Ebsco Host (Academic Search Complete, ERIC, SoINDEX, Health Source: Nursing/Academic Edition, CINAHL and PsycARTICLES), Science Direct, Springer Links, Wiley Online Library, JSTOR, and SAGE Journals, Pubmed, Cochrane Library, Sabinet, and NEXUS will be searched and retrieved for the period covering 2007 to 2017 using intervention and FASDs as search terms. Furthermore, the references of the retrieved article will be used to search for more articles.

The titles and abstracts of the articles will be screened using inclusion/exclusion criteria and duplicates will be removed. After screening, the articles that meet the inclusion criteria will be read and their methodological quality evaluated. In addition, the included articles will be appraised using a critical appraisal sheet. Narrative synthesis will be used for data analysis. Narrative synthesis can be described as a descriptive written summary of included studies and their findings [41]. The data extraction tool will consist of four distinct sections namely: the general description, a methodological appraisal, the content intervention, and the analysis of the results. While conducting and reporting the reviews, we will follow the guidelines for systematic reviews.

STEP 3: Exploratory qualitative research

Two exploratory qualitative studies will be conducted. The first qualitative study will explore the availability of guidelines/policies for the prevention and management of FASDs within the South African health system and the need to develop a policy for the prevention and management of FASDs. This study will target the policy makers (administrators involved in policy formulation and programme monitoring on FASDs and related disabilities) from three relevant departments: Department of Education, Department of Health, and Department of Social Development.

The second qualitative study will be aimed at identifying existing interventions and guideline/policy statements that are being used by the service providers (individuals involved in providing services and interventions on FASDs and related disabilities) for the prevention and management of FASDs. Relevant service providers include paediatricians, nurses, social workers, and occupational therapist. Policy makers and service providers from the Department of Education, Department of Health, and Department of Social Development will be interviewed as part of the processes of developing guidelines that will inform policy on FASDs. These groups of individuals are deemed knowledgeable to provide relevant information toward developing FASDs guidelines.

Purposive sampling will be employed in selecting participants. For the policy makers, we will include those individuals with about five years' experience in policy on FASDs or related disabilities. Similarly, service providers working in primary health care, hospital, schools, social development and NPOs with at least five years of experience in providing interventions and services to individuals with FASDs will be potential participants. The researchers intend to sample three policy makers from each department and conduct three

focus group discussions in each department with an average of 6-8 participants. Data saturation will be used to determine the sample sizes in the two qualitative studies.

In the first qualitative study, data will be collected through in-depth interviews with policy makers using an interview guide. Open-ended questions will be used to start the interviews and follow-up questions will be used to probe for additional explanations when required. The study participants will be asked various questions on available policies on FASDs, the coordination of FASDs interventions and relevant aspects of FASDs to inform the FASDs guidelines. Each interview will last for about 30- 60 minutes and each interview will be audio-recorded with the permission from the participants.

Data, in the second qualitative study, will be collected through focus group discussions with selected service providers using a discussion schedule to explore available interventions, services, and policies in practice. Focus groups use unstructured discussion subjects involving small ranges of subjects (6-10). It is cost-effective and time-efficient and participants are allowed to express their viewpoints in detail. Discussants are often motivated to contribute more in the presence of their co-participants [42]. In addition, focus group discussions can give rich information as discussants contribute in moderated discussions. An audio recorder will be used to capture information from the discussants. Notes will also be taken during the interview and the interview recordings will be transcribed for data analysis.

The data will be analysed using thematic content analysis technique [43]. Both inductive – making interpretations of the raw data – and deductive – using a framework – analytic approaches will be used to analyse the data [44]. The framework will be developed from the prevention and management aspects of FASDs. The researchers will use Creswell's data

analysis spiral [45]. This process involves reading and memoing, then, describing, classifying, and interpreting the data according to themes and categories, and finally visualising and representing the themes and categories. An independent coder will also be used to ensure trustworthiness.

The rigour in the qualitative studies, which is trustworthiness will be established through credibility, transferability, dependability, conformability, and reflexivity [46]. In addition, the reporting of the qualitative findings will follow the consolidated criteria for reporting qualitative research (COREQ) [47].

Phase 2: Development of prototype

In the second phase, information obtained from the document review, systematic review and in-depth and focus group interviews will be conflated to develop a prototype guideline for a FASDs policy. The researchers will engage and agree on consensus interpretations of the findings to develop the prototype guideline. We selected this approach because it has been successfully applied in the guideline development process by other researchers in developing guidelines for various issues [48–50].

Phase 3: Delphi techniques and developing of guidelines

In the third phase, we will apply the Delphi approach to refine the developed prototype. The use of the Delphi approach is recommended by the World Health Organization (WHO) Handbook for guideline development [35, 36]. The Delphi technique is a structured communication technique initially developed as a systematic, interactive forecasting method, which relies on a panel of experts [51]. It is common for experts to answer questionnaires in two or more rounds [52]. The researcher gives an anonymous summary of the experts'

predictions from the previous round, and the reasons they provided for their judgments. Experts are encouraged to revise their earlier responses in the light of the replies of other members in the forum. This process narrows the wide range of answers and converges the group answers towards an agreed answer [53]. The process is stopped after a predefined stop criterion and the mean or median scores of the final rounds determine the results [51].

There is no agreement on the sample size of participants in the Delphi discussion forum. We will identify relevant policy makers, health service providers and health policy advocates to participate in a Delphi contribution forum to refine the prototype. Some of the Delphi participants will include those who participated in any of the two qualitative studies. This is because experts on FASDs issues are few and it may be challenging to get a complete set of different participants.

We will start by identifying the experts and contacting them to explain the research. We will then purposefully select 30 experts using the following criteria: Top management decision makers; policy makers; FASDs service providers and FASDs researchers with about five years of experience relevant to FASDs policies, services, and intervention or research [54]. These experts will be contacted via email and telephone for their contributions.

The developed prototype will be used to engage in a Delphi technique with experts on FASDs to develop the proposed guidelines. Descriptions of prevention and management interventions identified in phase 1 will be used to design Likert statements to evaluate the perceptions of the experts on prevention modalities, screening components, and management methods. We will solicit their opinions on what should be included or excluded in the proposed guidelines for policies. Participants will be asked to rate their agreement with each

statement on a 5-point Likert scale, which will range from 'strongly agree' to 'strongly disagree', and a response option of 'no comment' will be provided to enable participants to indicate that a statement was outside their area of expertise [55].

The data generated will be analysed and these will be used to modify the prototype, which will be used as an instrument for the second round. The experts will be asked to review the items summarised by the researchers based on the information provided in round one and they will be asked to rate or rank - order items to establish priorities among items and to reach consensus. The information generated in the second round will then be analysed to refine the prototype to obtain the guidelines. The process of guideline development is expected to be evolving, thus, we plan to follow a thorough process and procedures as resource and time would permit.

Patient and public involvement

Patient and public will not be involved in the study. The findings of the study will be disseminated to the study participants through emails.

Discussion

The essence of the proposed guideline is to inform policy development on FASDs. While having informal discussions with the head of a non-governmental organisation, a leading organisation in conducting prevalence studies and rolling out prevention programme on FASDs, we understood that focused guidelines and policies to facilitate effective interventions on FASDs are non-existent. The administrators of the departments of Education, Health, and Social Development in the Western Cape Province also revealed that

drinking alcohol remains a major concern and FASDs are among the negative outcomes of irresponsible drinking practices.

These administrators pointed out that various efforts have been made, ranging from awareness programmes to funding of NGOs to response to FASDs, but these efforts are not being coordinated because there are no specific guidelines or policies on FASDs. Service providers consulted also raised the similar concerns. Nevertheless, the service provides suggested that generic programmes are in place to provide health education on alcohol use during pregnancy to pregnant women and that they would sometimes refer suspected cases of FASDs for proper diagnosis. Of course, these practices are not specifically targeting the problem of FASDs or addressing the issues around FASDs in an intended manner. The lack of focused attention accorded to FASDs prompted the conception to develop guidelines that will enhance the prevention and management of FASDs.

The strength of using the approach outlined in this protocol lies in the fact that it has been successfully used to develop various guidelines for interventions to combat diseases and improve services. In addition, the recommendations from the guidelines will be rooted in a comprehensive and objective assessment of the available evidence and a clear pathway on how recommendations are generated. The demerit of this process is that it requires a lot of resources, time and expertise. The limitation of this study might be that the researchers will not include the individuals with FASDs and their families directly in the study. Another limitation is that the study is restricted to the Western Cape region of South Africa, which might affect the generalizability of the study findings.

It has been reported that there are no guidelines for determining consensus, sample size, and sampling techniques when using the Delphi approach. This is because these aspects of the Delphi technique require a lot of time and participants' commitment, consequently, dropout is likely to happen, which leads to low response rate and a delay in data analysis between rounds [56, 57]. To mitigate the impact of these potential challenges, the researchers have decided to adapt the sample size to a manageable number to favour the in-depth engagement and discursive approach with fewer experts, rather than having a large sample of experts with less engagement with the guideline development process.

The finalised guideline will be distributed to all the participants in the study and to the departments of Health, Education, and Social Development as well as to the relevant NGOs working on FASDs. The guideline will also be published in a peer review journal to add to the literature on FASDs.

Ethics and Dissemination

The approval for the study was obtained from the research ethics committee of the University of the Western Cape (BM/16/4/4) and further approvals were obtained from the Western Cape Department of Education (20161212-6937), Department of Health (WC_2016RP29_862), and Social Development (12/1/2/4). Before the interviews and the FGDs, the study aims and objectives we will explain to the potential participants and they will be provided with an information sheet written in English explaining their roles. The potential participants will be requested to sign a consent form if they agreed to participant in the study. All participants for the interviews and FGDs will be asked to sign a consent form All information obtained during the study will be kept strictly confidential in a computer with a password known only to the researchers in this study.

Declarations

Consent for publication

Not applicable

Availability of data and material

Not applicable

Competing interests

The authors declare that there is no competing interest.

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Authors' contributions

BOA and AMB conceived and conceptualised the study and the paper. BOA designed and wrote the first draft of the manuscript. FCM redesigned the manuscript, contributed to the development of the paper and provided comments to improve the manuscript. KJO contributed to the development of the paper and provided comments to improve the manuscript. All authors read and approved the final manuscript

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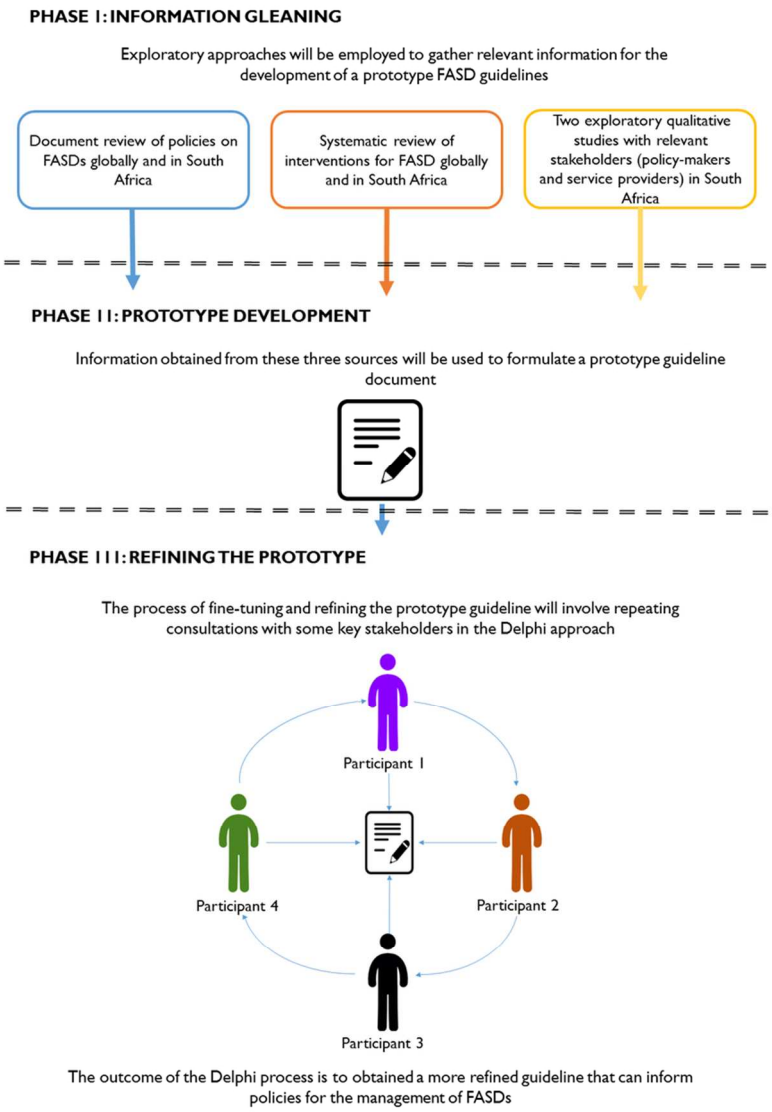
Figures

Figure 1: A proposed methodological approach for the development of guidelines for FASDs policy.



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