









STUDY PROTOCOL

Development and assessment of the feasibility of a Zika family support programme: a study protocol [version 1; peer review: 2 approved]

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Abstract

The Zika virus outbreak in Brazil in 2015 affected thousands of people. Zika is now known to cause congenital malformations leading to impairments and developmental delays in affected children, including Congenital Zika Syndrome (CZS). Children with CZS have complex care needs. Caregivers require significant levels of support to meet these needs, and there are large gaps in healthcare services.



This study aims to develop, pilot and assess the feasibility and scalability of a community-based Family Support Programme for caregivers of children with CZS. The programme is adapted from the Getting to Know Cerebral Palsy (GTKCP) programme for the context of CZS in Brazil. GTKCP is a 10-session programme held with 6-10 caregivers in the local community. It includes practical, educational, peer-support and psychosocial aspects, which aim to improve confidence and capacity to care for a child with CP, and quality of life and empowerment of caregivers.

The research project contains four components:


1. Ascertaining need for the caregiver programme: a mixed-methods approach that included two literature reviews, interviews with key stakeholders in country, and incorporation of findings from the Social and Economic Impact of Zika study.
2. Adapting GTKCP for the context of CZS and Brazil: undertaken with guidance from technical experts.
3. Pilot testing the intervention: deliver the 10-session programme to one group of caregivers of children with CZS in Rio de Janeiro and another in Greater Salvador.
4. Update the manual through fast-track learning from participant and facilitator feedback. Assessing the feasibility of the intervention for scale up: deliver the updated programme to two groups each in Rio de Janeiro and Greater Salvador, and evaluate the acceptability, demand, implementation, practicality, adaptation, integration,

Open Peer Review

Reviewer Status  

	Invited Reviewers	
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expansion, and limited efficacy, through questionnaires, direct observation, semi-structured interviews and cost calculation. The project has ethics approval in both the UK and Brazil.

Keywords

Zika, disability, microcephaly, early intervention, congenital Zika syndrome, family, caregiver, Brazil

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Introduction

Although several outbreaks of the Zika virus have occurred across the world since it was first identified in the 1950s, it was not until the sudden increase in numbers of cases were recorded in Brazil in 2015 that Zika started to garner significant international attention¹. Some 6 months after the first Brazilian cases, a spike in cases of microcephaly was noted and caused Brazil and the international health community to question whether there was a link between Zika and birth anomalies². Prior to that outbreak, Zika was thought to be relatively innocuous, causing few hospitalisations and was not believed to be fatal³. Zika was declared a Public Health Emergency of International Concern by the WHO in February 2016. This declaration was lifted in November 2016 with the recognition that Zika is likely to remain an ongoing challenge for the public health community and that “a robust longer-term technical mechanism was now required to manage the global response”⁴. The link between Zika and congenital conditions has now been proven⁵ and the group of impairments and developmental delays in affected infants and young children is known as “Congenital Zika Syndrome” (CZS)⁶.

CZS is a recognizable pattern of structural anomalies and functional impairments secondary to central and perhaps peripheral nervous system damage. In describing CZS, Moore *et al.*⁶ suggested five unique features:

- Severe microcephaly with partially collapsed skull;
- Thin cerebral cortices with subcortical calcifications;
- Macular scarring and focal pigmentary retinal mottling;
- Congenital contractures; and
- Marked early hypertonia and symptoms of extrapyramidal involvement.

More recent evidence suggests that not all children with developmental issues relating to Zika have CZS or present with microcephaly at birth. Some are born with a normal head circumference and go on to develop microcephaly later, and others show evidence of the other features without microcephaly⁷. Microcephaly, therefore may be the tip of the iceberg with regards the wider array of clinical and developmental features⁸. Consequently, the approximately 3000 cases of microcephaly with confirmed Zika infection may dramatically underestimate the true scale of the condition in Brazil. Additionally, it is not known what health conditions or impairments may yet manifest in young children as they continue their development. The oldest group of children from the Brazilian outbreak are 3 years old as of January 2019. For the purposes of this paper and project, we used CZS to describe any child with impairments that can be directly attributed to Zika.

What is known is that children with CZS are likely to require ongoing support and care from the health, social, education and other sectors as they grow and develop^{9–13}. Families experience heavy burdens of care raising children with similar neuro-developmental disabilities, such as cerebral palsy (CP)¹⁴. A

few studies have already shown high levels of anxiety amongst mothers of children with CZS^{15,16}.

In spite of the disabling impact of Zika, only a fraction of funding and research has been focussed towards meeting the care and support needs of children with CZS and their families. Perhaps reasonably, most programmes have targeted comprehending the nature of the virus in order to work towards a vaccine and future prevention¹⁷. Meanwhile, the health services for children with neuro-developmental disabilities that exist in Brazil, including physiotherapy, occupational therapy, speech therapy and other therapeutic services, alongside medical services, have been overwhelmed by an upsurge in demand^{18,19}. Against a background of generally stretched services, there is also inequity in the availability of these services, particularly in non-urban areas²⁰, and the non-clinical needs are often overlooked. Families often have only sporadic, limited or unstructured education and support with respect to the care of their child. True health promotion, however, requires meeting the holistic needs of families²¹.

A similar situation of unmet healthcare needs and unsupported families is also apparent for other types of complex childhood disability, such as CP. In response to this recognised need to provide for the holistic care of families of children with complex multiple impairments the London School of Hygiene & Tropical Medicine developed a participatory caregiver group programme for children with CP called ‘Getting to Know Cerebral Palsy’(GTKCP)²² (Figure 1). The programme sought to educate and empower these caregivers to optimise their situation, quality of life and the ability for their child to maximise his/her potential to participate within society. It was developed and pilot-tested in Bangladesh.

GTKCP has since been implemented in over 25 countries. In Ghana, an evaluation showed that the programme was positively received by families and had a positive impact on both quality of life and knowledge and confidence of caregivers of children with CP, as well as the reported physical and emotional health of the child²³. The programme was originally designed for children of age 2 and over, and a recent adaptation has been piloted in Uganda for children under 2, the Early Intervention Programme (EIP), and is currently being clinically trialled²⁴.

This current study aims to develop, pilot and assess the feasibility and scalability of a community-based *Family Support Programme* for caregivers of children with CZS. The programme is adapted from GTKCP for the context of children with CZS in Brazil. The specific objectives of the research are:

1. To undertake a needs assessment for the intervention
2. To adapt GTKCP and EIP for the context of CZS in Brazil.
3. To conduct a pilot programme in two sites in Brazil for families of children with CZS.
4. To assess the feasibility of the pilot programme for potential scale up and roll out across the country and beyond.

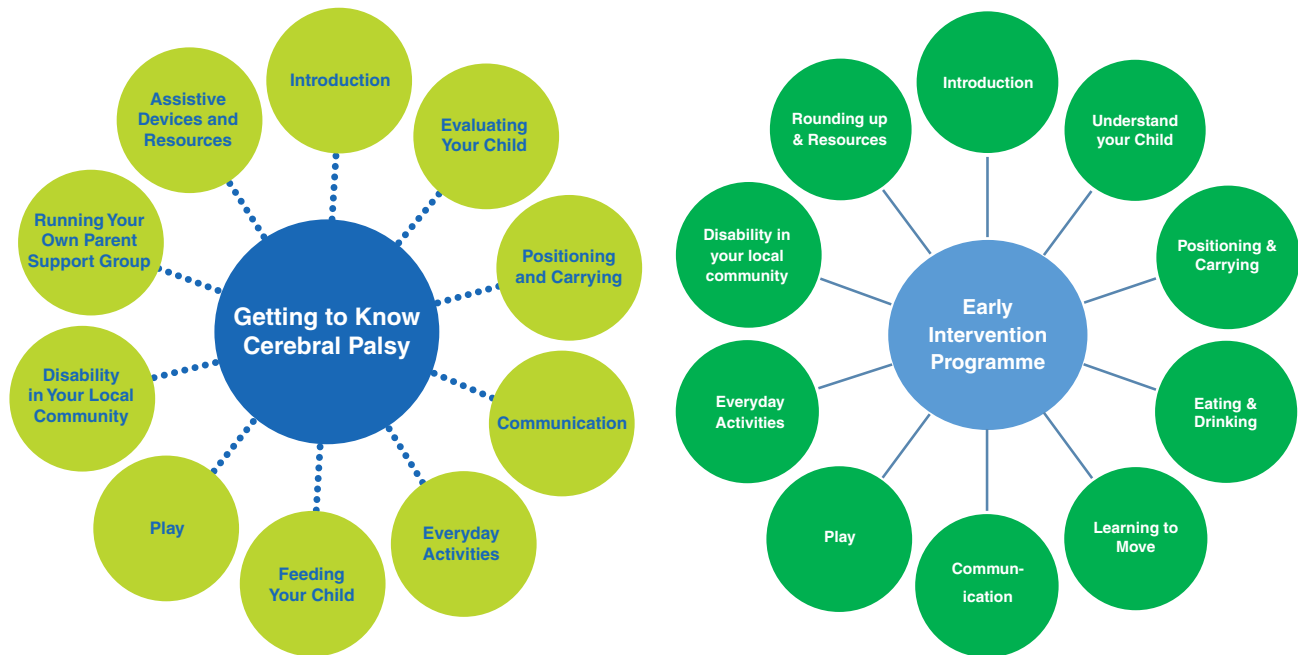


Figure 1. Structure of Getting to Know Cerebral Palsy and the Early Intervention Programme.

Protocol

Overview of study design and setting

The Family Support Programme is the implementation arm of two research initiatives undertaken by the International Centre for Evidence in Disability at the London School of Hygiene & Tropical Medicine. The sister study is a mixed methods evaluation of the social and economic impacts of CZS that took place concurrently²⁵ and data fed into the content of the Family Support Programme.

Two locations in Brazil were used to pilot the programme - Rio de Janeiro and Salvador, both of which had high numbers of Zika cases and children born with CZS⁹. The study is undertaken in partnership with Fundação Oswaldo Cruz (known as Fiocruz), the national institute for health research. In Rio de Janeiro, the partner was the National Institute of Women, Children and Adolescent Health Fernandes Figueira (IFF), part of Fiocruz, and in Salvador it was the Federal University of Bahia.

Ethics approval was acquired in both Brazil (IFF/FIOCRUZ - RJ/MS 2.183.547) and the UK (LSHTM Ethics number 13608). All participants who took part in the programme completed a consent form²⁶, relevant to their involvement in the study (e.g. survey, interview). Participants were also requested to provide consent for photographs or other media to be recorded during the group sessions, once it was explained that non-agreement to the media consent form would not impact their position in the groups.

Below we describe the methods for the four objectives:

1. To undertake a needs assessment for the intervention

A mixed-methods approach was adopted to identify the needs for a family support intervention. This approach included reviewing

emerging and associated literature, an in-country needs assessment with qualitative investigations, and incorporation of findings from the sister study²⁵.

A scoping review was undertaken in May 2017 to describe the clinical presentation of Zika-related impairments in children, including CZS and its similarities and differences with other neurodevelopmental disabilities. Studies published between October 2015 and April 2017 (i.e. since the onset of the Brazil outbreak) on CZS were identified through PubMed searchers using 'Zika', 'Microcephaly', 'Congenital Zika Syndrome' as search terms and reviewing the reference list of relevant papers. Data and evidence that contributed to information about the clinical presentation of CZS was compiled and presented to inform the programme structure, though the paper by Moore *et al.*⁶ provided a timely and comprehensive overview of the research and clinical features of CZS. These summaries were then compared to literature on the clinical presentation of other neurodevelopmental disabilities, most notably CP, guided by a paediatric neurologist.

A literature review was undertaken in summer 2017 to explore the implications for CZS and CP for support needed for affected families. Search terms included 'zika virus', 'congenital zika syndrome', 'cerebral palsy', 'family needs', 'parent needs', 'psychosocial', 'cost', economic impact' alone and in combination were used in CINAHL Plus, EMBASE, MEDLINE, PsychInfo, and PubMed with formats for search terms adapted for different databases. Eligibility criteria included any study published in peer reviewed journals that described the needs of families of children affected by CZS and/or microcephaly related to Zika, or CP (in low-middle-income countries only) and was published in English language in peer-reviewed journals between January 2000 and July 2017.

A scoping needs assessment was undertaken by members of the researcher team in Brazil in April 2017. The scoping visit involved reviewing the current context in Brazil and the project sites, in terms of structure, function and availability of health and social services that may be needed by parents, to ascertain the needs of the intervention. This assessment was undertaken by meeting with a range of clinicians in both Rio de Janeiro, Salvador and Recife, and included doctors, therapists, psychologists. Furthermore, meetings with organisations working to support families with children with CZS and informal consultations with families themselves were also undertaken. Meetings and discussions were not recorded, but annotated.

Finally, findings from the Social and Economic Impact of Zika study²⁵, funded by the Wellcome Trust, were incorporated into the needs analysis. This included data from semi-structured interviews²⁷ with families of children with CZS to ascertain their needs and the impact of CZS on their lives, specifically assessing need for psychological, social, financial and other supports. Data were provided through direct dialogue with the team and from published papers.

The team members considered evidence across these four sources to identify where the needs and service gaps were most substantial, which would need to be targeted by the intervention.

2. To adapt the GTKCP and EIP for the context of CZS in Brazil

The Family Support Programme is based on the existing structure of GTKCP²³ and the EIP²⁴, as no other relevant interventions for this target group were identified. Findings from the needs assessment, described above, were used to adapt the programme to meet the specified and identified needs of caregivers of children with CZS in Brazil and gaps in services (May–July, 2017). These adaptations were undertaken by a project team, which included individuals in Brazil and globally, with expertise in care for children with complex needs and their families, such as physiotherapists, paediatricians and social scientists. The team also included individuals who developed the GTKCP and EIP. One team member was assigned the role to lead on content development (AD). External support was sought where needs were identified that were outside of the expertise of the group (e.g. nutrition).

Further, two technical advisory groups (TAG) were established—one in Brazil and one in the UK. These groups contained members with a diverse background and experience related to Zika including researchers, health professionals and parent advocates. The role of the TAGs was to provide input and feedback during the development, review and finalisation of the programme. Most notably, the required tasks were to agree on the structure of the initial pilot programme, to review and agree on the changes made between phase 1 and 2 (see below) and review and agree on the final proposed programme after all pilot groups are concluded. The TAGs were also consulted on sample size and inclusion criteria, facilitator and researcher profiles, module structure, session frequency and other programme parameters.

The main implementation element of the programme is to deliver a series of sessions to a group of caregivers. Materials were

produced to support the programme including a manual for the facilitators providing guidance for the Family Support Programme content and structure, and images printed on durable textile for group work.

3. To conduct a pilot programme in two sites in Brazil on families of children with CZS

Once the initial adaptation of the programme was agreed by the two TAGs, it was pilot-tested within two groups in Brazil (August–November, 2017).

Eight facilitators for the caregiver groups were identified by the site coordinators (M.S. and S.F.), and included four mothers of children with CZS and four therapists (e.g. speech and language). Facilitators were enrolled in a training programme in July 2017, to be educated and capacitated on the content of the programme and aspects on delivering participatory groups. An expert consultant was used for this process, who had experience from the GTKCP programmes. The training programme lasted 5 days and included orientation to the content of the course, facilitation on adult based learning styles and practice sessions.

Subsequently, one parent group was established in Rio de Janeiro, and one in Greater Salvador. The sessions were guided by the facilitators through the structure and order of the adapted programme. Caregivers were identified using criteria agreed during the UK TAG—families of children with confirmed or suspected CZS who were residing at home (i.e. not receiving inpatient hospital care at the time of the start of the programme), who agreed to be involved in the programme and who were not participating in a conflicting group study (but could be receiving individual therapy). Participants were identified through clinical and therapy networks at the two sites and identified participants were contacted by site coordinators about joining the groups. Each group included 6–10 caregivers of children with CZS, and was held approximately weekly in the local community, for 10 sessions. Each session lasted approximately 3 hours, and a range of topics were covered (e.g. feeding, play, communication).

Researchers received pre-pilot training to review and familiarise with the questionnaires, observation and focus group procedures and interview schedules. Two researchers were assigned to each group and held focus group discussions with participants and facilitators at the end of each session to record data on a pre-designed format.

Fast-track learning was used during the pilot testing to hone and adjust the structure and content of the programme from the initial draft. Two main sources of information were utilised for Fast track learning:

- Researcher observation: researchers attended the sessions to observe and acquire feedback from participants and facilitators. Researchers observed the sessions using a checklist guide without directly intervening in the group. The completed form was sent to the coordinators (A.D. and T.S.) after each session, in English. The checklist contained information about the participatory approaches

used by facilitators, the level of interest and engagement of participants, and noted any aspects that went well or did not go well and require modification or improvement.

- End-of-session focus groups

The programme content and delivery was updated based upon the feedback received.

Facilitators and researchers received compensation for their involvement in the programme. The amount of compensation was ascertained and managed by the Fiocruz partners in Brazil based on their allocated budget. Participants did not receive any financial compensation for taking part in the programme.

4. To assess the feasibility of the pilot programme for potential scale up and roll out across the country and beyond

A further 3-day 'updating' training was undertaken in December 2017 to provide facilitators with information on the changes to the content and structure of the programme based on fast-track learning in the pilot phase. Two further parent groups were established in each setting as above, to ascertain the feasibility of the intervention, with identical procedures for fast-track learning and data collection (February–June, 2017).

The feasibility of the programme was assessed through the eight areas of focus proposed by Bowen *et al.*²⁸ for evaluating public health interventions (Table 1).

In total, four sources of data were collected to give an overall valuation of the programme and provide the appropriate information for the assessment into the feasibility. These include collecting data from participants of the programme, facilitators and other key stakeholders:

Participant data

Pre- and post-programme semi-structured questionnaires²⁹ were completed by all participants in the programme before the first session and after the final session of each group. This data was then logged into a password-protected Google Drive® document, shared with the content development lead, and discussed between the researcher and content lead within two days of submission. Questionnaires were developed in English and translated into Portuguese, and included the following items:

- Socio-demographic characteristics of the child and caregiver¹.
- Perceived unmet needs and main goals for the intervention¹.
- The PedsQL™ Family Impact Questionnaire Module³⁰ (using the official version translated to Brazilian Portuguese).
- Understanding and knowledge about the child's condition.
- Self-reported functioning of the child.
- Nutrition and feeding and drinking practices
- Subjective well-being of caregiver and child
- Review of goals achieved²
- Satisfaction with programme²

Minor adjustments to the translation and structure of the questionnaires was made by the researchers after the pilot groups to improve certain sections. Questionnaires were pilot-tested in a

¹Only asked at baseline

²Only asked at end-line

Table 1. The eight areas of focus. Adapted from Bowen *et al.*²⁸.

Area of focus	The feasibility study asks...
Acceptability	"To what extent is a new idea, program, process or measure judged as suitable, satisfying, or attractive to program deliverers? To program recipients?"
Demand	"To what extent is a new idea, program, process, or measure likely to be used (i.e., how much demand is likely to exist?)"
Implementation	"To what extent can a new idea, program, process, or measure be successfully delivered to intended participants in some defined, but not fully controlled, context?"
Practicality	"To what extent can an idea, program, process, or measure be carried out with intended participants using existing means, resources, and circumstances and without outside intervention?"
Adaptation	"To what extent does an existing idea, program, process, or measure perform when changes are made for a new format or with a different population?"
Integration	"To what extent can a new idea, program, process, or measure be integrated within an existing system?"
Expansion	To what extent can a previously tested program, process, approach, or system be expanded to provide a new program or service?
Limited efficacy	"Does the new idea, program, process, or measure show promise of being successful with the intended population, even in a highly controlled setting?"

sample of participants. Researchers administered the questionnaire to participants before commencing the first group and after completing the final group. Participants should have attended a majority of sessions (>50%) to complete the final questionnaire. Attendance of sessions was monitored through a simple registry.

The PedsQL™ Family Impact Questionnaire Module³⁰ was selected for a number of reasons. First, it contains a range of measures where we anticipated impact by the programme, for example emotional functioning, worry. Second, it has been validated in Brazilian Portuguese³¹. Third, it is also being applied in the sister study²⁵, which allowed some consistency and comparability for further analysis.

Semi-structured interviews were undertaken with two or three participants per group within 15 days of the final session of each group either in the setting of the group meetings or at the participant's home. Participants were selected at the discretion of researchers to reflect a broad range of perspectives (e.g. caregivers of children with different severities of disability, mothers and fathers). Interviews were undertaken in Portuguese by the local researchers, asking about satisfaction with and perceived impact of the groups. The interviewer recorded and transcribed the interviews.

Facilitator data

A semi structured interview of up to 30 minutes was undertaken at the location of the group sessions with each of the facilitators (total, seven) at the final session of the final group reflecting the whole process. These were undertaken by the local researchers, who recorded and transcribed the interviews.

Key stakeholder data

Semi-structured interviews were also conducted with identified key stakeholders in Brazil in April 2018. These included the two site coordinators and involved specific questions around practicality, adaptation, integration and expansion Bowen's areas of feasibility. Interviews were undertaken in English or Portuguese by either the study leads (AD/TS) or the local researchers, and were transcribed.

Other data

Cost of the sessions were assessed by analysing the budget and establishing an overall cost for delivery of the programme, and in addition the cost per participant. Training of facilitator costs were calculated and presented separately as this may not be reflective of the structure of a true training of facilitator programme if scaled up (number of facilitators, international travel etc). A costed plan for scale up was considered.

Data management and analysis

Data was sent to the research team in London by the site coordinators. Interviews were saved as word files and questionnaires in excel. All stored data was anonymized and password protected.

Data was stored by project site (Rio and Salvador) and by group number (i.e. Rio 1, Rio 2, Rio 3, Salvador 1, Salvador 2, Salvador 3). Each participant was given a unique number for pre-questionnaires, session notes and post-questionnaires. Participants interviews were not linked to their individual questionnaire responses.

Analysis of the interviews and session notes/focus groups was undertaken using NVIVO 12® software. A social scientist fluent in English and Portuguese coded the interview responses in NVIVO 12. Thematic analysis was structured around the eight areas of feasibility described by Bowen *et al.* (Table 1)²⁸ with an additional 'other' theme for information that the analyst found pertinent but did not fit into the eight feasibility themes. Analysis of the questionnaires was undertaken using Microsoft Excel, producing data on demographics of participants, change between baseline and endline in the areas described and reflection on the program.

As per Wellcome Trust data management plans, the data collected from this study will be made openly available to specific users (i.e. researchers in an academic environment) on request to the study lead (Antony Duttine) through e-mail (antony.duttine@lshtm.ac.uk). Data can be analysed only for the specific purposes compatible with the consent agreement. The data is not freely and open available since the sample size is relatively small and even though data is anonymised, there is a risk of establishing the identities of participants.

Dissemination of findings

A minimum of three additional papers are anticipated from the completion of the research: one on the needs of such an intervention, one on the feasibility analysis and one describing the whole intervention and final programme. Additional areas of potential interest which may be explored are the findings on using a mother as a facilitator and the engagement of fathers in the programme.

The subject is of interest to both the general public and the public health community given the attention that Zika gained. Therefore, it may be likely that there are opportunities for developing grey literature e.g. blog articles, media pieces regarding the work.

Opportunities for submitting abstracts and presenting the work at national (UK and Brazil) and international forums will be pursued. Dissemination events will be arranged in UK and Brazil, inviting key stakeholders.

Data availability

Underlying data

All data underlying the results are available as part of the article and no additional source data are required.

Extended data

Researchgate: Pre and post questionnaires. <https://doi.org/10.13140/RG.2.2.13700.17287>²⁹.

Researchgate: Qualitative interview questions for participants, facilitators and key informants. <https://doi.org/10.13140/RG.2.2.28380.2368627>.

Researchgate: Consent forms (interviews and questionnaires). <https://doi.org/10.13140/RG.2.2.30057.9584526>.

Extended data are available under the terms of the [Creative Commons Attribution 4.0 International license](#) (CC-BY 4.0), excluding the PEDS QL instrument which is © 1998 JW Varni, Ph.D. All rights reserved.

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Michael J. Griffiths 

Institute of Infection and Global Health, University of Liverpool, Liverpool, UK

This manuscript entitled “Development and assessment of the feasibility of a Zika family support programme: a study protocol” outlines a protocol to:

1. undertake a needs assessment on whether Brazilian caregivers of children with congenital zika syndrome may benefit from a family support programme
2. adapt two existing family support programmes , Getting to Know Cerebral Palsy (GTKCP) and Early Intervention Programme (EIP), for use among the above families
3. pilot these adapted programmes among two geographical distinct groups of caregivers in Brazil and alongside conduct an evaluation of the programme
4. re-pilot the refined adapted programme and alongside undertake a more detailed evaluation of the programme, including an assessment of the feasibility of the programme for potential scale up and roll out across the country and beyond.

I enthusiastically support the premise of the study. Such programmes need to be developed and rolled out among such patient groups and in such settings. On the whole the study protocol is well described and sufficiently detailed.

I offer the following comments to the author team:

Echoing the reviewer Michel Landry, the title could be changed to better describe the study as follows ‘Adaptation and assessment of a family support programme for use among caregivers of children with congenital zika syndrome: a study protocol’. This change would highlight that an existing programme was adapted to the target population rather than developed from scratch.

In the introduction, the initial description on the unique features of congenital Zika syndrome (CZS) as described by Moore and colleagues is probably not really necessary. The study does not recruit their target population based on these features. As the authors state, it has been reported that neurodevelopmental problems can occur in children of mothers exposed to Zika virus during pregnancy

that do not exhibit these features.

In the description of objective 2, the authors appear to 'jump' to selecting the existing GTKCP and EIP programmes. It would have been useful to describe in more detail why GTKCP and EIP programs were selected and why the authors believed these programmes were appropriate for use in the target group. For example the authors could have mapped the domains of need covered in these programs against the needs of caregivers of CZS children. This would have provided evidence to support the selection.

In the introduction, the authors could have described the range of neuro-developmental difficulties CZS children have been reported to experience in Brazil and how these difficulties overlap with other neurodevelopmental disorders, such as cerebral palsy. This comparison would have helped to orientate the readers to understand why the authors went on to choose to adapt a family support programme for caregivers of children with cerebral palsy.

As part of the description of objective two, it would be useful to have access to examples of the adapted GTKCP and EIP programmes in the extended data sets. As a minimum, a few examples of how the programmes' content or structure were adapted would be helpful.

In the description of objective four, '*To assess the feasibility of the pilot programme for potential scale up and roll out across the country*' the authors list a broad series of measures evaluated (including PedsQL, Nutrition and feeding and drinking practices). Several of these measures do not appear to relate to programme feasibility or scale-up. This objective could be written more clearly to explicitly state what measures and how the stated objective was assessed.

In the description of objective four, it would be useful to know how the researchers ensured the participants felt free to express any potential negative views of the course (or facilitators), particularly as some interviews were conducted at the programme site?
Details of the compensation given to facilitators would also be useful. Again this raises the question, how did the researchers/study design ensure compensation (e.g. if compensation was higher or lower than expected) didn't influence the facilitators' view of the programme.

At the start of the methods, the authors state 8 facilitators were trained. In objective four, they state 7 facilitators were interviewed. Details of why one facilitator was not interviewed would add completeness.

Echoing the reviewer Michel Landry, I fully support the focus on protection of personal identifiable information. However, I would also urge the authors to reconsider providing open access to the high level aggregated data. This would help provide context to the work on this protocol.

Overall this is a very useful study protocol. I very much look forward to the downstream publications.

Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Yes

Are sufficient details of the methods provided to allow replication by others?

Partly

Are the datasets clearly presented in a useable and accessible format?

Partly

Competing Interests: No competing interests were disclosed.**Reviewer Expertise:** Paediatric Neurology, Neuro-Infection, Global Health.**I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.**

Reviewer Report 20 May 2019

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**Michel D. Landry**

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This manuscript very nicely outlines a protocol that was implemented to explore the effects of Congenital Zika Syndrome (CZS) in Brazil, and the author team was inclusive of colleagues from 1 UK based institution and 2 independent institutes from Brazil. Ultimately the research team sought to describe their mixed-method approach to address the following areas/questions: is there a need for a caregiver program, and can a previously developed program (GTKCP) be adapted in Brazil for this population. Further, they sought to pilot test an adapted GTKCP, and update a manual (policies, procedures, structured and other such details I expect) related to the adapted program. The description of the protocol was detailed, and I offer the following suggestions to the author team.

1. It would seem to me that if the authors intend to adapt a previously developed program (i.e. GTKCP), it might be worthwhile to consider indicate such an ‘assessing the adaptability’ approach in the title. This would allow the reader to gain traction on the process and protocol rather quickly. It would also be picked up by others also seeking to adapt protocols in different settings.
2. Given the that CZS is mosquito-borne, and given that not all readers might be as familiar with the pathogenesis, it may be worth a very brief description of the transmission vectors.
3. While the stages of the protocol are very clear, it remains somewhat a complex interplay in time and space. The authors may wish to consider providing a flow chart/figure of the protocol (along with timing and duration) so that the reader can quickly refer back and better appreciate the methods.
4. Given that this is a student protocol submission, further details on the type and form of compensation that was provided from Fiocruz to the participants. This could be a helpful model (or at least consideration) for others to consider during their implementation of similar programs in lower resourced settings.
5. There appeared to be sufficient details on the first three objectives, but there was very little in regards to the fourth objective “Update the manual.” Please provide details on this final objective so as to make the study protocol complete.

6. While I recognize that this is a study protocol submission, I was left wishing to be able to easily access the findings. I followed the 'trail' to access all of the consents forms, and excel files, and they were all well developed in my opinion. I fully support the authors focus on protection of personal information that might be identifiable. However, I would suggest that the authors reconsider this position and provide open access to some basic information on the population that was involved in phases of the study. This high level aggregated information would provide context, and deepen the appreciation for all the work that has been inputted into this protocol. I have read that it was possible to contact Dr. Duttine (and all his information was well placed in the manuscript), but not all reader will likely do this extra step, and so I would encourage the authors to consider.

Overall, this is a well written study protocol, and I look forward to the subsequent publications on this important public and population health challenge.

Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Yes

Are sufficient details of the methods provided to allow replication by others?

Yes

Are the datasets clearly presented in a useable and accessible format?

Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Global Health, Policy and Disability.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.
