Assessing the feasibility for *Juntos*: a support programme for caregivers of children with congenital Zika syndrome in Brazil during the 2015-16 Zika epidemic

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DECLARATION OF OWN WORK

I, Antony Alan Duttine, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

ABSTRACT

Introduction

When the Zika virus emerged in Brazil in 2015, what also emerged was the previously unknown realisation that the virus causes microcephaly and other congenital abnormalities. Congenital Zika syndrome (CZS) affected thousands of children who may have lifelong impairments and disabilities. This DrPH project describes the development, piloting, and feasibility testing of a support programme for caregivers of children with CZS that took place between April 2017 and August 2018. Seven papers have been published in peer-reviewed literature with respect to the intervention and four are included in the main body of this research paper thesis: a study protocol paper; a needs analysis; an intervention description; and the final feasibility analysis. Introductory and discussion sections also elaborate on the work undertaken.

Methods

The needs analysis included a literature review, analysis of emerging evidence and a scoping visit. It identified important gaps for caregivers such as peer-to-peer support, mental health needs and education. *Juntos*, a ten-session programme led by two-co-facilitators was developed based on two pre-existing programmes for children with Cerebral Palsy – a neurodevelopmental disability that often has a similar presentation to CZS. Juntos was initially tested in two groups in Rio de Janeiro and Salvador, then adjusted based on feedback and piloted in four additional groups in the same cities. The feasibility analysis was undertaken using a public health framework based on qualitative and quantitative data.

Results

Juntos was successfully developed and piloted for 48 families. The feasibility analysis showed positive outcomes particularly in relation to acceptability, demand, and implementation. Some limited efficacy was also shown such as self-reported improvements in caregiver quality of life.

Conclusion

Despite positive initial piloting of Juntos there are many more implications to be considered if it is to be scaled up and considered a success, including policy, programme, and research actions.

ACKNOWLEDGEMENTS

There are many people I wish to thank for helping me through this DrPH, which has been one of the biggest challenges and endeavours I have ever undertaken.

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To Veronika, to Anna, and to Fernanda: without your passion and dedication, this work would never have been completed. Thank you.

To all the caregivers of children with Congenital Zika Syndrome or any other developmental disability – you are not going through this journey alone.

Last, but most importantly, to Emily who encouraged me to take on the DrPH, who has patiently supported me, loved me and guided me: انت حياتي يا حبيبتي. بحبك.

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ABBREVIATIONS

BRL	Brazilian Real (currency)
CDC	Centers for Disease Control and Prevention
CP	Cerebral Palsy
CZS	Congenital Zika Syndrome
DrPH	Doctor of Public Health
EBPHP	Evidence-Based Public Health Policy and Practice
ECD	Early Childhood Development
EIP	Early Intervention Programme
FIOCRUZ	Fundação Oswaldo Cruz
GBP	British Pound
GBS	Guillain-Barré Syndrome
GTKCP	Getting to Know Cerebral Palsy
GMFCS	Gross Motor Functioning Classification System
ICED	International Centre for Excellence on Disability
ICF	International Classification of Functioning, Disability and Health
LSHTM	London School of Hygiene and Tropical Medicine
NDD	Neurodevelopmental Disorders
NICE	National Institute for Health and Care Excellence (UK)

NMH	Noncommunicable Diseases and Mental Health (Department)
OPA	Organisational Policy Analysis
OT	Occupational Therapy or Occupational Therapist
PAHO	Pan American Health Organisation
PHC	Primary Health Care
PHEIC	Public Health Emergency of International Concern
PT	Physiotherapy or Physiotherapist
SLT	Speech and Language Therapy or Therapist
SUS	Sistema Único de Saúde (Brazilian Health System)
ToC	Theory of Change
ULMO	Understanding Leadership, Management and Organisations
WHO	World Health Organization
ZIKV	Zika Virus

OVERVIEW OF THESIS

Thesis Structure

This thesis is presented in a research paper style containing four publications which have been published in peer-reviewed journals. There are seven chapters in total:

Chapter 1 sets the context for the study. It gives an overview of the Zika epidemic in 2015-16. It then describes the neurodevelopmental consequences that were seen in some children born to mothers who had a Zika infection during pregnancy, a condition that has become to be known as congenital Zika syndrome (CZS). Focussing in on Brazil, where most cases occurred, the chapter then explores the context of the health and social system within the country, and the potential consequences for families and caregivers of children with CZS.

Chapter 2 presents the first paper, which was published in Open Wellcome Research in May 2020 [1]. It is a study protocol describing the intention and plans for conducting the research project. The research protocol is broken into four phases: ascertaining the need for a community programme for caregivers of children with CZS; adapting an existing programme, created for caregivers of children with Cerebral Palsy, for the context of CZS in Brazil; piloting the programme in two sites in Brazil; and assessing the feasibility of the pilot programmes for potential scale up.

Chapter 3 contains an overview of the Theory for Change for the project that was developed early in the process, reflected upon throughout and reviewed at its conclusion.

Chapter 4 presents the second paper, which was published in the *International Journal of Environmental Research and Public Health* in May 2020 [2]. It is a review of the need for a support programme, based on a systematic review, analysis of initial findings of a parallel study exploring the social and economic impact of Zika, and a scoping visit undertaken in April 2017.

Chapter 5 presents the third paper, which was published in *Global Health: Science and Practice* in December 2020 [3]. It provides a detailed description of the development and piloting of the family support programme - *Juntos* - across six groups in two locations (Rio de Janeiro and Salvador, Bahia) between August 2017 and June 2018.

Chapter 6 presents the fourth paper, which was published in *Open Wellcome Research* in March 2022 [4]. It focusses on an analysis of the feasibility of *Juntos* that was delivered in the six pilot groups. Feasibility is assessed across six areas: acceptability, demand, implementation, practicality, adaptation, and limited efficacy. A mixed-methods approach was used to assess the different areas of feasibility including caregiver, facilitator, and key informant interviews; baseline and endline participant questionnaires; session observations; and financial analysis of the project budget.

Chapter 7 explores the findings of the study and how these complement and extend our knowledge on provision of support for caregivers of children with neurodevelopment disabilities. Strengths and limitations of the research design are also considered, as is what may be required to scale up *Juntos,* or a similar intervention, to meet the needs of more families. In considering these questions, I draw upon learning from other aspects from the DrPH programme including policy context and political prioritisation. The discussion also provides a reflection on the utility of Bowen et al's model for assessing feasibility [5] and considers what might be required to assess the two additional areas of their framework: integration and expansion to begin to ascertain if *Juntos* 'does work'. Integrated into the chapter are recommendations for policy makers, for programme implementers and for researchers about the next steps for continued implementation, expansion, and integration of *Juntos*. The chapter also presents final conclusions to the study.

Annexes – Ethics approval (Annex 1); consent forms (Annex 2); interview guide (annex 3); three additional papers (Annexes 4-6) published in peer-reviewed journals related to the project (which I was a co-author (but not lead author); and comments made by the peer reviewers (Annexes 7 and 8) for the first and fourth papers (chapters 2 and 6)

Context of the DrPH programme

The Doctorate in Public Health (DrPH) programme at LSHTM is targeted towards health policy makers and public health practitioners, rather than researchers and academics. The DrPH programme has three distinct, but linked, components:

1. A taught component comprising two compulsory modules – Understanding Leadership, Management and Organisations (ULMO) and Evidence-Based Public Health Policy and Practice (EBPHP).
2. Research Study I: an Organisational and/or Policy Analysis (OPA) project, whereby the student undertakes a practical piece of work with a public health organization to explore and provide recommendations on the policy environment.
3. Research Study II: a research project leading to the production of a thesis.

The taught element of the programme, which I undertook in Autumn 2014, plaid a strong foundation for the other two components and was useful for my work. As a physiotherapist working for an international non-governmental organization (NGO) on improving recognition of rehabilitation services within the global health agenda, I utilized several of the approaches from these courses. For instance, consideration of how the three streams of policy making described by Kingdon [6] – problems, policies and politics - were extremely relevant to the work I was undertaking. This included developing an "advocacy framework" for rehabilitation which guided country teams on how they evaluate their advocacy and policy work relating to the three streams.

My OPA took place with the Pan American Health Organisation (PAHO) and drew from both the ULMO and EBPHP courses. The role involved undertaking an analysis of 10 policy documents or actions plans in the Department of Non-communicable Diseases and Mental Health (NMH) considering whether, and to what extent, the two issues of i) people with disabilities and ii) rehabilitation services were considered. The analysis also involved interviews with unit chiefs and other stakeholders to understand the policy setting context and understand how disability and rehabilitation can be better included within PAHO health policies. The results demonstrated that both evidence-based public health policy and organisational structure had important roles in shaping how PAHO policy documents are created and what gets included. On the one hand, it was clear that evidence was a crucial "informer" of policy creation within the organisation. On the other hand, both the internal and external dynamics of the organization had important influences on policy creation. The learning from the two taught courses was extremely valuable in developing insights into the policy creation processes within the organisation and providing a set of recommendations for PAHO to take forwards.

The third component of the DrPH is this research project described within this document. While the research project itself was a 'classic' implementation of primary research, involving collection and analysis of data (in addition to the programme management elements of establishing and launching a project initiative), there were additional layers of consideration which went beyond the scope of traditional research and drew from elements of the DrPH programme. For instance, I have used learning from both the taught courses and the OPA to reflect on what may be required from a policy perspective for growth and scale up of the programme. I also used insights from the taught courses and applied these to the use of Bowen's feasibility framework [5].

The COVID-19 pandemic impacted my ability to finish the final write up until this point due, in part, to commitments in my role with the Pan American Health Organisation.

CHAPTER 1: INTRODUCTION

Figure 1: Facilitators and researchers at the end of the initial training in Rio de Janeiro

The emergence of Zika as a public health issue: The 2015-16 Zika epidemic

Zika was not a new virus when it emerged in Brazil in 2015. Zika was first identified in Uganda in 1947 [7]. It was mapped as it spread to western Africa and Asia over the following decades, but received very little attention within the global health community, since it was thought to have caused no deaths and was seen as largely 'benign' [8]. Although a number of outbreaks of the virus had occurred, including in French Polynesia in 2013 [9], it was not until high numbers of cases were recorded in Brazil in February 2015 that Zika started to garner significant international attention [10]. Zika was declared a Public Health Emergency of International Concern (PHEIC) by the World Health Organisation in February 2016 [11], after it became clear that infection during pregnancy could result in microcephaly and other congenital abnormalities. By this time, 31 countries within the Americas had reported local transmission of the virus [12], and the virus had also spread to other parts of the world including Southeast Asia.

In Brazil, it was ascertained that the virus entered the Americas in late 2013, a full year before it was detected [13]. The Northeast of the country, containing nine states including Pernambuco, Bahía and Paraíba, and cities such as Recife, Salvador, Fortaleza and Natal, was the epicentre of the epidemic. The Northeast region of Brazil is one of the most economically disadvantaged with a high population density and high unemployment rates [14]. From the Northeast, genomic sequencing has shown that the virus travelled to southeast Brazil and to elsewhere in the Americas [15]. Figure 2, below, taken from Lowe et al [8] shows the geographic distribution of both Zika and microcephaly cases in Brazil.

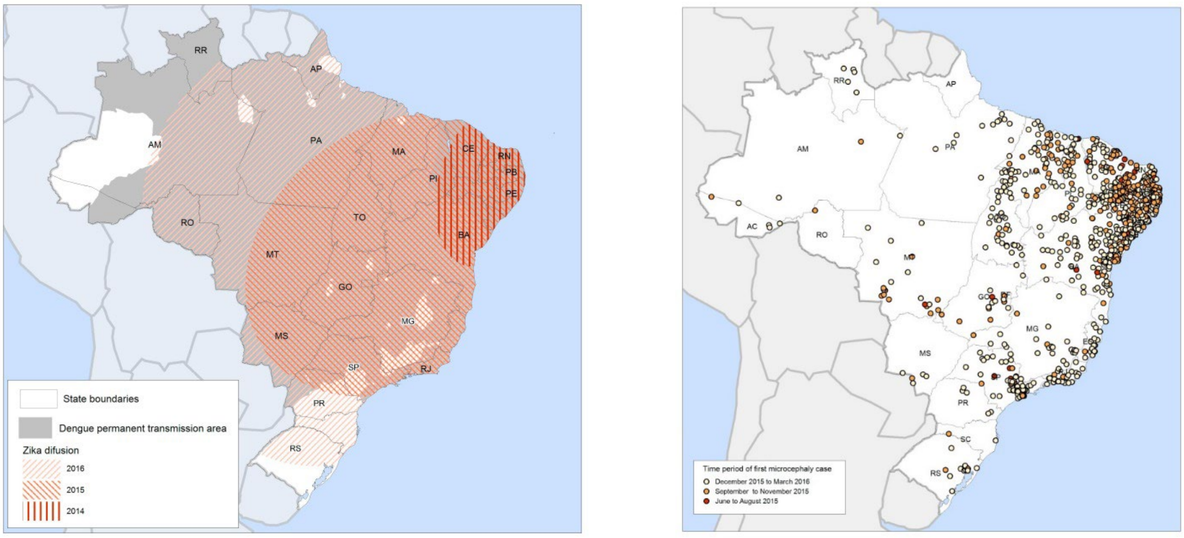


Figure 2: geographic distribution of zika cases (left) and microcephaly (right) taken from Lowe et al (2018)

The WHO lifted its declaration of Zika as a PHEIC in November 2016 with the recognition that Zika was likely to remain a challenge for the public health community and that “a robust longer-term technical mechanism was now required to manage the global response” [16]. By that time, more than 500,000 cases had been reported globally [17], but Latin American countries (and Brazil especially) were the most heavily impacted. Since then, there have been no further major outbreaks of Zika in Latin America or elsewhere. However Zika continues to cause low numbers of infections and subsequent consequences in occasional smaller outbreaks such as in Kerala, India in 2021 [18, 19]. Research has also suggested that a minor mutation of the virus could cause a new significant outbreak in the future [20].

What remains from the 2015-16 epidemic are thousands of children with what is now called “Congenital Zika Syndrome” (CZS) who experience a range of conditions and require support to maximise their development. This thesis will describe the rationale for and the process of developing and pilot-testing a group-based intervention to support children with CZS, through building the skills, capacity, and support networks of their caregivers.

Zika Virus and Transmission

Zika is a virus within the Flaviviridae family of positive-stranded RNA viruses [7]. Other viruses in this family include Yellow fever, Dengue, and West Nile virus [21]. There are two Zika lineages – an African and Asian one [22]. The South American outbreak was most closely connected to the Asian lineage [23]. The Zika virus is primarily spread by the Aedes genus of mosquitoes, particularly the female *Aedes aegypti* and (to a lesser extent) the *Aedes albopictus* mosquitoes, which are found in tropical and subtropical zones across the world [8] [24, 25]. Zika infects a human host through the bite of the mosquito who is carrying the virus (which it likely acquired from an animal host). Once introduced to a human, the virus is internalised by endocytosis, through the release of viral DNA into the endoplasmic reticulum [22]. From there, the virus can quickly replicate.

The Zika virus (ZIKV) can also be transmitted sexually [26]. This transmission is more likely to occur from a man to a sexual partner compared to a woman [27], in part because the virus seems to reside longer in seminal fluid (median duration 34 days) compared to the female genital tract (median duration 12 days) [28]. The risk of sexual transmission is relatively low, estimated per sex act at 1.3% for male-to-male transmission, 0.4% for male-to-female and 0.1% for female-to-male [27]. Sexual transmission is therefore thought to be a significantly less common cause as a route of transmission, compared to mosquito borne transmission [28]. Sexual transmission as a cause of Zika infection has, however, still been shown to be linked to CZS [29].

The final route for transmission is between a pregnant woman and her unborn foetus. This route of transmission was not known before the 2015 outbreak. It is now understood that (unlike many viruses) in around 20%-30% of cases [30] ZIKA is able to cross the human foetal-placental barrier and infect the brain and nervous system of a developing foetus [31].

Health impacts of Zika virus in the infected patient

The incubation period between being first infected with the virus and exhibiting any symptoms is between 3-14 days. However, the majority of people (thought to be around 4 out of every 5 people) do not exhibit any symptoms at all [24]. Prior to the 2015 outbreak, the Zika virus was not thought to be connected to any long-term health complications [32]. Indeed, Petersen notes that after its initial link to human illness was discovered in Nigeria in 1957, *“only 13 naturally acquired cases were reported during the next 57 years”* [7]. Those who do show symptoms often have very mild flu-like symptoms (fever, muscle and joint pain, headache) and possibly a skin rash which can last up to 7 days. Most cases only require self-management of symptoms at home and no significant medical interventions.

The 2015-16 Zika outbreak also proved the connection between ZIKV and Guillain-Barré Syndrome (GBS). GBS is a serious neurological condition which causes a rapid weakening of the limbs and reduction in reflexes. It leads to significant respiratory deterioration (often requiring intensive care intervention) in around 25% of cases [33]. GBS is the most common cause of acute flaccid paralysis and affects around one in every 100,000 people [34]. It has been shown that rates of GBS may have been 2.6 times higher than expected during the Zika outbreak [35], though recent modelling has also suggested a risk of developing GBS from Zika infection during the outbreak being around 0.0061% [36]. Most patients provided with the right medical interventions survive, but they may require significant rehabilitation and they may experience long-term disabling consequences [33].

Prevention of ZIKV infection

Prevention of ZIKV infection primarily involves prevention of the causes of infection, whether that through the bite of a mosquito or sexual transmission. These included (in the case of mosquito bite transmission) removal of stagnant water; population education and individual strategies to prevent being bitten (e.g. wearing of long-sleeved clothing, mosquito nets etc). For sexual transmission, people who had travelled to ZIKV zones were advised to refrain from unprotected sex for a period of time [37]. Vaccine trials and development have commenced, but to date there is no viable ZIKV vaccine on the market [38].

Congenital Zika Syndrome and other neurodevelopmental sequelae

Nine months after the initial onset in cases of Zika in Brazil, a significant increase in cases of microcephaly among new born children led the international public health community to suspect a connection between Zika and these disorders [39] [10]. Figure 3, below, shows this correlation of Zika infection and microcephaly, by region in Brazil.

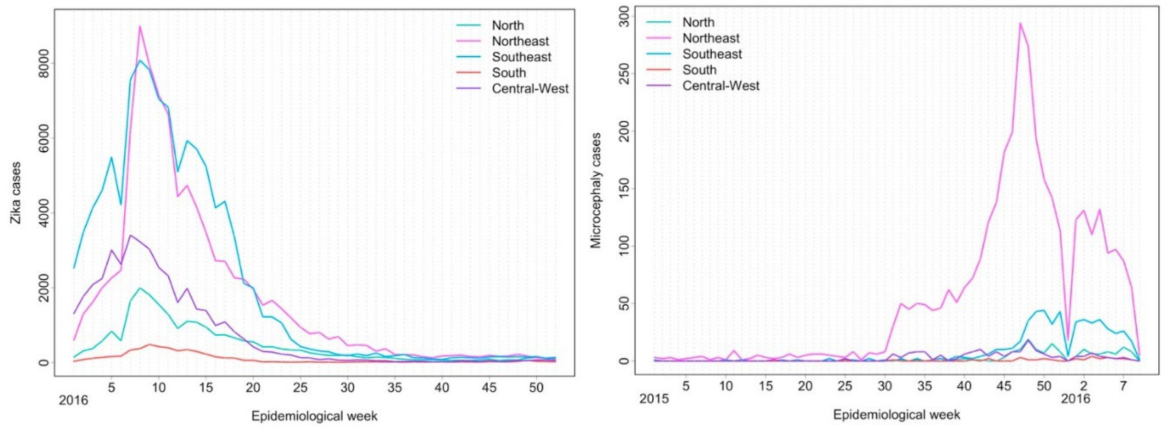


Figure 3: Notified cases of Zika infection (left) and of microcephaly (right) by epidemiological week in 2016, taken from Lowe et al (2018) [8]

This causal link has since been confirmed through several epidemiological and basic science studies [40]. A retrospective analysis of the French Polynesia outbreak also confirmed an increase in cases of microcephaly after Zika infection [41].

Microcephaly is a condition where the head size of a baby is small, as compared to other babies of the same age and sex. This may be immediately apparent at birth or only during infancy if the head grows at a lower-than-expected rate. Microcephaly can often lead to long-term physical, sensorial and intellectual impairments [42, 43]. While the precise physiology of what causes these alterations to the foetus is not fully known, it is likely that microcephaly results from the disrupted centrosome organization and mitotic abnormalities observed in cells that are infected with ZIKV [22].

Congenital Zika Syndrome (CZS) began to be used as the term to describe the neurodevelopmental sequalae that was affecting children born to mothers who had been infected with the Zika virus as it was clear that the impacts were greater than just microcephaly. Moore *et al* presented the first comprehensive overview of the characteristics of CZS in 2017 [44]. They described CZS as a recognizable pattern of structural anomalies and functional disabilities secondary to central and perhaps peripheral nervous system damage. Further, they suggested that there are five features unique to congenital Zika infection:

- 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.

Recent evidence suggests that not all children with CZS present with microcephaly at birth. Some children are born with a normal head circumference and go on to develop microcephaly later while others show evidence of other features without microcephaly [45]. Mortality of live-born children with CZS in Brazil during the pandemic has been estimated to be 11.3 times higher than those born without the syndrome over the first three years of life [46]. A broad range of impairments related to CZS have been described in the literature including several cohort studies that have undertaken longitudinal analyses within cohort groups [47, 48]. These impairments include:

Motor impairments: A systematic review by Freitas et al [49] describes both neurological and osteoskeletal abnormalities reported in the literature. Many of these impact on a child's capacity for gross and fine movements such as walking, sitting, grasping items etc. Common neurological abnormalities described include hypertonicity and hyperexcitability/hyperreflexia and osteoskeletal abnormalities include arthrogryposis (joint contractures), hip dysplasia and clubfoot.

Intellectual impairments: Intellectual impairments, or the likelihood of long-term intellectual impairments appears frequent. This includes autism spectrum disorder [50].

Dysphagia (difficulties swallowing): Dysphagia and other feeding and swallowing difficulties have been frequently reported, which has had a significant impact on the babies' ability to breastfeed [51] [52].

Epilepsy: A high incidence of epilepsy and spasm was reported early in the pandemic [53]. A more recent study, which looked at outcomes at 24 months suggested that there was a cumulative incidence of epilepsy of 71.4% [54] and another suggested 54% [55].

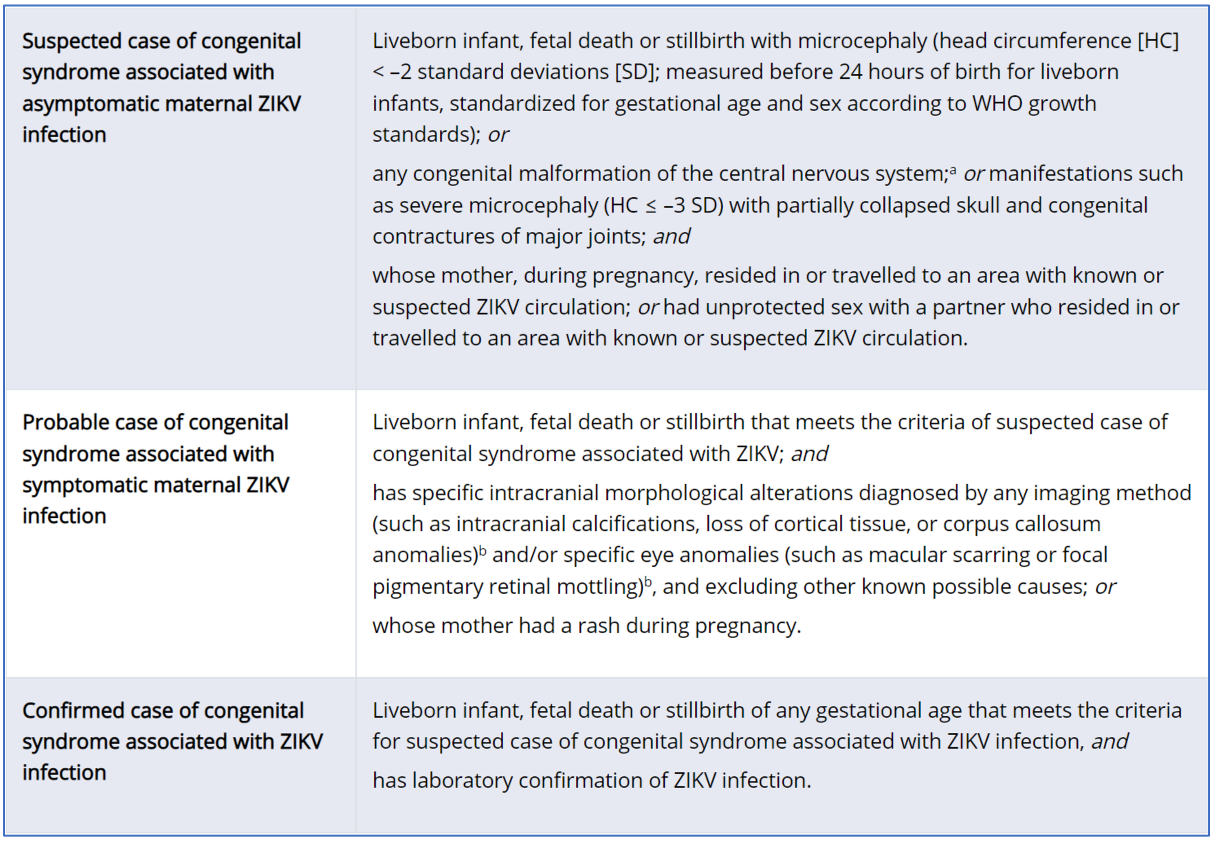
Irritability: which was reported as the most common symptom in one study of 48 infants, with 85% of cases showing irritability [52]

Hearing loss: Sensorineural hearing loss was reported relatively early as one of the potential symptoms of CZS [56]. A three year follow up study has suggested a 9.3% prevalence of hearing loss [57]

Vision abnormalities: Ophthalmic changes seem to be relatively common [58] and one paper suggests that these are seen in around 7% of cases [59].

Testing and classification

Formal testing for Zika is done through RT-PCR testing [60, 61]. However, since the window for confirming the presence of the virus is relatively short, there have been challenges developing a reliable test. Therefore, cases of congenital syndromes associated with Zika virus have three different classifications: suspected, probable and confirmed (as shown in Figure 4, below, which is taken from the CDC webpages describing congenital Zika syndrome [62]).



Suspected case of congenital syndrome associated with asymptomatic maternal ZIKV infection	Liveborn infant, fetal death or stillbirth with microcephaly (head circumference [HC] < –2 standard deviations [SD]; measured before 24 hours of birth for liveborn infants, standardized for gestational age and sex according to WHO growth standards); *or* any congenital malformation of the central nervous system;[a] *or* manifestations such as severe microcephaly (HC ≤ –3 SD) with partially collapsed skull and congenital contractures of major joints; *and* whose mother, during pregnancy, resided in or travelled to an area with known or suspected ZIKV circulation; *or* had unprotected sex with a partner who resided in or travelled to an area with known or suspected ZIKV circulation.
Probable case of congenital syndrome associated with symptomatic maternal ZIKV infection	Liveborn infant, fetal death or stillbirth that meets the criteria of suspected case of congenital syndrome associated with ZIKV; *and* has specific intracranial morphological alterations diagnosed by any imaging method (such as intracranial calcifications, loss of cortical tissue, or corpus callosum anomalies)[b] and/or specific eye anomalies (such as macular scarring or focal pigmentary retinal mottling)[b], and excluding other known possible causes; *or* whose mother had a rash during pregnancy.
Confirmed case of congenital syndrome associated with ZIKV infection	Liveborn infant, fetal death or stillbirth of any gestational age that meets the criteria for suspected case of congenital syndrome associated with ZIKV infection, *and* has laboratory confirmation of ZIKV infection.

Figure 4: Case Definition for ZIKV infection taken from the website of the CDC

Epidemiology and frequency of CZS

As previously described, it is estimated that mother to child transmission occurs in around 20-30% of cases [30]. In Brazil there are over 3000 reported cases of children with CZS [63]. As it is very difficult to ascertain the number of children with a wider spectrum of more mild developmental disabilities due to challenges with testing and diagnoses, most cases recorded are likely to be children with microcephaly. As was shown in Figure 1, a higher proportion of cases of microcephaly occurred in the Northeast region of Brazil. Children with CZS have also been noted to be more likely to come from lower income households [64].

Healthcare needs of children with CZS

Much of the specific healthcare needs relating to medical management of CZS depends on the severity of the condition and the specific impairments that present in the child. Children may be under the care of a paediatric specialist or neurologist and have regular appointments, diagnostic tests or other interventions. Depending on the child's capacities in feeding and swallowing, a child may need to have adapted diets (e.g., thickened fluids) or, if a child is unable to safely swallow or cannot adequately eat/drink enough to meet their nutritional needs, they may require a feeding tube or Percutaneous endoscopic gastrostomy to supply additional nutrients directly into the stomach. This is a surgical procedure and caregivers would require training and a supply of the appropriate nutrients.

Bladder or bowel management may need to be incorporated into the medical management also, depending on a child's level of continence. If a child has epilepsy they may require ongoing medication, monitoring and additional testing. If a child has auditory or visual difficulties, they may require specialized services such as ophthalmology or audiology to fully diagnose and manage these impairments.

Habilitation, rehabilitation and assistive products

There are additional health and social interventions which may be required to ensure that a child has the opportunity to optimise their functioning and is able to fully participate within society. There is an important role in the management of CZS for services like rehabilitation [65]. Although CZS was a new condition, habilitation and rehabilitation approaches for children with microcephaly or other neurodevelopmental disabilities (NDD) is not new.

Rehabilitation interventions that may be required are also guided by the specific functioning needs of a child. They may be likely to involve one or more of: 1) Physiotherapy, to promote movement and functioning [66]; 2) Occupational Therapy to enhance the child's ability to engage in their everyday activities [67]; 3) Speech and Language Therapy, to support difficulties in communication or with eating, drinking and swallowing [68]; 4) Orthotic devices and assistive products such as wheelchairs, pressure mattresses or communication devices to support the child in his or her day-to-day functioning.

General healthcare needs

In addition to any healthcare needs associated directly with impairments caused by CZS, it is important to remember that children with CZS have the same healthcare needs as all other children of a similar age. In younger children, that might mean access to routine childhood vaccines, treatment of day-to-day health issues (e.g., small abrasions) or dental check-ups. Unfortunately, general health facilities are often extremely inaccessible for people with disabilities, or they may have negative experiences in accessing general healthcare [69]. This can lead to additional health consequences [70].

Healthcare services and availability in Brazil during the Zika outbreak

Brazil strives for a health system of universal health care. The health system is managed by the federal ministry of health, the state departments of health, and the municipal departments of health [71]. The unified health system – SUS – which is available for all Brazilian citizens, is the mechanism by which health services are delivered.

Despite Brazil's bold ambitions to have comprehensive universal health coverage, even prior to the outbreak, coverage of services was noted to be variable [71] and healthcare needs may have often been unmet. This was true also for rehabilitation services, since there was a quite centralised model [72] which didn't optimize service delivery. The additional strain that the Zika outbreak caused was noted to add to the existing challenges within the health system [72-74].

Gaps in rehabilitation [75] services in Brazil were noted as was access to general healthcare services for children with CZS [74]. Caregivers also reported high economic burdens in obtaining healthcare services for their children [76]

CZS in context of Neurodevelopmental disability

Although CZS is a relatively newly defined condition, much can be learned about its implications and managements from other similar conditions. CZS can be classified as a neurodevelopmental disorder (NDD). NDD is an umbrella term for a range of conditions that as a group of conditions *"with onset in* *the developmental period, inducing deficits that produce impairments of functioning"* [77]. The main categories of NDDs are intellectual disabilities, autism spectrum disorders and Attention-Deficit/Hyperactivity Disorder. NDDs are often irreversible and can lead to lifelong impairments and disability. Developmental disorders are common – approximately 8.4% of children under 5 have one, according to the Global Burden of Disease studies (which includes vision and hearing loss) [78]. Another systematic review estimated that the prevalence of NDDs is 7.6% in children under 19 [79].

Among the NDDs, CZS potentially shares most similarities with Cerebral Palsy (CP). CP is *"a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by a seizure disorder"* [80]. Cerebral palsy develops during pregnancy, after birth or in early childhood due to a variety of causes such as birth asphyxia, hyperbilirubinemia, infections with most causes occurring during pregnancy *[81]*. As such, it is likely that many children with CZS may also have Cerebral Palsy [48, 53]. Furthermore, even for those children with CZS, but no clinical presentation of Cerebral Palsy, there are likely to be large overlaps in terms of approaches to manage the impairments relating to the condition and even more so in terms of addressing societal barriers relating to having a child with a disability.

CZS in the context of disability and holistic support

So far, CZS has been described in relation to its medical presentation and associated symptoms, but it is also important to describe it in the context of disability. Disability "results from the interaction between persons with impairments and attitudinal and environmental barriers that hinder their full and effective participation in society on an equal basis with others [82]. There are approximately one billion people with disabilities in the world, constituting 15% of the global population [83].

There are different ways of conceptualizing disability. One key approach is through the International Classification of Functioning, Disability and Health (ICF). ICF is a tool developed by the WHO to describe the relationship between a health condition and the impact on the body, on a person's capacity to perform functions such as walking or command societal levels that interact to create (or minimise) disability. It is valuable in helping to identify, visualise and measure the interactions at different levels. Within ICF, 'impairments' describe changes at the body structure or function, such as movement of a limb. Activities and participation then describe a person's capacity and performance in relation to their personal situation and tasks for example, walking (activity) or going to school (participation). These two areas are often highly variable based on a person's personal factors such as age or educational level or environmental factors such as accessibility of spaces or attitudes and beliefs of the society in which they live. The ICF is important as it considers impacts beyond health/impairment, but also factors that alleviate disability beyond medical interventions alone. It provides a framework for a more holistic approach to understanding disability. Figure 5, below, shows the model of the ICF:

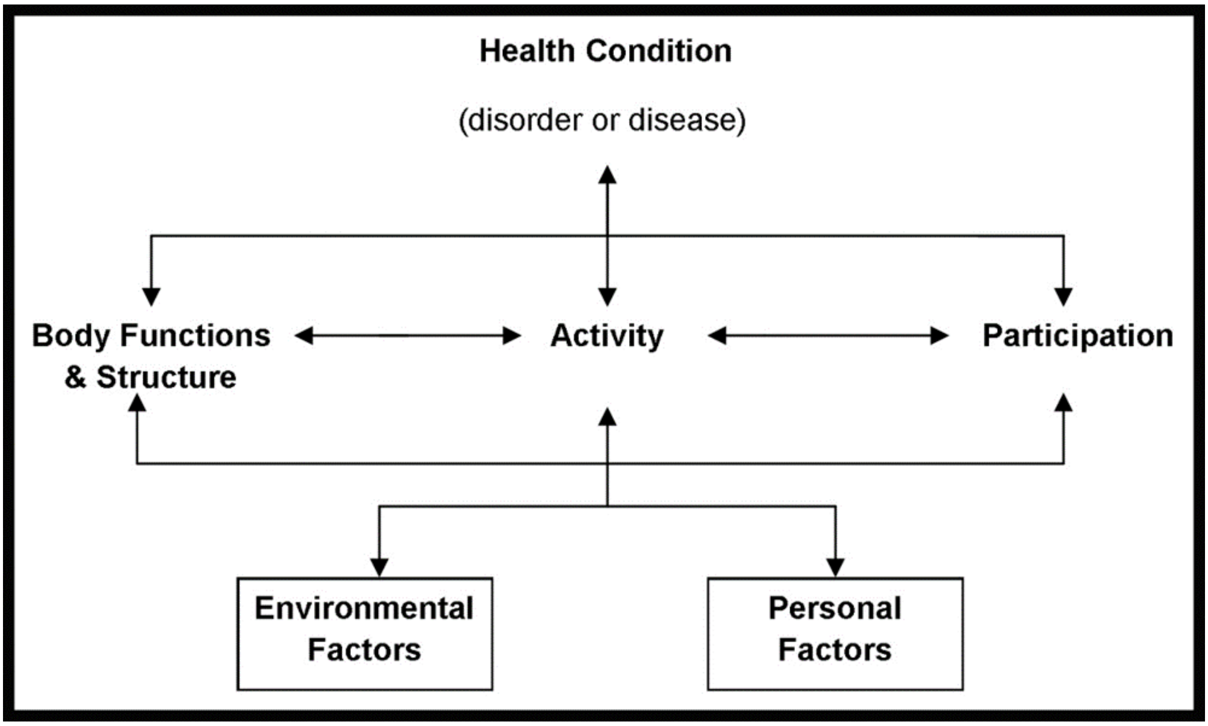


Figure 5: The International Classification of Functioning, Disability and Health

Three studies undertaken in Brazil have used the ICF in relation to CZS using the ICF. Campos et al [84] provides a comprehensive overview of CZS in relation to domains of the ICF based on focus group discussions with 32 caregivers. Figure 6, below, shows the main categories commonly linked across the different elements of the ICF. In relation to body functions and structures, a range of motor, sensory and other functional impairments were described such as seeing, hearing, movement, sleeping, breathing and pain. Activity limitations and participation restrictions were reported in relation to areas such as eating, speaking, walking and family relationships. Finally, in relation to environmental factors, the immediate family, society attitudes and transportation were among those noted in relation to CZS. The three most reported categories of difficulties and needs associated with CZS were related to *health services systems and policies (e580* - environmental factors*)*, *control of voluntary Movement (b760* - body structure and function) and *products and technology for personal use in daily life* (e115 - environmental factors).

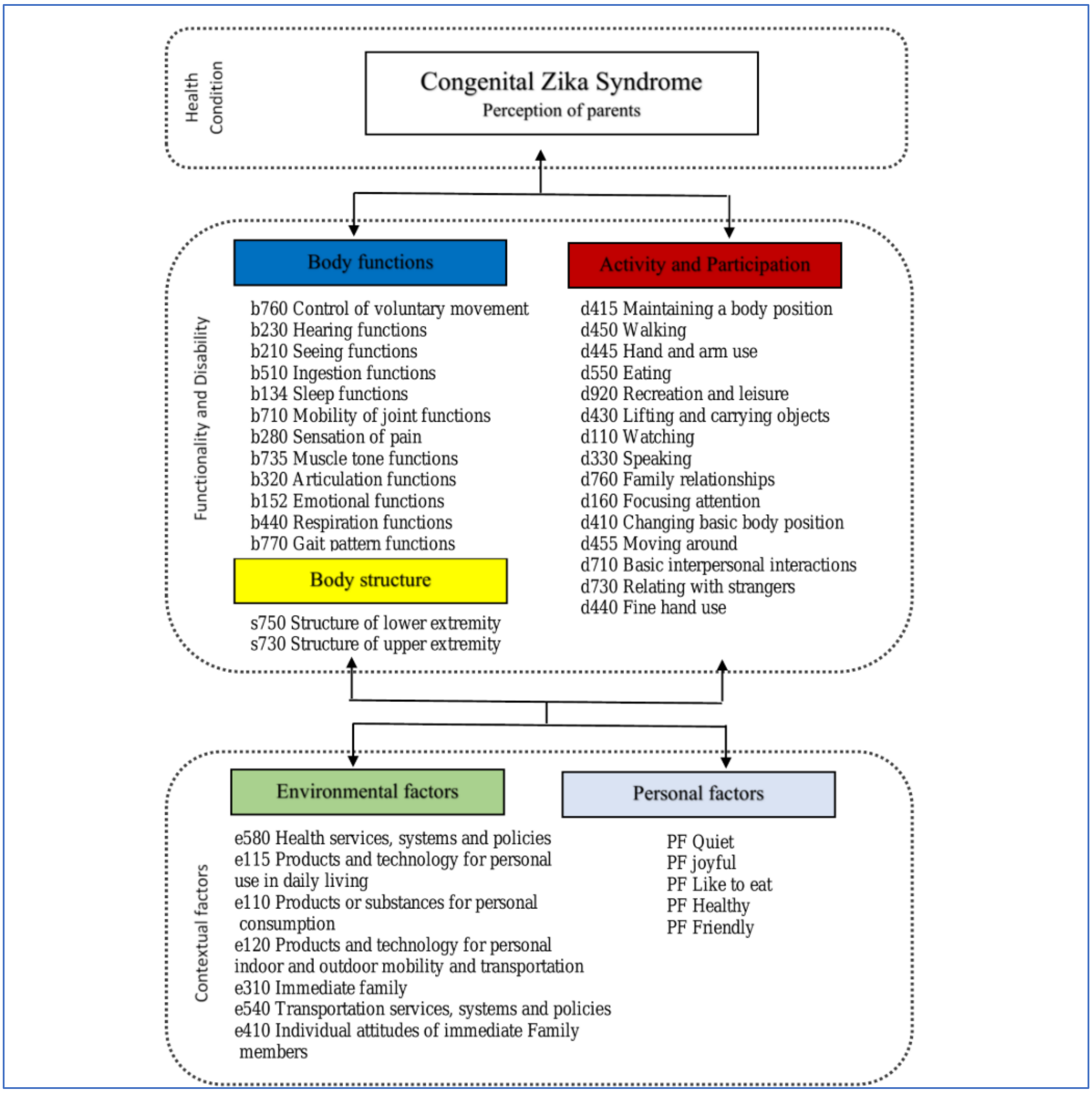


Figure 6: ICF categories representing the parents' perspective taken from Campos et al (2020)

The Campos study provided an update of domains that Fereira et al [85] had published two years prior when documenting 34 children with CZS, which highlighted categories of functioning difficulties including *intellectual functions* (b117) and *mental functions of language* (b167) Also highlighted were *difficulties with fine hand use* (d440) and *needs for products and technology for communication* (e125).

Finally, Longo et al [86] considered how mapping CZS to ICF can help to develop and standardise rehabilitation interventions and approaches including data collection. They showed a successful pilot where this has been done, identified challenges to implementing the ICF at present including lack of tools to implement the ICF, lack of accurate diagnostics and cultural restrictions. However, if these challenges were to be overcome, Longo et al highlight how its implementation can build and guide rehabilitative interventions across the life course.

Applying the ICF to conceptualise CZS in relation to broader disability, therefore widens the lens beyond healthcare interventions alone. However, this necessitates consideration of the key challenges facing children with CZS and their families as discussed below.

Wider socio-economic impact of CZS

In addition to the immediate medical and rehabilitative needs of children, CZS was widely reported to have broader socio-economic impacts for families of affected children. A sister study to the current study, undertaken by LSHTM and Brazilian partners, explored some of these areas in further detail [87]. Three areas of the socio-economic impact of CZS have been particularly raised in research and literature: poverty, stigma, the mental health of caregivers, and education for children.

Poverty

Caregivers of children with CZS are both more likely to come from low-income neighbourhoods as well as have limitations in being able to work due to caring duties [64]. Added to already challenging pre-existing socio-economic statuses has been the burden of catastrophic expenditures related to the care of a child with disabilities [32, 88-91]. Different types of support are therefore needed for carers on account of these financial pressures, such as vocational rehabilitation and social protection.

Brazil has a system of Social Assistance Benefit called (Benefício de Prestação Continuada – BPC) which supports people with disabilities and older persons [92] (Portuguese). Anecdotal findings from the scoping visit to Brazil [2] suggested that being able to receive this benefit was based on a medical diagnosis where children with microcephaly were able to access the fund. A recent study made it clear that children with CZS but with less severe impairments and disabilities were less able to access to the benefit, and therefore experienced more profound economic impacts than children with microcephaly [76].

Stigma

Disability can often be associated with stigma, myths and societal attitudes which can vary greatly based on the context [93]. Stigma can arise for many reasons, including a lack of understanding or awareness, misconceptions, or discriminatory legislation and policies [94]. However, stigma is a complex and nebulous concept, variably defined and measured. "Stigma" may include negative attitudes of others towards people with disabilities (including from family members), and self-stigma. Discrimination and exclusion can also be considered as under the umbrella of the word "stigma". Despite the issues with the classification, it is clear that children born with disabilities and their parents, can be victims of particularly harmful stigma and stereotypes. In Ghana, for example, Zuurmond et al. describe social stigma at the familial levels, whereby rejection and blame were apportioned by families towards the mother and child, as well as internalised stigma, whereby mothers expressed negative feelings about themselves and expected others to stigmatise them [95]. In India another study showed a clear link of poorer psychological wellbeing associated with affiliate stigma among mothers of children with learning disabilities [96]. Poorer mental health of parents may contribute to the high levels of paternal abandonment of children with disabilities. People with disabilities in Brazil continue to face significant levels of exclusion, barriers and ableism in being able to fully participate in society [97]. The most recent Brazil reports to the UN Committee on the Rights of Persons with Disabilities highlighted that while progress is being made in Brazil on creating more equal societies for people with disabilities, there is still a long way to go before this is fully realised [98].

There was some emerging evidence, however, that stigma experienced by children with CZS may be less than for children with other conditions. There was some anecdotal evidence that fathers of children with congenital Zika syndrome were more likely to be present and involved in raising their children as compared to fathers of children with other neurodevelopmental disabilities. This was a finding that emerged during the in-country visit in April 2017, however other research has described father abandonment [99]. This may have been due to the high level of media attention that came during the Zika outbreak. Interviews and articles written about the children and their families put their cases in the spotlight. As a result, their fathers may have been likely to abandon them since they were in the public eye. That said, there doesn't seem to be a significant amount of concrete evidence to support this or research that has specifically looked into father abandonment of children with congenital Zika syndrome compared to other neurodevelopmental disabilities. One study did interview fathers and discovered significant gaps in relation to support and knowledge [100].

The high levels of media attention on Zika also seem to have had a role in potentially improving societal understanding that this was no fault of the mother, and she should not be the bearer of any blame. Sadly, mothers are often the targets of blame within society when a child with a disability is born. Interestingly, in Brazil a counter-movement of caregivers labelling their children as ‘angels’ also strongly emerged [97].

Mental health of parents

The psychosocial wellbeing of caregivers has also been explored. They have higher levels of anxiety, depression, and stress [101-104]. This echoes what has been shown in terms of mental health impacts of caregivers of children with NDD more generally [105]. In Brazil there were low levels of social support were linked to higher rates of depression and anxiety among caregivers [106].

To address this, there is an important need for caregivers to have an opportunity to receive professional psychosocial support when indicated. However, the important of peer to peer and family support for psychosocial needs is also crucial.

Education

Considerations about the education of children with disabilities is important, particularly as they reach the age for schooling. The UN Convention on the Rights of Persons with Disabilities reinforces that all children have a right to education, including those with disabilities [82]. Considering how children with

CZS may access their local school will be an important factor for the education sector to ensure that these children do not get left further behind [89] [107].

Supporting the needs of children with CZS and their caregivers

This literature review shows that medical support and rehabilitation are vital to address some of the health needs, impairments, activity limitations and participation restrictions associated with CZS. However, alone they will not address the holistic needs of children and their caregivers. In particular, medical interventions may be unlikely to target some of the environmental factors that were listed when describing CZS though the ICF, such as stigma. Wider support is therefore needed to ensure that children, and their caregivers, are supported, able to address some of these environmental factors, and optimise activities and participation. Providing education and support to caregivers is being increasingly recognised for NDD, including through the establishment of support groups and training. The topic of this DrPH is to develop and pilot-test such an intervention.

The scoping visit to Brazil and assessment of needs that was undertaken as part of this DrPH [2], and discussed in more detail in Chapter 4, ascertained that different options were available. Although CZS is relatively new, there is a lot of learning possible from other NDD – in particular CP - which have more well-developed programmes. The needs analysis, described in Chapter 4, ascertained that although there were networks of carers who may meet via virtual platforms (e.g. WhatsApp) or face-to-face (e.g. Associação aBRAÇO a Microcefalia [108]), these did not necessarily run through a pedagogical approach and tended to be more “informal” in their support nature. There were examples where individual support was often offered (e.g., holistic services offered at Ventura in Recife), but these did not allow for significant exchange between caregivers. There were some virtual or theory-based caregiver training such as Caregiver Skills Training from the WHO [109], but the uptake was low. Lastly some pilot parent support programmes had been established but did not have a detailed curriculum and tended to be therapist led.

A formal parent support programme was selected as the most appropriate intervention for this project to address the wider unmet need of carers of children with CZS, for several reasons. First, there was a very positive response from parents and health teams during our scoping visit to Brazil. Second, it was felt that this would fill a significant gap that was not currently being addressed. Many caregivers were accessing individual rehabilitation or other health services but did not yet have the peer-to-peer support. Third, it was seen as feasible to pilot – the two partner groups (FIOCRUZ in Rio and Universidad de Bahia in Salvador) both felt that sourcing a location and identifying and training facilitator teams would be achievable. Finally, Brazil has a proud history of promoting pedagogy for vulnerable and marginalised groups stemming from the country's affection to the philosopher Paolo Freire, who wrote the famous literature *Pedagogy of the Oppressed [110].*

Parent support programmes

Parent support programmes are a common way of imparting knowledge and skills to parents of children. There are many parent support initiatives that have been successfully created and implemented in a range of contexts. Somewhat less available are specific programmes for parents of children with neurodevelopmental disabilities. This is especially true in low- and middle-income contexts, where services such as rehabilitation may be limited.

In 2011, ICED launched a programme called *Getting to Know Cerebral Palsy* (GTKCP) in response to a project on child disability in Bangladesh [111, 112]. The research had identified large numbers of children with Cerebral Palsy who had extremely limited or no access to habilitation or rehabilitation services such as physical therapy or occupational therapy. A parent training project was developed and launched following a participatory process to interview families to better understand their lives and priorities [113]. This pilot training programme was then further implemented and tested in Ghana where it underwent more rigorous analysis [114].

GTKCP is a programme containing a guided framework of 10 sessions, led by local facilitators, which aims to *"deliver a model rehabilitation training programme for families of children with cerebral palsy*

…[and] to develop a training resource that empowered families, and thus place[d] greater emphasis on use of a participatory approaches to training at community level". It is based on a previous programme in South Africa called *Hambisela* [115]. The Ghana analysis has revealed positive impact on caregivers' quality of life and knowledge and confidence of caring for their child [114]. The evaluation identified some positive outcomes in relation to feeding and nutrition [116]. Since its origins in Bangladesh and Ghana, GTKCP has been implemented in many low-resource countries globally [117]. GTKCP was adapted in Uganda in 2014 into an *Early intervention Programme* (EIP) for children younger than 2 years of age [118]. EIP has shown feasibility, acceptability, and impact in initial evaluations [119]. The structure of GTKCP and EIP is shown in Figure 7, below. These two programmes have recently been rebranded as *Ubuntu* and *Baby Ubuntu* and a website has been created to track their implementation together with *Juntos,* the programme developed and described in this research project [120].

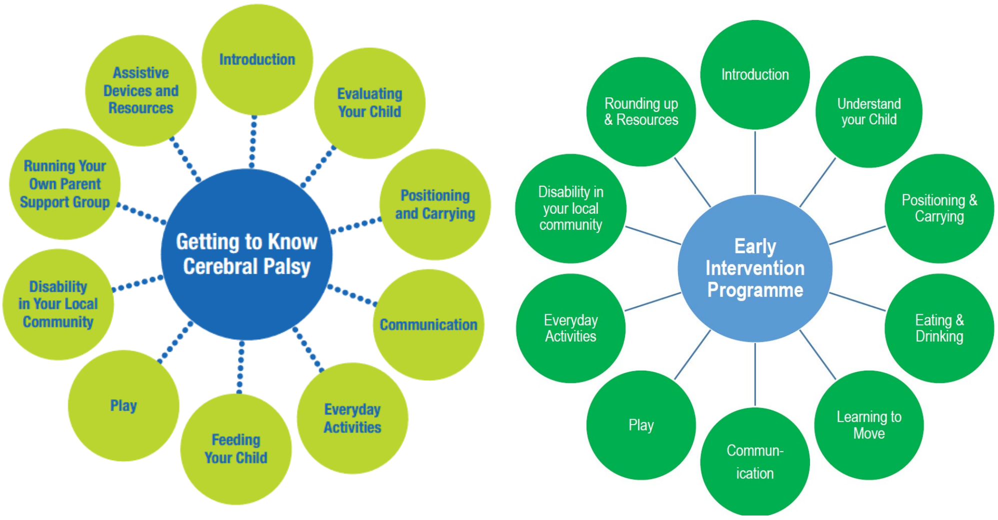


Figure 7 ***Structure of original Getting to Know Cerebral Palsy programme (left) with the newly developed programme for under twos (right)***

Both GTKCP and EIP are examples not only of facilitating the empowerment of parents or caregivers, but also of health promotion more generally. By targeting an intervention to the caregiver, it is likely to have an impact on the health and wellbeing of a child both in the short and longer term [121]. For the context of CZS in Brazil, *GTKCP* and the *EIP* were seen as a good starting point for providing parent support as they were already established programmes for caregivers of children with not dissimilar NDDs. Indeed, as described earlier, it has been suggested that many children with CZS also have CP [48]. Time was of the essence in this emergency, and there was not time to create a new support programme, and so starting with existing material was seen as preferable.

Although *GTKCP and EIP* were available for use, it was also felt that there was a need to adapt them for their implementation in Brazil. First, this was in relation to some of the new and emerging issues that were arising in how CZS may be different to CP and the implications of this for the content of the course. For example, irritability was frequently noted to be high, but neither GTKCP or EIP contained any guidance on soothing highly irritable babies. Second, an adaptation was required for the context of Brazil, a Latin American country with a very different culture and society that Ghana, Uganda or Bangladesh where the *GTKCP* and *EIP* programmes had been previously implemented.

Aims and objectives of the study

The **overall aim** of the research project was *to assess whether a community intervention aimed at families of children with CZS in Brazil is needed, acceptable and feasible.*

The **specific objectives** of the research project were:

1. To undertake a needs assessment for the intervention.
2. To adapt *Getting to know Cerebral Palsy* for the Zika context in Brazil.
3. To pilot test the intervention with 2 parent groups in Rio de Janeiro and Salvador.
4. To assess feasibility of the intervention with 4 additional parent groups in Rio de Janeiro and Salvador.

The subsequent chapters present a sequence of papers which have been published in peer-reviewed literature. The first presents the research protocol, which gives an overview of what we wanted to achieve by establishing a programme and how we intended to measure it [1]. The following chapter gives a short overview of the initial Theory of Change (ToC) that was developed in the early stages of the project. This is reflected upon and further elaborated on in the discussion chapter. Following this chapter is a paper that described the needs assessment that was undertaken to better understand the precise need and value add that a family support initiative could provide [2]. After the needs assessment is an overall description of the intervention including the fast track learning approach that we used to hone and adjust the programme from the first groups to the subsequent ones [3]. Finally the feasibility paper is the cumulative analysis of the research and data to ascertain how feasible the programme could be to expand and integrate more widely [4].

CHAPTER 2: FIRST RESEARCH PAPER (published)

Development and assessment of the feasibility of a Zika family support programme: a study protocol.

Figure 8: Room set up ahead of a group session.

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RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congenital Zika syndrome in Brazil during the 2015-16 Zika epidemic		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	Wellcome Open Research		
When was the work published?	13 May 2019		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion	n/a		
Have you retained the copyright for the work?*	**No**	Was the work subject to academic peer review?	**Yes**

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SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	
Please list the paper's authors in the intended authorship order:	

Stage of publication	Choose an item.

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I developed the study protocol and drafted the manuscript, coordinated with coauthors collating their feedback and submitting the paper.

SECTION E

Student Signature	
Date	19th September 2022

Supervisor Signature	
Date	20 September 2022

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 1	 2
version 1 published 13 May 2019	✓ report	✓ report

1 **Michel D. Landry**, Duke University, Durham, USA

2 **Michael J. Griffiths**, University of Liverpool, Liverpool, UK
Alder-Hey Childrens NHS Trust, Liverpool, UK
University of Liverpool, Liverpool, UK

Any reports and responses or comments on the article can be found at the end of the article.

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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Author roles: Duttine A: Conceptualization, Formal Analysis, Methodology, Project Administration, Supervision, Writing – Original Draft Preparation; **Smythe T**: Methodology, Project Administration, Software, Writing – Review & Editing; **Calheiro de Sá MR**: Project Administration, Writing – Review & Editing; **Ferrite S**: Project Administration, Writing – Review & Editing; **Moreira ME**: Conceptualization, Funding Acquisition, Project Administration, Writing – Review & Editing; **Kuper H**: Conceptualization, Funding Acquisition, Project Administration, Supervision, Writing – Review & Editing

Competing interests: One of the researchers (AD) joined the Pan American Health Organisation (PAHO) during the research period. Work on the research study was undertaken outside and separate to his PAHO duties.

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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 attention[1]. 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 anomalies[2]. Prior to that outbreak, Zika was thought to be relatively innocuous, causing few hospitalisations and was not believed to be fatal[3]. 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 proven[5] 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 microcephaly[7]. Microcephaly, therefore may be the tip of the iceberg with regards the wider array of clinical and developmental features[8]. 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 neurodevelopmental 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 prevention[17]. 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 areas[20], 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 families[21].

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 child[23]. 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 trialled[24].

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.

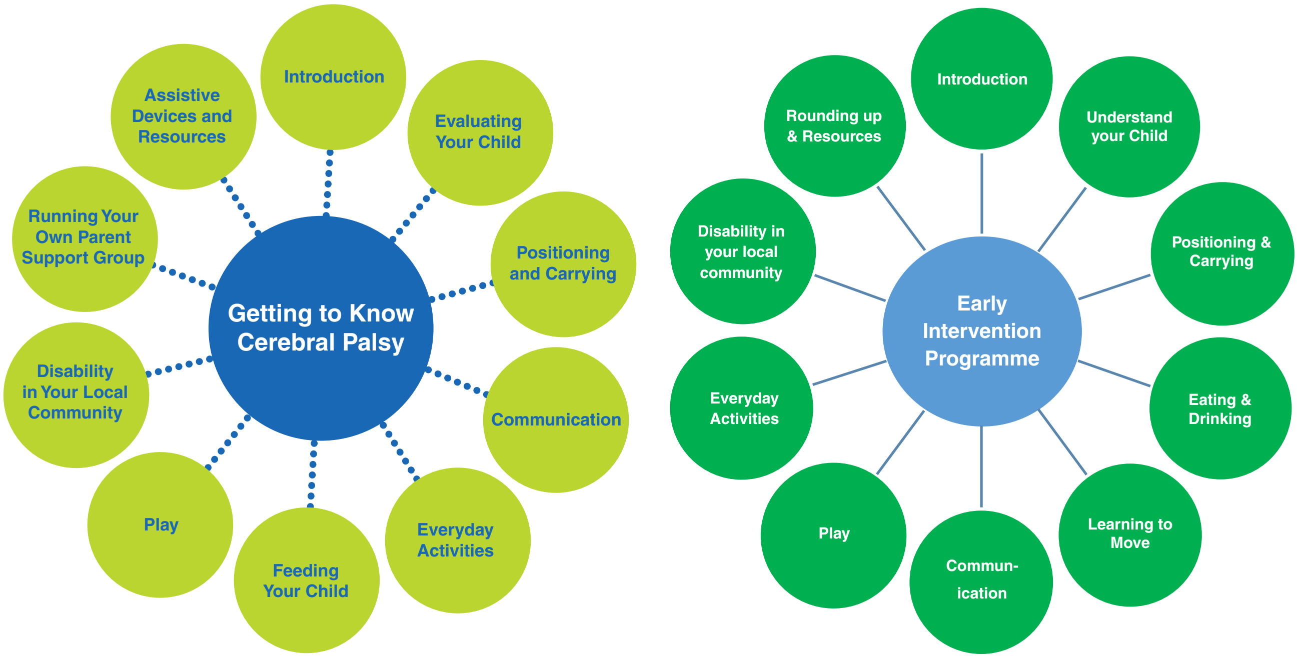

Figure 1. **Structure of Getting to Know Cerebral Palsy and the Early Intervention Programme.**

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 concurrently[25] 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 CZS[9]. 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 form[26], 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 study[25].

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 study[25], funded by the Wellcome Trust, were incorporated into the needs analysis. This included data from semi-structured interviews[27] 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 GTKCP[23] and the EIP[24], 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 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 questionnaires[29] 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 caregiver[1].
- Perceived unmet needs and main goals for the intervention[1].
- The PedsQL™ Family Impact Questionnaire Module[30] (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 achieved[2]
- Satisfaction with programme[2]

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

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[1] Only asked at baseline

[2] Only 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 Module[30] 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 Portuguese[31]. Third, it is also being applied in the sister study[25], 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 a 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.2.2.28380.23686[27].

Researchgate: Consent forms (interviews and questionnaires). https://doi.org/10.13140/RG.2.2.30057.95845[26].

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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CHAPTER 3: THEORY OF CHANGE (ToC)

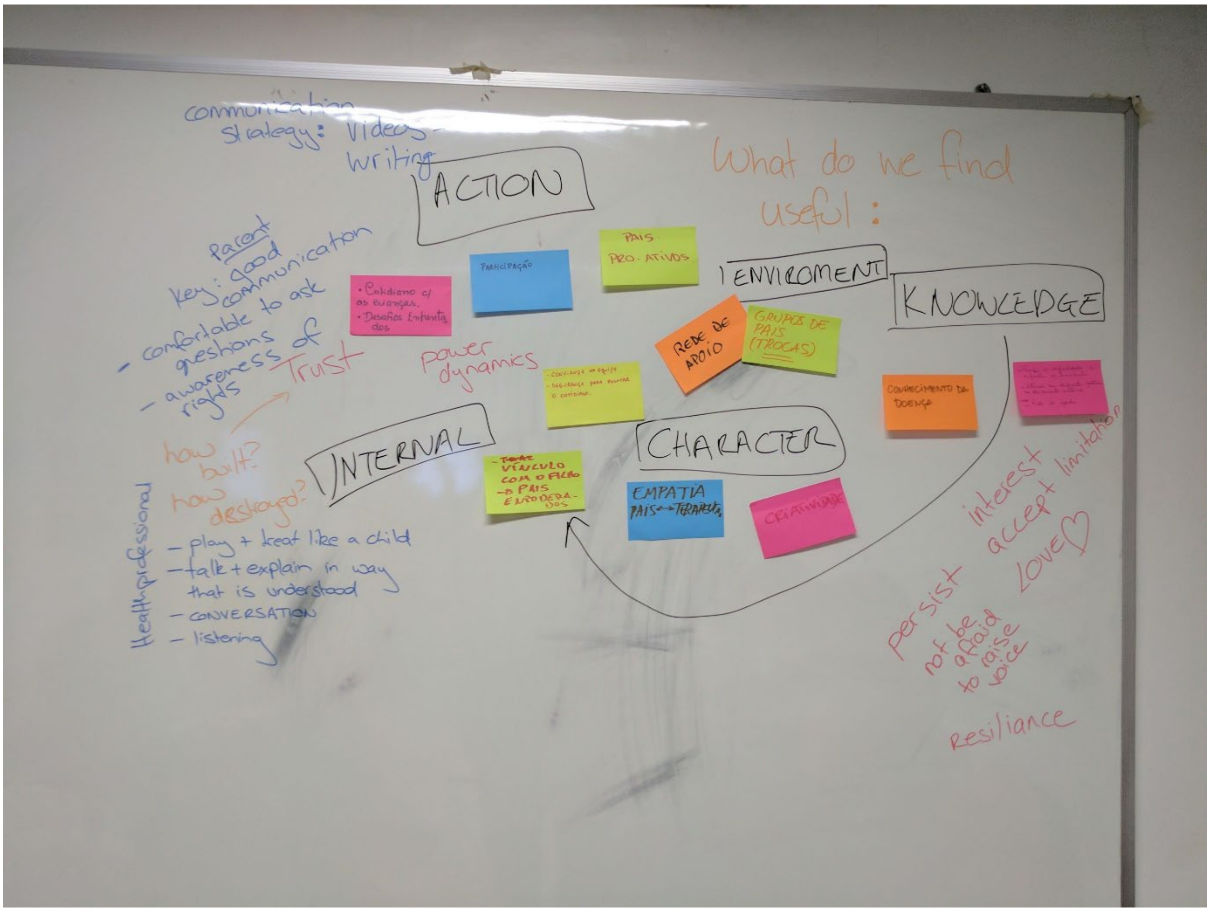

Figure 9: brainstorming session during a working group meeting

After establishing the protocol, several members of the project team including myself, Hannah Kuper (Principal Investigator), Maria Zuurmond (who coordinated the GTKCP programme), Miriam Ribiero Calheiro de Sá and Silvia Ferrite (site coordinators in Brazil) and Joerg Weber (Community Based Rehabilitation expert) developed a ToC for the project to visualise the outcomes and impacts of the intervention. This was undertaken during a workshop in May 2017 and is shown in Figure 10, below. A theory of change is commonly used in public health to describe a project and the change it hopes to achieve [122]. According to De Silva et al [123], development of a theory of change involves *"stakeholders first agree[ing] on the real- world impact they want to achieve. They then identify the causal pathways through which this change can be achieved in that context using the available resources. These are articulated as a series of preconditions leading to outcomes, the order of which can be adjusted as the pathway develops"* (p.4).

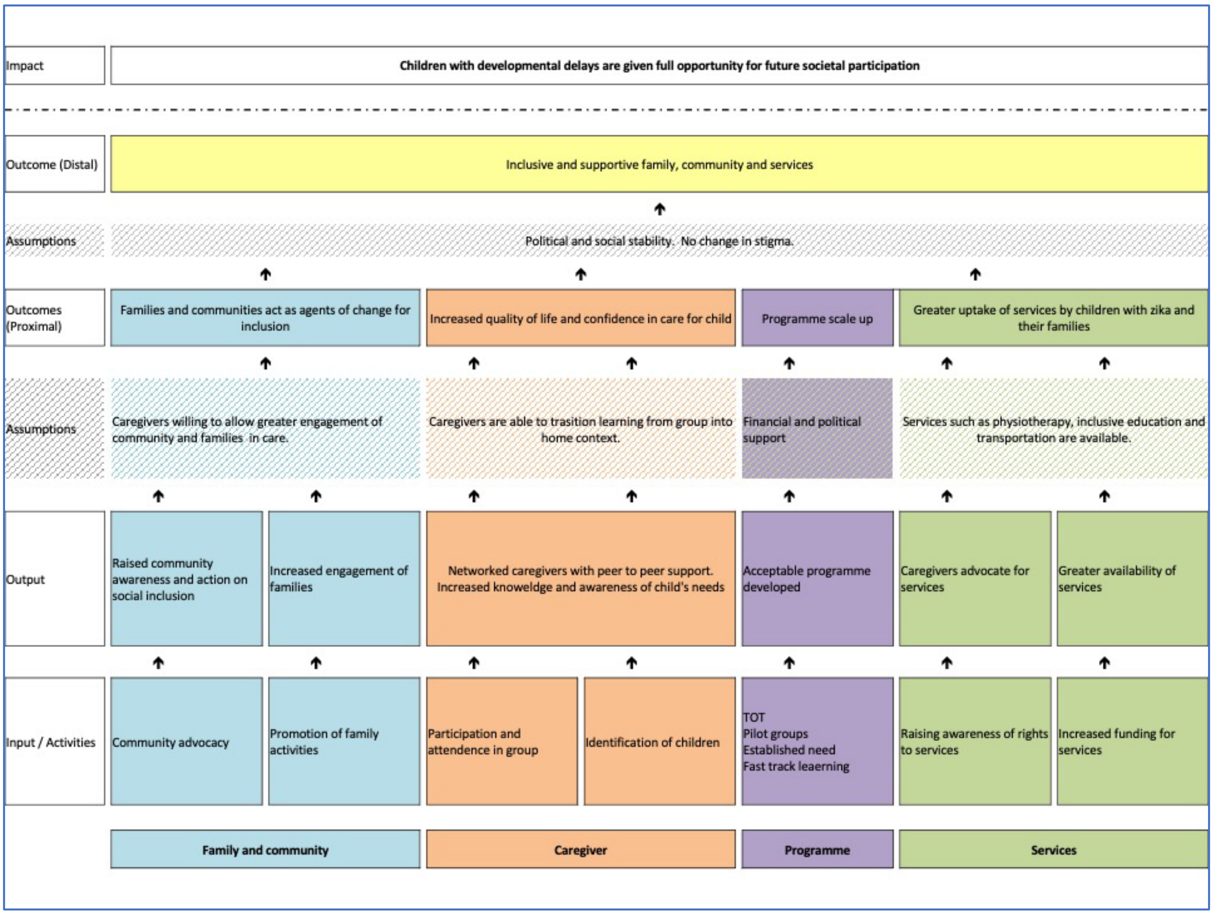

Figure 10: Theory of Change for Juntos

In our ToC, we identified that the impact we intended that *"Children with developmental delays are given full opportunity for future societal participation"*. This would be achieved though creating inclusive and supportive family, community and services (distal outcome). The impact statement, focusing on children with developmental delays rather than just CZS, already reflected the fact that we did not believe that children with CZS should be unique in their specific needs and there was likely to be significant overlap with other NDDs such as CP.

Four columns of individual, but complementary, 'stakeholders' were identified as key to achieving the impact: family and community, caregivers, the Juntos programme, and services such as rehabilitation, education, etc. The two outer columns – family and community and services depend on factors outside of the programme intervention such as health services, community engagement. This is further elaborated on in the adjusted ToC presented in Chapter 7.

At the proximal outcome level for the caregivers' column – which is the extent we may expect to see a tangible direct impact through the intervention – the aim is to achieve *"increased quality of life and confidence to care for child".* At the same level the *Juntos* programme is scaled up, thus having a wider reach and greater impact. The two outer columns, *families, and communities,* would act as agents of change for inclusion (thus demonstrating greater empowerment) and *services* will be available and be utilised by caregivers and children (thus reflecting their greater availability and accessibility).

The lower two rows – output and activities –describe the programme being established and implemented, with the participation of caregivers leading to more peer-to-peer support networks being developed through regular participation in the programme. The outer two columns show that there would be initial efforts for greater advocacy and development of services and actions to raise awareness and sensitise the community being undertaken.

Used in this way, the ToC helps to frame, develop, and ultimately evaluate the programme both impacts in the short and long term.

In developing our ToC, and the revised version shown in Chapter 7, we did not directly request or receive input from the participants. Two mothers of children with CZS, who run a support organisation, were members of the Brazil based working group, however. The reasons for not directly sharing with participants in the focal groups were in part since their feedback in the end of session focus groups and from the interviews were used to adapt and validate the ToC. Furthermore, the complexity and nuance of the ToC may not have been useful for the participants to have directly contributed to this discussion and the feedback from participants relating to the content and delivery of the programme was deemed as more relevant.

CHAPTER 4: SECOND RESEARCH PAPER (published)

Congenital Zika Syndrome—Assessing the Need for a Family Support Programme in Brazil.

Figure 11: Facilitators practice an exercise on feeding during the first training.

RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic.		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	International Journal of Environmental Research and Public Health		
When was the work published?	19 May 2020		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion			
Have you retained the copyright for the work?*	**No***	Was the work subject to academic peer review?	**Yes**

*If yes, please attach evidence of retention. If no, or if the work is being included in its published format, please attach evidence of permission from the copyright holder (publisher or other author) to include this work.

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SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	
Please list the paper's authors in the intended authorship order:	

Stage of publication	Choose an item.

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I led the design, structure, authoring and corrections of the manuscript including collating feedback from co-authors.

SECTION E

Student Signature	
Date	19th September 2022

Supervisor Signature	
Date	20 September 2022

International Journal of
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Article

Congenital Zika Syndrome—Assessing the Need for a Family Support Programme in Brazil

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Abstract: The Zika outbreak in Brazil caused congenital impairments and developmental delays, or Congenital Zika Syndrome (CZS). We sought to ascertain whether a family support programme was needed and, if so, could be adapted from the Getting to Know Cerebral Palsy programme (GTKCP) designed for children with cerebral palsy (CP). We conducted a systematic review of the needs of families of children with CZS or CP in low- and middle-income countries and reviewed the findings of the Social and Economic Impact of Zika study. We undertook a scoping visit to three facilities offering services to children with CZS in Brazil to understand potential utility and adaptability of GTKCP. The literature review showed that caregivers of children with CZS experience challenges in mental health, healthcare access, and quality of life, consistent with the CP literature. The scoping visits demonstrated that most support provided to families was medically orientated and while informal support networks were established, these lacked structure. Caregivers and practitioners expressed an eagerness for more structure community-based family support programmes. A support programme for families of children with CZS in Brazil appeared relevant and needed, and may fill an important gap in the Zika response.

Keywords: congenital zika syndrome; Zika; family support; Brazil; cerebral palsy; community programme

1. Introduction

Since the peak of the Zika epidemic in 2015–2016, the number of new cases of Zika infection and of confirmed Congenital Zika Syndrome (CZS) has gradually declined across the Americas [1], although the virus is now considered endemic to the region [2]. Brazil was the most heavily impacted country, accounting for 47.9% of total cases between 2016 and the end of 2019 [3]. The Ministry of Health in Brazil reports that since 2015, there have been 3474 confirmed and 743 probable cases of CZS with a further 2659 cases under investigation [4]. There are likely to be many more cases that have not been designated as caused by Zika, given the emerging evidence of more mild and later onset impairments and the lack of a reliable retrospective test for Zika.

Int. J. Environ. Res. Public Health **2020**, *17*, 3559; doi:10.3390/ijerph17103559

CZS was defined by Moore et al. [5] as a syndrome of congenital anomalies associated with Zika virus (ZIKV) infection during pregnancy including; severe microcephaly with a partially collapsed skull; thin cerebral cortices with subcortical calcifications; eye anomalies, including macular scarring and focal pigmentary retinal mottling; congenital contractures or a limited range of joint motion; marked hypertonia; and symptoms of extrapyramidal involvement.

While many of these characteristics are common features of congenital central nervous system infections, it was the epidemic of microcephaly and children with severe neurodevelopmental sequelae that initially raised alarm during the Zika epidemic [5]. Subsequently, during the course of the Zika crisis, there was increasing recognition of the broader spectrum of anomalies occurring in children with CZS [6]. For example, while microcephaly was common, it was by no means always present for cases of CZS, and while ophthalmologic manifestations often co-occurred with other neurological features, there were case reports of these occurring in isolation [6].

Emerging evidence on CZS suggested that affected children appeared to have motor abnormalities consistent with internationally accepted definition of cerebral palsy (CP): "A group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain" [7]. The presence of microcephaly in CZS is strongly associated with severe neurological disabilities, such as hearing problems, epilepsy, and learning disabilities, which are also common in children with CP [8,9]. Indeed, in situations such as the Zika epidemic, when a specific cause for the occurrence of cerebral palsy is known, there is international consensus that the term cerebral palsy be used in addition to naming the underlying cause [10].

The focus of research and response to Zika in the Americas has fallen heavily on understanding the pathophysiology of the virus, prevention of the spread of the virus, and development of a vaccine and treatments [11]. In contrast, relatively little focus or specific research considered how to meet the needs of children born with CZS, or the impact of CZS experienced by their families, and they are likely to experience widespread unmet needs.

Interventions are therefore needed to meet the broader needs of children with CZS. A number of family-based support programmes have been established for families of children with neurodevelopmental disability in low- and middle-income contexts to respond to the unmet needs experienced by these families [12,13]. One such programme is Getting to Know Cerebral Palsy (GTKCP), developed to educate and empower caregivers in the care of their child [14]. GTKCP is a community based participatory programme for caregivers in a support group setting, and has been shown to improve caregiver quality of life and knowledge and confidence in caring for a child [15,16]. By targeting the caregivers, the programme aims to have an impact on the long-term health, wellbeing, and participation of children with cerebral palsy. An Early Intervention Programme (EIP) has also been developed from GTKCP to address the needs of younger infants [17]. Considering the commonalities of CZS and CP, it may be plausible to use existing CP programmes as the basis for new interventions. Therefore, the aim of the current study was to ascertain whether a similarly structured family support programme to GTKCP was (a) needed and (b) relevant for the post-Zika Brazilian context.

Specific questions that this study sought to answer were:

a) What are the needs of families of children with CZS (or related conditions) in Brazil, and are they being met by the existing support services?;
b) Would a family support programme be potentially useful in the post-Zika context in Brazil?;
c) Are the similarities between CP and CZS sufficient to suggest that GTCKP/EIP could be used as a basis for a Brazil family support intervention?

2. Materials and Methods

2.1. Data Extraction

Data were extracted to a custom-made spreadsheet in Microsoft Excel. We did not perform a meta-analysis, as the data were highly heterogeneous and included qualitative and quantitative data.

2.2. Ethical Approval

Ethics approval was acquired in Brazil (IFF/FIOCRUZ-RJ/MS 2.183.547) and the UK (LSHTM Ethics number 13608).

2.3. Methods

The methods involved 3 processes:

2.3.1. Systematic review on unmet needs of families of children with CZS and CP

A systematic search of the literature was performed in June 2017 to identify articles that considered the unmet needs of families of children with CZS [18]. The review was led by one researcher (AC), with a second researcher (AD) acting in a supervisory capacity. We followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Statement [19] in the conduct of this systematic review. We searched CINAHL Plus, EMBASE, MEDLINE, Psychinfo, and PubMed, and search terms are included in Appendix A1. Inclusion criteria were: Articles related to the needs or wellbeing of families with children with CZS, published from 2000–2017 in peer reviewed journals, full text available in English. No exclusion criteria were set in terms of study design, due to the lack of research into the topic at the time.

Few articles were identified as being relevant. Therefore, a second search was conducted, expanding search terms to include articles related to CP. Inclusion criteria were the same as above, however this time related to the needs or wellbeing of mothers and families with infants or children with CP and only research set in lower middle- or upper middle-income countries was considered.

In both parts of the search, articles were first reviewed for relevance by title, followed by abstracts and full texts by one researcher (AC). Duplicates and articles that did not meet inclusion criteria were excluded. Identified texts were confirmed for relevance by a second researcher (AD). Once identified, references were saved and managed using Mendeley Web and Mendeley Desktop. A PRISMA flow chart is included in Appendix B.

2.3.2. Findings from the Social and Economic Impact of Zika Study

Emerging themes that were being raised as part of a parallel study, *The Social and Economic Impacts of CZS on Families and Caregivers* [20] were reviewed. This was a mixed methods study, conducted in Recife and Rio de Janeiro. It included in-depth qualitative interviews in each setting with approximately 30 families of children with CZS and 10–12 healthcare providers, as well as a case-control study of 163 children with CZS and 324 unaffected controls. Through both approaches, information was collected on economic, mental health, and social impacts, using standardised tools in the quantitative component (e.g., Depression Anxiety Stress Scales (DASS) for depression, anxiety and depression, and Medical Outcomes Study Social Support Scale for social support). An additional statistical analysis was conducted using data from this study, comparing the results of the Pediatric Quality of Life Inventory (PedsQL) Family Impact Module between a subset of participants from the study—155 mothers of children with CZS and 47 mothers of unaffected children—in order to assess the broader impacts of CZS on the quality of life of families. For logistical reasons, the PedsQL data were not collected from all participants. The PedsQL Family Impact Module is a questionnaire that measures parent self-reported physical, emotional, social, and cognitive functioning, communication, and worry. The module also measures parent-reported family daily activities and family relationships. It is scored on a 5-point Likert scale where 0 is "never" and 4 is "almost always". The results are then transformed to a 0–100 scale to enable/allow scoring and data analysis. We used the PedsQL Family Impact Module to compare the parent self-reported physical, emotional, social, and cognitive functioning, and communication and worry between mothers of children with CZS and mothers of children with unaffected children.

2.3.3. Scoping visit in Brazil

In April 2017, 3 researchers (AD, MZ, HK) undertook a week-long visit to Brazil. The researchers visited a range of facilities offering services to children with CZS and their families. The sites were identified and selected by the local research partners in Brazil (SF, MS, EM) and included, a tertiary facility in Rio de Janeiro (Instituto Fernandes Figueira (IFF)), which offers clinical services including habilitation and psychosocial support in a hospital based setting in Central Rio de Janeiro; the Altino Ventura Foundation, an NGO in Recife providing support and care to families, including group programmes, in a hospital-based setting; and Associação aBRAÇO a Microcefalia, a parent support programme in Salvador, Bahia which offered twice monthly meetings of carers, including both formal lectures and social activities, as well as therapeutic support and donations (e.g., nappies/diapers). At each site, the researchers consulted with caregivers of affected children, and health care professionals (doctors, psychologists, physiotherapists, occupational therapists, social workers, speech therapists, and lactation specialists). Caregivers (n = 7) were consulted about the services offered, the main perceived barriers and gaps, and level of interest in a formal parent support programme. Consultations with healthcare professionals (n = 12) included mapping the flow of service delivery to meet child and family support needs within the existing structures of Brazil, in order to better understand and contextualise how services are currently delivered to families of children with CZS.

3. Results

3.1. Findings from the Literature Review of Families and Caregivers of Children with CZS and CP

Only seven eligible papers were identified that assessed the needs of children with CZS [21–27], and 31 eligible papers focussed on the needs of children with CP [18]. Tables 1 and 2 below summarise the findings.

Table 1. Overview of findings from Congenital Zika Syndrome articles.

Article	Country	Measures Used	Overall Findings/Topics
Anxiety, depression, and quality of life in mothers of newborns with microcephaly and presumed congenital Zika virus infection [21]	Brazil	World Health Organisation Quality of Life-BREF (WHOQoL-BREF)	Lower scores in psychosocial domain of WHOQoL-BREF of women with babies with microcephaly in first 24 h after birth.
Babies with microcephaly in Brazil are struggling to access care [22]	Brazil	Anecdotal evidence	Struggle of families to access care, transportation, investigation, and medication. Financial cost of bringing up an infant with congenital Zika syndrome.
Congenital Zika virus infection: A developmental- behavioural perspective [23]	Brazil	Anecdotal evidence and recommendations	Stigma surrounding congenital zika syndrome in Brazil. Broad range of outcomes and potential interventions needed.
Engaging human rights in the response to the evolving Zika virus epidemic [24]	Brazil	Relationship between human rights principles and Zika response with relation to discrimination, participation, accountability of Brazilian health system, equity	Health system may need to divert resources to areas of greatest need, given that Zika was concentrated in areas that may have less health providers. Need to address structural and social determinants of health.
Integrated reproductive health: The Zika virus [25]	Brazil	Anecdotal evidence	Psychological impact on women and need for support and communication. Social inequities within Brazil.
Infants with congenital zika virus infection: A new challenge for early intervention professionals [27]	Brazil	Recent literature and recommendations	Social stigma and media attention may affect parents' psychological wellbeing. Poor sleep patterns of infants may contribute to poor emotional health of parents. Parents may need education and explanation of child's condition. Adequate psychosocial services will be necessary, as well as possibly respite opportunities. Consultation with lactation specialists may be useful.
Brazil struggles to cope with zika epidemic [26]	Brazil	Anecdotal evidence	Highlights lack of available finances and services in the Brazilian health system.

Table 2. Overview of findings of CP studies [18].

Article	Country	Scale or Questionnaire Used	Main Findings
Understanding the lives of caregivers of children with cerebral palsy in rural Bangladesh: Use of mixed methods	Bangladesh	PedsQL Family Impact Questionnaire	Lower quality of life in all domains of PedsQL in families of children with CP ($p < 0.001$). Parents experienced fatigue, stigma, lack of social support.
Assessment of family environment and needs of families who have children with cerebral palsy	Turkey	Family Needs Score (FNS) and Family Environment Score (FES)	Vast majority (91.8%) of primary caregivers were mothers. More families cited information needs (84.3%) than support or financial needs. Many families had assistance from elders.
An investigation of parents' problems according to motor functional level of children with cerebral palsy	Turkey	Author written questionnaire	Families with children with more severe CP had more problems than those with mild CP (no p-value given). Major difficulties were economic, lack of health services, and communication. Many families had assistance from elders.
Comparative quality of life of Nigerian caregivers of children with cerebral palsy	Nigeria	World Health Organisation Quality of Life score (WHOQoL-BREF), Gross Motor Functional Classification System (GMFCS)	Caregivers of children with CP have a lower quality of life than those without children with CP ($p = 0.003$). Quality of life scores improved over time as children's motor function improved, suggesting that early intervention and therapy may help with caregiver's quality of life long term. No significant correlation between child's GMFCS and severity of depression ($p = 0.339$).
Depression in mothers of children with cerebral palsy and its relation to severity and type of cerebral palsy	Iran	Beck Depression Inventory-II (BDI-II), GMFCS	Greater risk of mothers caring for children with CP having depression ($p = 0.003$). No significant correlation between the GMFCS and severity of depression.
Depression and anxiety levels in mothers of children with cerebral palsy: A controlled study	Turkey	Beck Depression Inventory (BDI), Beck Anxiety Inventory (BAI)	Higher levels of depression and anxiety in mothers of children with CP ($p = <0.001$). Statistically significant difference in effect of speech defects and higher GMFCS score on mothers' depression ($p < 0.05$ with 95% CI) based on logistic regression.
Depression in parents of children with cerebral palsy in Bosnia and Herzegovina	Bosnia and Herzegovina	Zung self-evaluated method for depression	No significant difference in levels of depression between mothers and fathers of children with CP, and mothers of healthy controls ($p = 0.09$).
Factors associated with caregiver burden among caregivers of children with cerebral palsy in Sri Lanka	Sri Lanka	WHOQoL-BREF 'Caregiver Difficulties Scale' (CDS)	Majority of caregivers (97%) were mothers. Majority of caregivers were from a rural area and low socioeconomic background (72% and 70% respectively). Living in a rural area ($p = 0.001$), having a lower income ($p < 0.023$), male sex of the child ($p = 0.017$), and more significant functional impairment of child ($p < 0.001$) were associated with a higher caregiver burden in multivariate analysis. Social support was associated with a lower caregiver burden ($p < 0.001$).
Functional priorities reported by parents of children with cerebral palsy	Brazil	Questionnaire	In all age groups, 'personal care' was the highest rated functional goal by parents (42.99%–52.38%). In 3–6 year olds, play was second highest rated (20.56%), in 7–10 year olds and in 11–16 year olds, school was the second highest rated (23.16% and 22.22% respectively).
Higher Levels of Caregiver Strain Perceived by Indian Mothers of Children and Young Adults with Cerebral Palsy Who have Limited Self-Mobility	India	Caregiver Strain Index (CS)	Caregivers of children with higher scores on GMFCS had higher levels of caregiver strain ($p < 0.01$).
Life quality among mothers of children with cerebral palsy living in Armenia	Armenia	BDI-II and Norakidze's modification of Taylor manifest anxiety scale	High levels of depression (74%) and anxiety (95%) in mothers of children with CP. Mothers with lower level of education had higher rates of anxiety.
Mental health and quality of life of caregivers of individuals with cerebral palsy in a community-based rehabilitation programme in rural Karnataka	India	General Health Questionnaire (GHQ) WHOQoL-BREF	Majority (87%) of caregivers were mothers. No statistically significant difference in GHQ-28 score in relation to functional status of child. No statistically significant difference in children's needs in relation to mothers' mental health score.

Table 2. *Cont.*

Article	Country	Scale or Questionnaire Used	Main Findings
Coping with stress and adaptation in mothers of children with cerebral palsy	Serbia	Family Crisis Oriented Personal Evaluation Scale (F-COPES)	No difference in methods of coping between urban and rural mothers, reframing was the strategy most commonly used. Only statistically significant difference in methods of coping in relation to severity of child's functional impairment was use of institutions in more severe impairment.
Fatigue in the mothers of children with cerebral palsy	Turkey	Fatigue Symptom Inventory (FSI), Beck Depression Scale (BDS), and Nottingham Health Profile (NHP)	Mothers of children with CP scored higher in all groups of FSI ($p < 0.00001$). Mothers of children with CP had higher scores on BDS ($p < 0.00001$). Mothers of children with CP had higher scores on BDS ($p < 0.026$ or less in all domains). No impact of GMFCS on outcomes in mother when regression analysis applied. Fatigue correlated with higher NHP and BDS scores.
Predictors of stress in mothers of children with cerebral palsy in Bangladesh	Bangladesh	Judson Scale, Family Support Index (FSI)	Higher levels of stress in mothers living in rural areas ($p = 0.02$). Higher levels of household income associated with lower levels of stress ($p = 0.02$). Level of child's functional impairment not associate with higher levels of stress. Child's behavioural issues (including sleep, bet wetting, hyperactivity) associated with a higher level of stress (goodness of fit 75.46%).
Psychological distress and perceived support among Jordanian parents living with a child with cerebral palsy: A cross sectional study	Jordan	GMFCS, Perceived Stress Scale (PSS), BDI, Strengths and Difficulties Questionnaire (SDQ), and Multidimensional Scale of Perceived Social Support (MSPSS)	Many parents of children with CP have perceived levels of stress. Parents of children with higher GMFCS had higher levels of stress ($p = 0.03$). Parents of children with more behavioural issues had higher levels of perceived stress. Parents with lower social supports had higher levels of stress ($p < 0.0005$).
Psychological adversities and depression in mothers of children with cerebral palsy in Nigeria	Nigeria	Psychosocial Adversity Scale (PAS) and Patient Health Questionnaire (PHQ)	Additional psychosocial stressors associated with depression (all except unemployment and mother's education). Majority of mothers (89%) had some degree of depression.
Quality of life in mothers of children with cerebral palsy: The role of children's gross motor function	Iran	Short Form Health Survey (SF-36), GMFCS	Mothers of children with better GMFCS had better QoL scores. When compared with general population mean, mothers of children with CP has statistically significant lower scores in all QoL domains.
Quality of life in parents/caretakers of children with cerebral palsy in Kampong Cham, Cambodia	Cambodia	Comprehensive Quality of Life Scale (ComQOL-A5) scores	Lowest scoring QoL domains were health, emotional wellbeing, and material well-being.
Social support provided to caregivers of children with cerebral palsy	Brazil	Sarason's Social Support Questionnaire (SSQ)	Majority of caregivers (88%) are mothers. Husband, mother, and brother are those cited most frequently as sources of social support.
The effect of having a children with cerebral palsy on quality of life, burn-out, depression and anxiety scores: A comparative study	Turkey	WHOQoL-BREF, GMFCS	Higher levels of depression in CP group compared to control group (58.0% vs. 46.7%). Higher levels of anxiety in CP group compared to control group (71.4% vs. 51.7%). Highest scores in WHOQoL were in domains of physical, psychosocial, and environment. Correlation between higher GMFCS and higher total WHOQoL and BDI scores ($p = 0.04$ and 0.01 respectively).
Quality of life and anticipatory grieving among parents living with a child with cerebral palsy	Jordan	Marwitand Meuser Caregiver Inventory Quality of Life Index	62.7% reported stress, 78.3% reported drastic life changes, 71.0% reported anxiety, 73.4% reported excellent family support. Personal sacrifice burden score highest. Negative correlation between anticipatory grief and QoL scores ($p < 0.0005$).

Table 2. *Cont.*

Article	Country	Scale or Questionnaire Used	Main Findings
Psychosocial impact of caring for children with cerebral palsy on the family in a developing country	Nigeria	Impact on Family Scale (IFS) and GMFCS	Majority of caregivers (80.3%) were mothers. Although 46.2% of CP children had speech impairments, only 2.6% received speech therapy. Correlation between higher GMFCS and higher IOF scores, but not statistically significant ($p = 0.16$). Higher IOF scores in families of children with CP ($p = 0.000$).
Psychosocial challenges for parents of children with cerebral palsy: A qualitative study	Iran	Semi-structured interview	Lack of financial support, transportation, medical services. Sense of guilt, stigma. Lack of social support.
Investigation of quality of life in mothers of children with cerebral palsy in Iran: Association with socio-economic status, marital satisfaction and fatigue	Iran	WHOQoL-BREF, Socioeconomic Status Questionnaire (SES), Index of Marital Satisfaction (IMS) and Fatigue Severity Scale-Persian (FSS-P)	Mothers in CP group has lower SES categories. Mothers in CP group had higher fatigue levels ($p < 0.001$) and higher marital dissatisfaction ($p < 0.001$). Mothers in CP group had lower QoL scores in all domains ($p < 0.001$).
Frequency and severity of depression in mothers of cerebral palsy children	Pakistan	Siddiqui -Shah Depression Scale (SSDS)	50.62% of mothers had depression.
Experiences shared through the interviews from fifteen mothers of children with cerebral palsy, sexuality and disability	Turkey	Semi-structured questionnaire	Majority of caregivers were mothers, often blamed for child's condition. Out of 12 who had other children, 3 reported difficulties in sibling relationships. All mothers reported financial difficulties. Lack of suitable support for child's education. Concerns for child's future.
An evaluation of quality of life of mothers of children with cerebral palsy	Turkey	Turkish version of SF-36	Negative correlation between SF-36 QoL scores and GMFCS; significant in domains of role physical ($p = 0.001$), bodily pain ($p = 0.023$), general health ($p = 0.031$), social functioning ($p = 0.0320$, role emotional ($p = 0.003$), and mental health ($p = 0.004$). Statistically significant difference between mothers of children with CP and controls in domains of mental health ($p = 0.002$), social functioning ($p = 0.002$), general health ($p = 0.001$), bodily pain ($p = 0.005$), and role physical ($p = 0.008$).
Assessment of the quality of life of mothers of children with cerebral palsy (primary caregivers)	Turkey	Nottingham Health Profile-1, BDI, BAI, GMFCS	Higher NHP score in mothers of children with CP in sleep, energy, social isolation ($p = 0.000$), pain ($p = 0.007$), physical activity ($p = 0.004$), and emotional reactions ($p = 0.001$). BDI scores higher in mothers of children with CP ($p = 0.000$). 78.2% of mothers of children with CP had depression compared with 21.7% in control group.
Coping strategies and resolution in mothers of children with cerebral palsy	Serbia	Reaction to Diagnosis Interview (RDI) and classification system used and modified version of F-COPES and Functional Status II (FS-II)	59% mothers remained unresolved. Reframing was the coping strategy used most, followed by passive appraisal. No difference between resolution and non-resolution depending on coping strategy. Mothers with children with better functional status who utilised institutional support had better resolution.
Depression in mothers of children with cerebral palsy and other related factors in Turkey: A controlled study	Turkey	BDI, GMFCS	More mothers in the CP group (61.2%) were depressed compared with control group (36%). Depression did not vary depending on CP type. Depression correlated with speech deficits ($p = 0.036$). No correlation between GMFCS level I, II, III and groups IV, V, and depression ($p = 0.260$). Higher BDI score correlated with lower household income ($r = -0.384$, $p = 0.007$).

Studies focussing on psychosocial aspects of caring for a child with CZS or CP found higher levels of anxiety and depression and poorer Quality of Life (QoL) scores in primary caregivers of children with these conditions, which was usually the mother. CZS literature was still emerging, but one study provided specific information on the impact on the psychosocial domain of caregivers with CZS had interviewed mothers within 24 h of birth of a child [21]. Lack of sleep of parents of children with CZS due to severe cerebral irritation noted in these children may compound psychosocial distress [27].

Expanding to literature on CP, we noted findings around financial hardships, difficulties with transport and services, and stigma. These challenges were associated with a significant impact on the caregivers' psychosocial wellbeing. Difficulties were also reported by caregivers of children with CP in terms of lack of access to services, in particular on account of distance, cost and lack of availability. The information needs of parents were cited in one study as being greater than financial or other support needs [28]. Of the different types of information gaps, information about the 'child's condition' and information about the 'institutions that the child can benefit from' were the two most frequently reported [28].

3.2. Findings from Social and Economic Study

The social and economic impacts of the CZS study also highlighted the needs of parents of children affected by CZS. The quantitative data showed that mothers of children with CZS were more likely to experience depression, anxiety, and stress than mothers of unaffected children [29]. Mothers of children with CZS reporting low social support were particularly likely to experience depression, anxiety, and stress, indicating that social support may buffer adverse mental health effects.

This study also showed through qualitative and quantitative data that affected children had very high health care needs, and had to make frequent visits to services to attend to specific conditions related to CZS (neurology appointments, physiotherapy etc.), co-morbidities (e.g., chest infection, epileptic seizure) and routine health care needs (e.g., vaccines) [30]. Services were often far away, fragmented, and uncoordinated. As a consequence, healthcare professionals felt that it was difficult to adequately meet the holistic needs of these children and their families. Visits by families to therapy and medical appointments focussed almost exclusively on the therapeutic or medical interventions and parents felt they had little opportunity to discuss their own needs. Parents also reported issues of distrust with healthcare professionals based on difficulties in communication with the health care provider.

Some identified gaps in services included provision for children with less severe developmental disability and delays. Concerns were expressed by several health professionals about children with mild CZS being lost from the system, either due to parents not believing that the impairments warranted intervention or because the systems in place had been established for more severe cases. In addition, families reported high household expenditures to meet the healthcare needs of their child. This impact was particularly difficult since the families were on average poorer than families of a child with a disability (paper in submission).

The PedsQL analyses conducted for this paper showed that mothers of children with CZS had worse quality of life scores across all domains (Table 3). These differences reached statistical significance in relation to problems with communication and problems with worrying, showing that these are important needs that should be addressed.

Table 3. The results of the PedsQL Family Impact Module comparing mothers of children with CZS to mothers of unaffected children.

Dimensions of PedsQL	Mothers of children with CZS (n = 155)	Mothers of Children with Unaffected Children (n = 47)	*p*-Value (*t*-Test)
Physical Functioning	53.6 (1.8)	54.6 (3.5)	0.39
Emotional Functioning	57.6 (1.9)	62.1 (3.3)	0.13
Social Functioning	56.7 (2.3)	61.6 (3.9)	0.15
Cognitive Functioning	60.5 (2.0)	66.1 (3.7)	0.09
Communication	58.9 (2.4)	71.6 (4.5)	0.006
Worry	33.6 (1.4)	38.9 (3.3)	0.04
Daily Activities	35.2 (2.1)	38.3 (4.2)	0.24
Family Relationships	60.5 (2.2)	58.0 (4.6)	0.70
Total	52.5 (1.3)	56.4 (2.7)	0.08

Note: Mean scores out of 100, with a higher number equating to higher reported quality of life; Standard deviation noted in parentheses.

3.3. Findings from Scoping Visit

Our mapping of services indicated that the response to CZS had a largely medical/therapy-based focus. The main structure in Brazil is the provision of interventions at specialised tertiary level health centres, which tended to be based in large urban settings (e.g., Rio, Salvador, Recife). The Brazilian Unified Health System, Sistema Único de Saúde, has an extensive health network reaching out to primary level settings. However, rehabilitation teams are not always available at the primary level, either due to lack of specialised rehabilitation staff or services not being developed. Those staff that are based at primary level may be more likely to be generalists, and lack the specialised paediatric knowledge and experience required to meet the needs of many of the children with CZS.

Other forms of support were available for some families in some settings. Similar grassroot family-support initiatives to those seen at Associação aBRAÇO a Microcefalia existed in other settings, and it was reported by both caregivers and healthcare providers that families often made informal social networks to be connected with others. These initiatives varied in focus and structure. Some groups had more of a focus on advocacy and promoting children's rights, rather than on caregiver education and support. Additionally, mothers almost universally reported being part of WhatsApp groups with other carers, which provided some social and emotional support, but was unstructured and on an ad hoc basis.

Health professionals reported that the concept and approach of GTKCP was highly relevant for the situation being faced by many Brazilian families. Most caregivers said that having support groups would be acceptable to them and welcomed the idea of having an opportunity to learn and share from one another. However, practical considerations were also raised with respect to the parent support groups. The security situation, particularly in Rio de Janeiro, was a concern, because levels of urban violence meant that the logistics of safely planning community-based interventions were more complicated. There was also awareness that additional programmes should complement clinical services and ideally be integrated with caregiver networks already in place.

4. Discussion

This paper aimed to ascertain whether a structured family support programme to GTKCP was needed and relevant for the post-Zika Brazilian context. Specifically, this mixed-method study generated evidence to respond to the three following questions posed, as follows:

a) What are the needs of families of children with CZS (or related conditions) in Brazil, and are they being met by the existing support services?

We found a need for caregivers to receive a higher level of informational, psychosocial, and emotional support than was currently available. Health and specialist medical needs, including rehabilitation, were largely available and being accessed by families. However, meeting the health care and specialist needs of children was onerous, especially given the need to travel long distances to access the relevant services. This finding is echoed in recent publications, which have described several family impacts, notably isolation, stress, lack of access to services, and powerlessness [31] and the importance of a holistic approach to meet the broad needs of children with neurodevelopmental disabilities and caregivers [32].

b) Would a family support programme be potentially useful in the post Zika context in Brazil?

A family support programme may be useful in the post Zika context in Brazil as a complement to clinical services and existing caregiver groups and networks. Both caregivers and health professionals agreed that a support programme could be an important adjunct to the existing services and fill gaps in the existing support mechanism, such as a focus on mental health of caregivers and holistic needs of children. The needs of caregivers of children with less severe or later onset impairments are an important group to consider.

Parenting programmes have been shown to have a positive impact on self-efficacy for parents of children with developmental disabilities [33]. Targeting parent and caregiver skills and behaviour can have an important foundational impact on child health and wellbeing. If a family support programme in Brazil can have some similar impacts on children and caregivers as GTKCP has shown [15,16,34], these foundations can potentially impact more long-term health and wellbeing outcomes of children with CZS across the life-course.

c) Are the similarities between CP and CZS sufficient to suggest that GTCKP/EIP could be used as a basis for a Brazil family support intervention?

The evidence on CZS that was emerging at the time of this study highlighted the similarities of the physical presentation of CZS to those of CP. More recent literature has further described the overlapping between CZS and CP [35–37]. The review of the literature and findings of the social and economic impact study also suggested that there are similarities between CP and CZS, particularly in terms of needs and unmet needs of the caregiver. This led us to suggest that the GTKCP/EIP programmes had potential utility for being a basis of a programme in Brazil. However, it was also clear that adaptations to the existing programmes (GTKCP and EIP) to the context of Zika and Brazil would be required, to cater to the specific circumstances in Brazil and to address the mental health impacts on caregivers.

The needs of children with developmental disabilities will change over their life course, and consequently, family support programmes need to be adapted for different age groups. In the early years, programmes may need to focus on maximising development of the child, and supporting carers in looking after the child. As children reach the age of 5 or more, attention needs to be given to helping carers support their child's inclusion in education. In the next stage, as children with developmental disabilities reach adulthood, focus of programmes should shift towards supporting independent living, employment, and maintaining health and function. Throughout the life course, programmes should address social inclusion and supporting carers, especially with respect to mental health.

Strengths of the mixed methods approach of the study include obtaining data from a range of sources and ensuring inclusion of different perspectives to create an overview of the needs of caregivers. For instance, data from the parallel study on social and economic impacts of Zika provided information that helped to confirm and elaborate findings from the scoping visit. Limitations include the fact that the Brazil scoping visit was a rapid, pragmatic stakeholder assessment in the setting of the epidemic, rather than a detailed qualitative evaluation, and a limited number of facilities were visited. Other limitations were that double screening was not undertaken within the systematic review, and the PedsQL was not collected on the full sample of cases and controls, which may be introduced biases. Finally, data on CZS was newly emerging when the literature review was undertaken, therefore giving a narrow range of useful information specific to caregiver needs relating to CZS.

5. Conclusions

A family support programme could potentially fill a gap in the range of services provided in Brazil in the wake of the Zika outbreak and could address unmet holistic needs of families of children affected by CZS. The literature review coupled with site specific needs assessment demonstrated an important gap in support for children and families affected by CZS. Further implementation research regarding contextual design and adaptation of psychosocial support programmes for caregivers of children with CZS and other neurodevelopmental disabilities at community level in low- and middle-income countries is urgently needed. However, given the similarities of CP and CZS, there could be justification to use GTCKP as a basis for a Brazil family support intervention if cultural and practical adaptations to the existing programme are made.

Author Contributions: Conceptualization, A.D. and H.K.; methodology, A.D., H.K., M.E.M.; validation, H.K., T.S., M.Z., M.E.M., S.F., M.R.C.d.S., A.C., K.M.; formal analysis, A.D., A.C., H.K.; investigation, A.D., A.C., M.Z., K.M., H.K.; resources, A.D., H.K., M.E.M.; data curation, A.D., A.C., H.K. writing—original draft preparation, AD.; writing—review and editing, H.K., T.S., M.Z., M.E.M., S.F., M.R.C.d.S., A.C., K.M.; visualization, A.D., A.C., H.K.; supervision, A.D., H.K., M.E.M.; project administration, A.D., M.R.C.d.S., S.F.; funding acquisition, H.K. All authors have read and agreed to the published version of the manuscript.

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Conflicts of Interest: One of the researchers (AD) joined the Pan American Health Organisation (PAHO) during the research period. Work on the research study was undertaken outside and separate to his PAHO duties. Otherwise the authors declare no conflict of interest. The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript, or in the decision to publish the results.

Appendix A Search Terms Used for Literature Review

Appendix A.1 Search on CZS

Zika virus; Congenital zika syndrome; Zika + family needs; Zika + family; Zika + parent; Zika + parental needs; Zika + psychosocial; Congenital zika syndrome + cost; Congenital zika syndrome + economic impact.

Appendix A.2 Search on CP

Cerebral palsy + family needs; Cerebral palsy + family; Cerebral palsy + parents; Cerebral palsy + parental needs; Cerebral palsy + psychosocial; Cerebral palsy + economic impact; Cerebral palsy + cost.

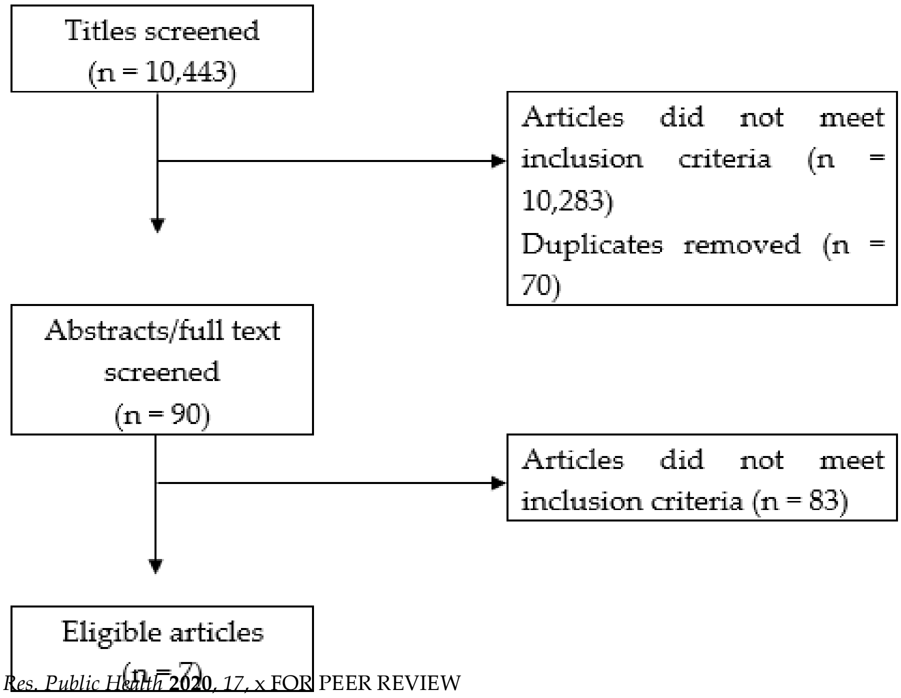
Appendix A.2 Search on CP

Cerebral palsy + family needs; Cerebral palsy + family; Cerebral palsy + parents; Cerebral palsy + parental needs; Cerebral palsy + psychosocial; Cerebral palsy + economic impact; Cerebral palsy + cost.

Appendix B. PRISMA Flow Charts of the CZS and CP Literature Review.

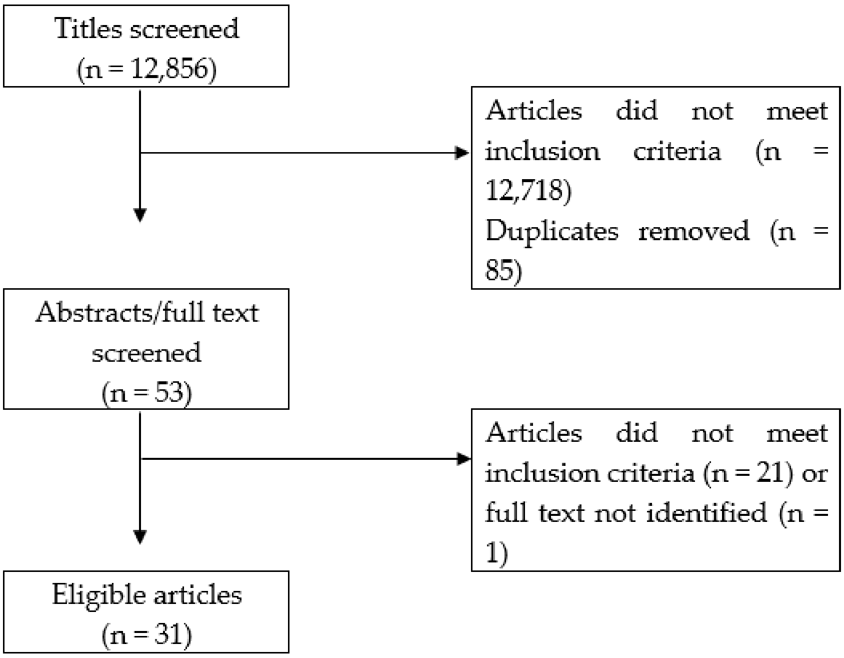
Appendix B.1 Search on CZS

Appendix B.1 Search on CZS



Appendix B.2 Search on CP.

Appendix B.2 Search on CP



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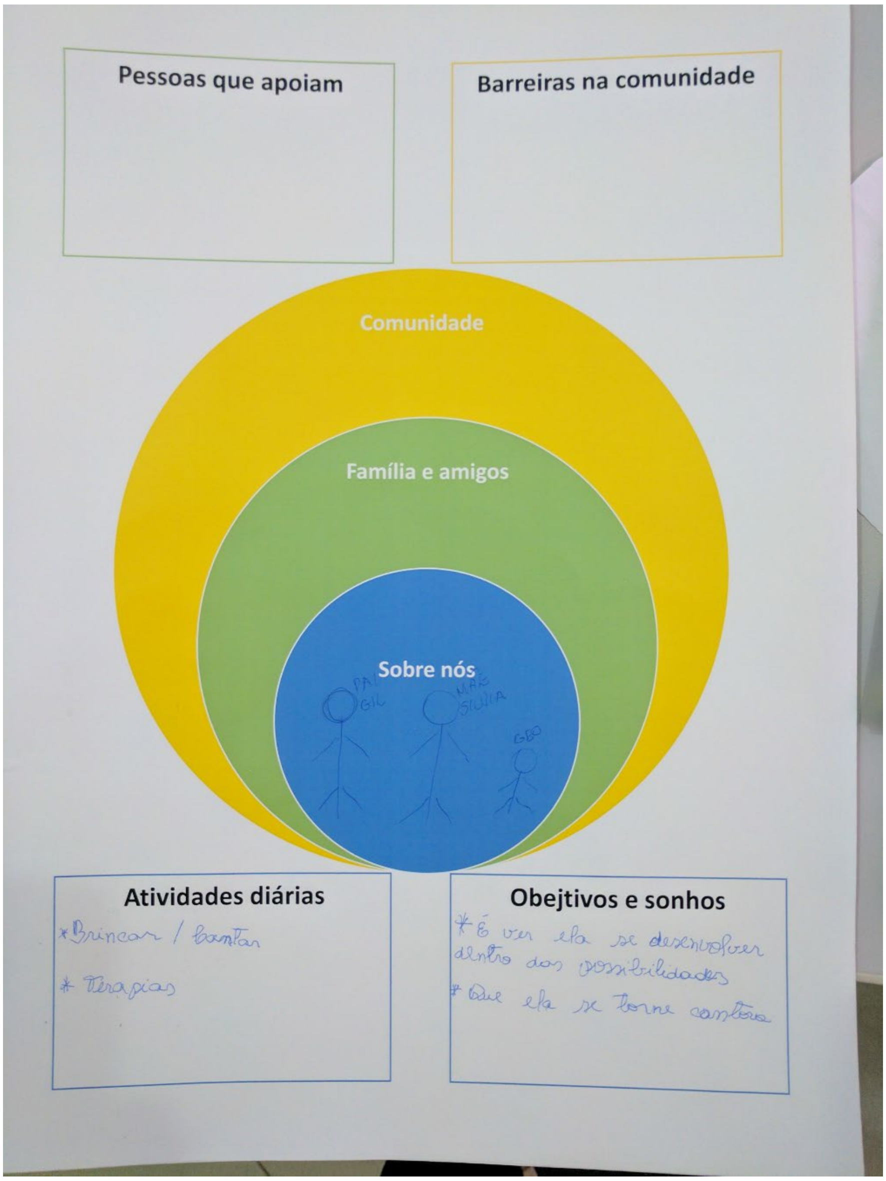
CHAPTER 5: THIRD RESEARCH PAPER (published)

Juntos: A Support Program for Families Impacted by Congenital Zika Syndrome in Brazil



Pessoas que apoiam

Barreiras na comunidade

Atividades diárias

*Brincar / cantar

* Terapias

Obejtivos e sonhos

* É ver ela se desenvolver dentro das possibilidades

* Que ela se torne cantora

Figure 12: The personal poster that families would create during the course of the programme, describing their circumstances, goals, community and activities

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SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic.		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	Global Health: Science and Practice		
When was the work published?	November 4, 2020		
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GLOBAL HEALTH: SCIENCE AND PRACTICE
Dedicated to what works in global health programs
OPEN ACCESS

PROGRAM CASE STUDY

Juntos: A Support Program for Families Impacted by Congenital Zika Syndrome in Brazil

Antony Duttine,[a] Tracey Smythe,[a] Miriam Ribeiro Calheiros de Sa,[b] Silvia Ferrite,[c] Maria Elisabeth Moreira,[b] Hannah Kuper[a]

Key Messages

- A community-based family group program for caregivers of children with congenital Zika syndrome (CZS) was developed based on an existing program for families of children with cerebral palsy and pilot tested in Brazil.
- Program managers developing group initiatives should consider fast-track learning approaches to adjust their intervention to make it more useful to participants.
- Clinicians and managers of Zika initiatives should consider that caregivers of children with CZS will likely benefit from the ability to engage and exchange with caregivers of children with other neurodevelopmental disabilities.
- Program managers delivering community interventions targeting caregivers should consider engaging expert mothers as group cofacilitators.
- Integrating emotional support activities into groups that address child development is important, and adds value.

➔ *Resumo em português no final do artigo.*

ABSTRACT

Background: The 2015–2016 Zika virus outbreak in Brazil was unprecedented and resulted in the birth of more than 3,000 children with congenital Zika syndrome (CZS). These children experience multiple complex health conditions and have limited services to support them and their family's needs.

Program Development and Piloting: An existing family support program for children with cerebral palsy (Getting to Know Cerebral Palsy) was adapted to the Zika context in Brazil through expert consultation. The program was pilot tested at 2 sites among 6 groups of caregivers (total of 48 families) from August 2017 to June 2018. Group observation and focus group discussions with facilitators and participants at the end of each session informed fast-track learning, which was used to tailor the program for future groups. Fast-track learning—adjusting the intervention in real time based on gathered feedback—was found to be a helpful process to inform and hone the program from its initial concept.

Program Description: The intervention, Juntos, is a facilitated participatory group program for caregivers of children who have CZS. The group sessions are cofacilitated by a parent of a child who has CZS and an allied health professional. The group meets for 10 sessions that last 4 hours. Each session includes an icebreaker, activities, and group discussions. Content covers practical information on caring for a child with a developmental disability including that caused by Zika. Psychosocial support forms an important component, and families are guided from the first week to define and develop their own communities of support. Six pilot groups were successfully run in Rio de Janeiro and Greater Salvador, Bahia. The groups gave positive feedback on acceptability and demand.

Conclusions: The program has the potential to be an important tool for community health and social support services in South America in response to Zika. The program can also be applied to children with neurodevelopmental disabilities other than those caused by the Zika virus, which could be important in ensuring families of children with CZS are less isolated.

BACKGROUND

The Zika outbreak of 2015–2016 in South America caught the international health community unaware. There had previously been no severe health consequences associated with the virus, despite Zika having been known since the 1940s.[1,2] Zika has now been

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proven to cause developmental impairments in children[3,4] collectively known as congenital Zika syndrome (CZS).[5] This syndrome includes microcephaly as the most pronounced and documented symptom, which is linked with severe and multiple impairments. Evidence is emerging that Zika also causes an array of other cognitive and physical impairments that may not be immediately apparent at birth. Microcephaly is likely to be the tip of the iceberg in terms of affected children, as more mild or moderate impairments stemming from in utero Zika infection appear to be far more frequent.[6] Brazil was the most affected country in the outbreak. As of March 2020, Brazil had 3,559 confirmed cases of CZS with an additional 2,871 cases under investigation (total 6,430 cases).[7]

Although CZS and cerebral palsy are separate conditions, because they have similarities, programs designed for caregivers of children with cerebral palsy could provide a strong foundation to adapt a program for the Zika context in Brazil.[13] One such program, Getting to Know Cerebral Palsy (GTKCP), was developed by the London School of Hygiene & Tropical Medicine (LSHTM) after a childhood disability survey showed that caregivers of children with cerebral palsy in Bangladesh had very little access to information or support regarding the best way to care for their child and that available services were extremely limited.[14] GTKCP is a 10-session parent-support program held in the community that aims to improve parents' knowledge and skills in caring for their child and improve the quality of life of parents and children with developmental disabilities. It is hard to estimate the exact reach of the program, but an online community of practice established in 2014 to support the rollout of GTKCP has 412 members across 72 countries who share knowledge and experiences.[15,16] GTKCP focuses on parents of children aged 2 years and older; a new version, the Early Intervention Program (EIP), was developed for parents of children aged younger than 2 years.[17] Program material is available from www.ubuntu-hub.org.

Needs Analysis

From April to August 2017, we conducted a needs analysis to assess the potential value of a community-based program, based on GTKCP, for caregivers of children with CZS in Brazil. The needs assessment involved: (1) tracking and comparing emerging literature on the clinical presentation of CZS with existing literature on cerebral palsy; (2) conducting a literature review on the needs of caregivers of children with CZS and cerebral palsy in middle-income contexts; (3) meeting with caregivers, specialists, and other local stakeholders in Brazil to identify key gaps, challenges, and needs; and (4) reviewing emerging data from a sister study measuring the social and economic impact of CZS on caregivers. A full description of the needs analysis is available.[8]

We found that providing some services for children with complex multiple impairments at the community level could be crucial to address the unmet needs experienced by families of children with CZS in Brazil and may be more affordable than centralized services (which may be difficult or costly to access). Families of children with CZS, particularly those children with more severe impairments, did not have enough access to specialized health and rehabilitative services and informal support groups, and formalized support for caregivers was also limited. There was some concern raised by clinicians that children with mild to moderate impairments stemming from Zika infection were less likely to attend rehabilitation and that these caregivers were an important group to be targeted. Other researchers have also reported on the additional services required to fully address the care needs of children with CZS and their families.[9–12]

Families of children with CZS lacked adequate access to specialized health and rehabilitative services, informal support groups, and formalized caregiver support.

Because CZS and cerebral palsy have similarities, programs designed for caregivers of children with cerebral palsy could provide a foundation to adapt a program for the Zika context.

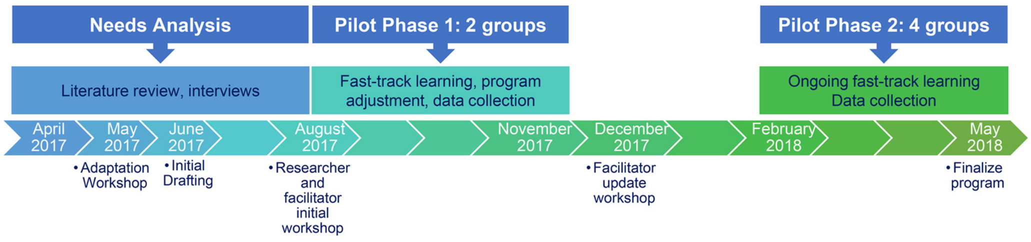
Given the results of the needs analysis that identified the unmet support needs of parents in Brazil and the positive reception of the principle of GTKCP for Brazil among local stakeholders, researchers at the LSHTM who had been involved in GTKCP and EIP felt that adapting GTKCP and EIP for the Zika context and Brazilian culture could be potentially useful. Partnership for the project was established between the LSHTM and 2 Brazilian institutions: the Instituto Nacional de Saúde da Mulher, da Criança e do Adolescente Fernandes Figueira (IFF) in Rio de Janeiro, and the Universidade Federal da Bahia (UFBA) in Salvador.

This article describes the process of developing and piloting the intervention in Brazil, as well as the final program that was developed (Figure 1). We also reflect on lessons learned as key recommendations from this innovative program may be useful for other global health practitioners designing community-based family group interventions.

PROGRAM ADAPTATION AND DEVELOPMENT

After conducting the needs analysis, we developed and adapted the program through expert consultation, and then piloted the intervention using a

FIGURE 1. Timeline of Juntos Program Development for Caregivers of Children with Congenital Zika Syndrome, Brazil



The theory of change linked outcomes with activities to explain how and why the desired change was expected to occur.

multiphase approach. A protocol was established by the lead project researcher (AD) to measure feasibility of the pilot intervention.[13]

Ethics Approval

Ethical approval was obtained from the Instituto de Saúde Coletiva/UFBA Ethics Ref 2.369.348, IFF/FIOCRUZ RJ/MS Ethics Ref2.183.547, and LSHTM Ethics Ref 13608. Informed consent was acquired from all participants.

Initial Adaptation of Program

To support the adaptation, advisory groups were established in Brazil and in the United Kingdom and included a range of specialists, as well as mothers of children with CZS.

The GTKCP and EIP curricula were reviewed by the lead project researcher (AD) with other LSHTM colleagues (TS, HK), Brazilian colleagues (SF, MS), the GTKCP and EIP teams, and other key identified experts (including specialists). The project lead is a physiotherapist with 15 years of programmatic experience, including in qualitative and participatory research and community-based rehabilitation in low- and middle-income countries. During a May 2017 workshop in London, the experts convened to discuss the preliminary findings of the needs assessment and to develop consensus on a first draft outline of the program, an initial timeline, constituency of the facilitators to lead the caregiver group sessions, and participant inclusion criteria.

The project group developed a theory of change to describe how the program relates to broader societal participation of children with developmental delays, including CZS, and the pathways that determine the extent to which this intervention may be successful. The theory of change describes what changes are needed and the assumptions underlying the achievement of these changes.[18] Therefore, the theory of change linked outcomes with activities to explain how and why the desired change was expected to occur and was useful in providing a more comprehensive understanding of steps to improve services to be more inclusive and supportive of family and community. Throughout the program development process, the theory of change was refined to reflect ongoing understanding and research findings (Figure 2).

Several areas of adaptation were identified through the emerging literature, clinical experiences of managing children with CZS, development of the theory of change, and by the GTKCP/EIP teams. These areas included recommendations to further strengthen and develop specific approaches to recognize and address caregivers' psychosocial needs and other clinical issues in children with CZS that were not covered within GTKCP or EIP (e.g., irritability; challenges with breastfeeding or weaning; management of gastrostomy including feeding, low vision, or blindness).

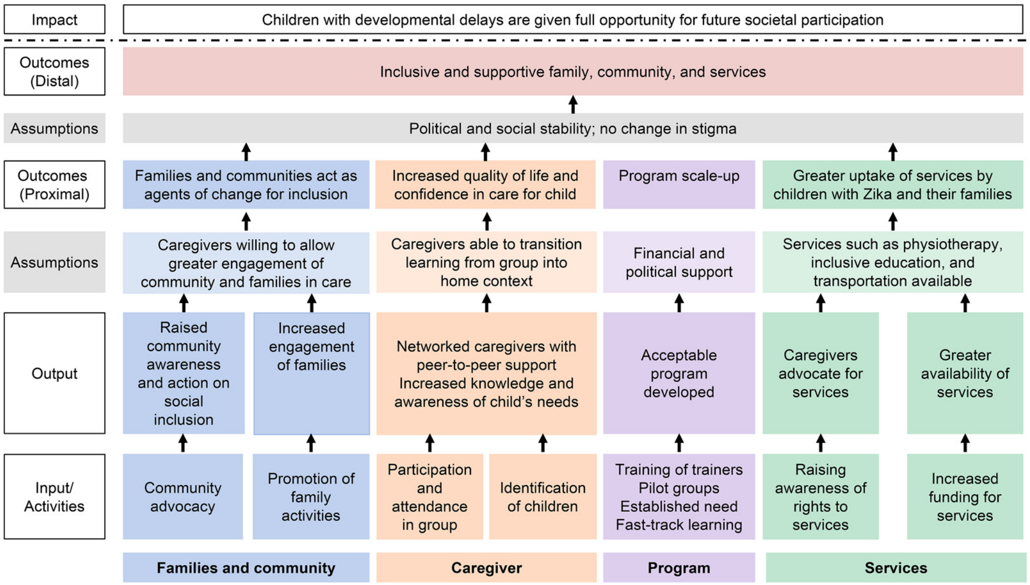
The EIP groups are cofacilitated by an expert mother who has experience caring for a child with cerebral palsy and a rehabilitation professional (e.g., physiotherapist, occupational therapist, or speech and language therapist) who is experienced in working with children who have developmental disabilities. This approach had not been used in GTKCP. A decision was made to pilot test group facilitation by an expert mother combined with a therapist and assess whether this would be effective.

There is a wide range in type and severity of symptoms among children affected by Zika.

It was agreed that program inclusion criteria would be:

- Caregivers of children who have confirmed or suspected CZS but not other types of neurodevelopmental disabilities
- Caregivers of children residing at home and not currently requiring inpatient hospital care

FIGURE 2. Theory of Change for the Juntos Program for Caregivers of Children with Congenital Zika Syndrome, Brazil



- Caregivers willing to attend the whole program and living within 1 hour of the group meeting location
- Children of any age (although given the nature of the epidemic in Brazil in 2017–2018, they were all aged 3 years and younger)
- Children who may be receiving rehabilitation services to address individual needs

More than 1 caregiver of a child (e.g., mother and father, mother and grandmother) was allowed to attend the group meeting.

From June to August 2017, the project lead researcher (AD) drafted the initial program, adapting the GTKCP and EIP materials with input from expert committee members, project teams in Brazil, and other experts (TS, MS, SF, EM, HK).

PILOTING

We piloted the approach during 2 phases with 6 different groups and used this information to finalize the program. A future analysis will report the feasibility of the program using qualitative and quantitative data analysis.

Program Establishment in Brazil

The partners in Brazil (IFF and UFBA) identified a site coordinator (MS and SF) for each of the 2 pilot sites, the states of Rio de Janeiro and Bahia. The site coordinators' main responsibilities were to manage the logistic components of the pilot groups, including identifying an appropriate location for the groups, recruiting facilitators, recruiting researchers, identifying participants, and liaising with local health providers.

Rio de Janeiro and Greater Salvador, Bahia, were selected as pilot sites because they had a large population of children impacted by Zika. Recife, which the LSHTM team visited during the initial country visit, was not selected because several other intervention projects by other organizations were already taking place and contamination of outcomes was a concern. Three sites within Rio de Janeiro and 3 municipalities of Greater Salvador (Simões Filho, Lauro de Freitas, and Camaçari) were selected because of their proximity to families of children with CZS, availability of an appropriately sized venue, and willingness of the local relevant authorities to accommodate a group.

Facilitators were identified by the site coordinators and approved by the project team lead. A total of 8 local facilitators were selected (4 therapists with experience in pediatrics and CZS and 4 expert mothers). In August 2017, a week-long facilitator training was conducted in Rio de Janeiro and led by a trainer who has taught the GTKCP program extensively. The trainer was international, and we used a translator for the sessions as well as materials in Brazilian Portuguese. The training involved education on facilitating a group, practice sessions with reflective learning and feedback, and opportunities for discussion. The project leads and site coordinators selected 2 pairs of facilitators to lead the first pilot groups based on their performance during the training week.

Two researchers were identified by the site coordinators and approved by the project team lead. All the researchers had a background in psychology, but this was not a prerequisite for the role. The researchers participated in a 2-day training in July 2017 on the research approaches and data collection methods and on the fast-track learning approach that would be used to update and adjust the program content based on weekly feedback that they collected from the groups.

Pilot Phase 1

In August 2017, the first 2 pilot support groups—1 in Rio de Janeiro and 1 in Greater Salvador—started meeting weekly. The Rio group had 7 families, and the Greater Salvador group had 8. There were 10 sessions for each group with a different topic each week. Researchers used 3 techniques to collect data to inform real-time feedback and fast-track learning about the content and processes of the session. First, researchers directly observed the sessions and noted the session flow, participants' responses, and behaviors of participants and facilitators. Second, researchers conducted focus group discussions at the end of each of the 10 sessions with participants and (separately) with facilitators to obtain immediate reflections and feedback on the session content. The researchers recorded detailed observation notes about the session and comprehensive notes about focus group discussions that they uploaded to a password-secured Google Drive document for the content developer (TS) to analyze. Third, researchers recorded pertinent comments from participants, facilitators, and site coordinators on images, content, activities, practicalities, and logistics, which were made outside of the sessions. Weekly calls within 48 hours of the session occurred between the researchers and TS, which allowed for further explanation and contextualization. Content issues were recorded and reviewed to update the program in real time and for 4 weeks after the conclusion of phase 1 in November 2017.

Pilot Phase 2

In December 2017, a 3-day training session provided facilitators and site coordinators with information on the changes to the program content and structure based on fast-track learning in the first pilot phase.

Two additional support groups were established in each pilot setting (4 total), with the primary aim of ascertaining the feasibility of the intervention. These support groups had identical procedures for data collection, real-time feedback, and fast-track learning (February–June 2018). After the delivery of the groups, the intervention was further updated, improved, and finalized using the same processes as before. The 2 groups in Rio had 7 and 9 families, respectively, and the 2 groups in Greater Salvador had 10 and 7 families, respectively.

Summary

Six groups ran between August 2017 and June 2018 across 2 phases. The children of the caregivers were 25 males and 23 females with an average age of 23 months (standard deviation=9 months) at their first session. Of the families included in all 6 pilot groups, all (n=48) stated the mother as the primary caregiver. The ages of the mothers (n=48) were 15–20 years (3), 21–25 years (17), 26–30 years (5), 31–40 years (18), and 41–50 years (3). Thirty-six mothers reported they were married, 3 divorced, and 9 reported they were single. Only 6 mothers reported being in work, with the most common reason for not being in work being that they cared for their child (n=34).

During the second and third groups in Greater Salvador, held between January and June 2018, several children with non-Zika related developmental disabilities participated in the sessions. This was done for 2 reasons: (1) to increase the number of children participating because the number of children with CZS who met the inclusion criteria was quite low, and (2) to assess whether combining caregivers of children with CZS and those with other neurodevelopmental disabilities would be a positive experience.

We focused primarily on the caregiver and the program, with some interaction with the family, community, and services at the activity and output levels as informed by our theory of change (Figure 2). The proximal outcomes of the program are expected to be (1) increased participant quality of life and confidence in caring for a child with CZS, and (2) an intervention that is feasible to scale up and replicate in other contexts. Core to the theory of change is empowering the caregiver to improve care for their child through developing support networks and increased knowledge and awareness of their child's needs.

Fast-track learning meant that the intervention was updated and improved as new information was gathered each week about what was working or not. For example, practical or administrative issues, such as organization of transport for participants, were changed and updated in real time each week.

As a result of rapid participant feedback, we made several changes to the program. For example, we changed the title of session 8 (highlighting advocacy and empowerment) to "uniting our voices"; the original title "raising our voices" translated to "shouting out loud." In a second example, participants felt that the images used in the first 2 pilots, which used images from GTKCP and EIP, did not adequately reflect phenotype, family behavior, and environment in Brazil. Therefore, as participants requested, we included images that reflected their lives to create identification and favor more adherence. A local artist was engaged to draw more culturally appropriate images for the later groups, which were perceived more positively. More representation of fathers in caring roles was also incorporated at this stage.

New innovations in Juntos, which were not in GTCKP or EIP, include information on the Zika virus, strengthened participatory approaches to engage participants with community inclusion and disability rights, and a concerted effort to improve male engagement,[19] which was successful to a degree (though the female engagement was still much higher). Additional content includes group discussion on gastrostomy (dysphagia was a common problem), creating trousers stuffed with padding to support children in sitting, using an elasticated cloth to rock children who are irritable, and activities to promote understanding of disability rights. In addition, each session includes reflection and discussion on the session and on the past week through an emotional support activity at the end of the session. The facilitators work as a pair together throughout the session; however, the emotional support activity is facilitated by the expert mother. The first 5 sessions include the same activity with facilitated questions:

- How did you find talking about today's subject?
- Did it raise any emotions or feelings that you did not expect?
- How have you been feeling this week?

The predictability of the questions helps participants to become comfortable with sharing. By week 5, participants have explored much of their thoughts on emotions and feelings, and this then progresses to reflecting on the future.

The feasibility assessment is not detailed in this article and will be described in a future article on the findings.

Through fast-track learning, we updated and improved the intervention as new information was gathered each week about what was working or not.

Finalization of the Program

Consensus on the final content of the program was reached through 2 workshops (London, United Kingdom, and Rio de Janeiro, Brazil) in May 2018. One group in Greater Salvador was still running. However, feedback that had already been collected from the groups was deemed sufficient to be able to finalize the content. The workshops included the technical advisory committees, study site coordinators, and researchers (psychologists).

PROGRAM DESCRIPTION

The final program intervention is called Juntos, which means together in Portuguese and Spanish, to emphasize the importance of inclusion and mutual support. Intervention materials comprise a facilitator manual and participant materials, such as photographs, animations, and video footage. An allied health professional and an expert mother cofacilitate groups that meet once a week for 10 sessions. Support and guidance for facilitators is provided by project coordinators via telephone, email, and/or WhatsApp.

Groups are held at local community facilities, such as health centers, offices of local organizations, or schools, to minimize participants' travel time and to foster relationships between people who lived relatively near to each other. Nine sessions are only for the caregivers and their children, and 1 session is open for other community members to attend. The children who come are looked after in a separate room or space by volunteers, but they are present for some of the practical aspects whenever relevant. Table 1 describes each Juntos module.

The sessions are participatory and use principles of adult learning theory.[20] Participants learn by sharing their own experiences and realities about topics that are important to them, which promotes peer support, critical thinking, and mutual problem solving. The groups start with a light-hearted icebreaker to welcome and warm up the conversation and to encourage comfortable interaction. Participants are then guided through a series of activities, open discussions, pair work, explanations, and demonstrations. Tables 2 and 3 provide examples of session content from session 4 and 6, respectively.

Supportive information was developed for the program that includes short videos on the program

The Juntos logo, which facilitators, psychologists, and site coordinators created to complement the program name's meaning.

and different aspects of care. The individual modules, full manual, and supportive materials are available in English, Portuguese, and Spanish: https://www.ubuntu-hub.org/resources/juntos.

LESSONS LEARNED

The fast-track learning process demonstrated how participant feedback was valued and reinforced the program's focus on caregivers, a group that has been overlooked in the wider Zika response.

Fast-track learning added value to the intervention development because it allowed inclusion of language, logistics, content, and culturally specific changes in real time. Participants' feedback during the first pilot phase was utilized to revise the content (for example, providing case studies, images, and videos of fathers undertaking practical tasks), which may have made the overall content more useful for the later groups. The later groups were aware of this process and recognized some of the changes based on early peers' feedback. In a context of relative distrust and research fatigue,[21] this process helped to demonstrate how participant feedback was valued and reinforced that the program was genuinely and specifically intended for caregivers, an area that had been largely overlooked in the wider Zika response.[22] This could be an important point of learning for global health practitioners implementing community-based group programming: bringing together participants, implementers, and researchers to adapt interventions rapidly as feedback is received. In our approach, although not by initial design, the use of psychologists as researchers and observers provided a unique opportunity for nuanced feedback. This was particularly useful for developing and crafting the messaging and discussions on emotional well-being and psychosocial support.

The integration of a component of caregiver emotional well-being in this group intervention demonstrates a novel approach to including psychosocial support to better promote emotional well-being as an integral part of health work, rather than being seen as a standalone effort. There is no single recognized theory of how participatory groups achieve their health impacts[23] and few studies evaluate how and why different support networks improve caregiver and child outcomes. Examples in resource-limited settings include self-help groups for people with mental health conditions, which demonstrate positive impacts on both the people with mental health conditions and their caregivers.[24] Additionally, women's self-help groups have resulted in improved maternal and neonatal survival.[25] Our integration of a mental health component in Juntos illustrates that groups that address child development can practically integrate emotional support activities. Facilitators reported that they valued having a dedicated space each week to raise issues of emotional well-being. The practical components of the sessions often raised some emotions for a participant, but there would be little time to explore these, so the final section allowed further exploration and discussion between the group. Evaluation of whether such a strategy can work in other settings is necessary, and negative and unanticipated consequences warrant further evaluation in future work. Having an expert mother facilitate these sessions was particularly important and helped form group connections that might not have been possible with an allied health professional alone.

In understanding pathways to change, the role of the expert mother appears to offer crucial

TABLE 1. Finalized Module Topics Included in Juntos, A Community-Support Group for Caregivers of Children with Congenital Zika Syndrome in Brazil

Module	Topics
1. Introduction	• About the program • Information about Zika and Congenital Zika Syndrome • How to find information • Personal stories
2. Our child	• Introducing your close family and friends • Development milestones for young children • Determining your child's progress • Managing irritability and crying
3. Positioning and moving	• How to position children who need assistance • How to assist children to learn to move
4. Eating and drinking	• Feeding challenges • Practical skills to address challenges for your child
5. Communication	• Importance of communication • Practical advice to help your child communicate
6. Play and early stimulation	• Importance of play for children to develop and learn • Early stimulation • Making simple toys • Inclusion of play in the family and broader community
7. Everyday activities	• How to use everyday activities to help your child develop • Managing seizures
8. Uniting our voices	• Understand the context of disability rights • Education • Communicating with your health team • Advocating
9. Our community	• Who is in your community? • Common barriers to inclusion • Addressing negative attitudes and exclusion • Social activity
10. Next steps	• Summing up • Planning next steps for yourself and the group

encouragement to shared learning between caregivers and contributes to developing an egalitarian atmosphere, expanding care practices beyond traditional rehabilitation models.[26] Relating this common ground and a sense of belonging through a social support network provides an environment to improve the knowledge and skills of caregivers.[15] It was critically important that the 2 cofacilitators were equals, each bringing their own experiences to the process and an expertise and insight that the other did not possess. The allied health professionals immediately saw the value in this, and there was no sense of protectionism or defensiveness that they needed to be the lead or expert given their professional training.

Groups were held in the local community so that caregivers could build strong local networks. This also increased interest from caregivers of

The expert mother appears to offer crucial encouragement to shared learning between caregivers and contributes to developing an egalitarian atmosphere.

TABLE 2. Example of Content From Facilitated Group Session 4 on Eating and Drinking from Juntos, A Community-Support Group for Caregivers of Children With Congenital Zika Syndrome in Brazil

Example	Discussion	Aim
Icebreaker In pairs: One person tries to give the other a drink of water in different positions (e.g., head leaning back, turned to one side, or flopping forwards)	How easy or difficult is it to swallow in each position? How does it feel to be fed?	To understand a range of issues that your child may experience with eating and drinking
Discussion As a large group to share experiences	What is a nutritious or "balanced" diet?	To know what a balanced diet is and how to maximize your child's nutritional intake and prevent malnutrition
Activity Show a banana and a biscuit and other common food	Discuss—Are the items hard or soft? Can they be made into a smooth puree? How?	To learn ways to feed your child safely

TABLE 3. Example of Content From Facilitated Group Session 6 on Play and Early Stimulation from Juntos, A Community-Support Group for Caregivers of Children With Congenital Zika Syndrome in Brazil

Example	Discussion	Aim
Icebreaker In groups of 3: each group is given one inexpensive everyday item (e.g., cup, piece of cloth, container, ball) and everyone uses their imagination to transform the object into something else and acts it out	What is play?	To understand how our imagination works with play and how children have an even greater imagination than adults
Discussion As a large group to share experiences	What have you found play helps your child to do? Does your child need to play?	To know that play gives children an opportunity to explore, learn about their environment and to use and develop their senses
Activity Toy making, such as making bells and rings with ribbons	Discuss—How can you involve short periods of play in your daily activities? How can you involve other members of your family in playing with your child?	To learn ways for play to be fun, and to see how fun can motivate children to move and learn and how other family members can be included

children with developmental disabilities other than CZS and highlights the importance of de-isolating Zika from other causes of neurodevelopmental disability when developing community support programs. Juntos does not replace health care services but rather seeks to complement services by empowering other caregivers to optimize their child's care and upbringing.

We received positive feedback during the sessions that combined caregivers of children with CZS with caregivers of children with other neurodevelopmental disabilities. There was a recurrent expression of comfort among the caregivers when engaging with other caregivers in similar situations and circumstances that they were not as alone, unique, and isolated as they had perhaps feared. This was also seen in the sessions where non-CZS caregivers engaged and, in fact, there was a value perceived to understand that the challenges being faced were not unique to only caregivers of CZS. This was also reinforced frequently in session 8 of the Rio sessions, where an external speaker came from a local Down's Syndrome organization to discuss their advocacy approaches; the sessions were always extremely well received by participants. Although the challenges facing children with CZS and their caregivers remain unique and, to a certain degree, still unknown, there may be an important value to ensure that there are also many common issues faced and a shared approach may be both efficient and useful.

RECOMMENDATIONS

The needs analysis that we undertook at the beginning of the project[8] as well as more recent literature[27,28] has highlighted overlaps and similarities between cerebral palsy and CZS. We suggest that children with CZS and their caregivers may benefit by integrating and linking with services and programs for children with other neurodevelopmental disabilities. Rehabilitation/therapy services were already doing this to a large extent, and there seems a good scope for other health and social service providers to also ensure service integration. Conversely, newly formed services as a result of the attention to CZS shouldn't be exclusive to this population group and should seek to include all families and children who may benefit.

By the nature of its design, Juntos can potentially be implemented by a range of stakeholders, from nongovernmental organizations to public community services to primary health settings. This flexibility may mean that there is a stronger opportunity for Juntos to be scaled up. The universal primary health structure in Brazil—the Sistema Único de Saúde—could be an avenue to further explore. We see opportunities for public/private partnerships also. Cost is clearly a major factor in the potential for scale up. Facilitator training can be done in larger groups to reduce costs. In addition, if the facilitator therapists undertake the role as part of their existing work, these costs may be further reduced. However, we do feel that it is important to remunerate parent facilitators for their work and other costs, such as transport and refreshments, to ensure full participation of families.

Strengths

Strengths of this pilot include the development process being informed by a theory of change and reflective practice and robust methodology that allowed integration of rapid feedback. Real-time feedback and adaption enabled the development of a culture-specific and language-specific intervention, and the program was developed and refined to meet the needs of caregivers of children with CZS in Brazil. Running the program in 2 sites concurrently (Rio de Janeiro and Greater Salvador) was an important methodological choice for achieving better final version program. Brazil is huge and diverse, and although these 2 sites do not cover the breadth of diversity, piloting in more than 1 site and acquiring different feedback added to the strength of the study.

Limitations

Our study has limitations. We describe the intervention development, but assessment of feasibility and evaluation of replication and scale-up in other countries is now needed. More work is needed on forming a comprehensive facilitator training program, and further development of the intervention to include all children with developmental disabilities is warranted. If Juntos is found to be feasible, robust studies to evaluate the cost-effectiveness of the intervention will be needed.

CONCLUSIONS

We developed and refined a participatory community-based group intervention to meet the needs of caregivers of children with CZS. Juntos has the potential to be an important resource for community practice. There is scope to expand across Brazil and in other South American countries and to children with other developmental disabilities.

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En português

Juntos: Um Programa de Apoio às Famílias Afetadas pela Síndrome Congênita do Vírus Zika no Brasil

ABSTRATO

Histórico: O surto, sem precedentes, do vírus do Zika em 2015–2016 resultou no nascimento de mais de 3.000 crianças com a Síndrome Congênita do Vírus Zika (SCZ). Essas crianças experenciam múltiplas e complexas condições de saúde com limitado acesso a serviços de apoio tanto para elas quanto para as suas famílias.

O desenvolvimento de um programa piloto: um programa existente de apoio às crianças com paralisia cerebral (*Getting to Know Cerebral Palsy* -Conhecendo a Paralisia Cerebral) foi adaptado para o contexto do Zika no Brasil através de uma consultoria especializada. O programa piloto foi testado em dois locais com 6 grupos de cuidadores (um total de 48 famílias) entre agosto de 2017 e junho de 2018. Grupos focais e de observação com facilitadores e participantes receberam avaliações ao final de cada intervenção que foram utilizadas para adequar o programa para grupos futuros, através da metodologia de aprendizagem rápida. Isso permitiu ajustar as intervenções em tempo real, o que provou ser um processo útil para informar e aprimorar o programa desde a sua concepção inicial.

Descrição do programa: a iniciativa Junto é um programa de facilitação e participação para grupos de cuidadores de crianças com SCZ. São dez encontros com a duração de 4 horas - cada um inclui uma dinâmica inicial de quebra gelo, atividades e discussões em grupo. O conteúdo cobre informações práticas sobre os cuidados com crianças com problemas de desenvolvimento, incluído aqueles causados pelo Zika. O apoio psicossocial abrange um componente importante no qual as famílias são orientadas desde a primeira semana sobre como definir e desenvolver suporte em suas comunidades. A realização com seis grupos no Rio de Janeiro e na aérea metropolitana de Salvador ocorreu de forma exitosa e em ambos os locais houve um retorno positivo em termos de aceitação e demanda.

Conclusões: o programa tem o potencial de ser uma ferramenta importante para as aéreas de saúde e prestação de serviços sociais na América do Sul em resposta ao vírus do Zika. Além disso, pode ser adaptado para crianças com problemas no neurodesenvolvimento para além daqueles causados pelo Zika, o que por sua vez, pode ser importante para garantir que as famílias de crianças com SCZ sintam-se menos isoladas.

Aspectos Principais

- Um programa de orientação comunitária para grupos de família desenvolvido para os cuidadores de crianças com a Síndrome Congênita do vírus Zika (SCZ), baseado em programa anterior focado em crianças com paralisia cerebral, foi testado como uma experiência piloto no Brasil
- Gestores que desenvolvem atividades de grupo devem considerar as abordagens de aprendizagem rápida para adequar as suas intervenções, tornando-as mais úteis para os participantes. Médicos e gestores de iniciativas para o apoio às vítimas do Zika devem atentar para o fato de que cuidadores de crianças com CZS podem se beneficiar da interlocução e troca com cuidadores de crianças com outras problemas de neurodesenvolvimento.
- Gestores de programas focados em intervenções comunitárias devem levar em consideração engajar as mães como um grupo de cofacilitadoras.
- Integrar atividades de apoio emocional em grupos para abordar a questão do desenvolvimento infantil é algo prático importante e agrega valor.

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CHAPTER 6: FOURTH RESEARCH PAPER (published)

Assessment of the feasibility of Juntos: A support programme for families of children affected by Congenital Zika Syndrome

Figure 13: Mid-session discussions led by a facilitator

RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic.		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	Wellcome Open		
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For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I led the design, structure, authoring and corrections of the manuscript including collating feedback from co-authors.

SECTION E

Student Signature	
Date	19th September 2022

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RESEARCH ARTICLE

Assessment of the feasibility of Juntos: A support programme for families of children affected by Congenital Zika Syndrome

[version 1; peer review: 2 approved]

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2. **Sarah Mulkey**, Children's National Hospital, Washington, USA
George Washington University School of Medicine, Washington, USA

Any reports and responses or comments on the article can be found at the end of the article.

Abstract

Background: The 2015-16 Zika epidemic resulted in thousands of children born with congenital Zika syndrome (CZS). In Brazil, gaps in the health system often caused parents to be left with insufficient information and support. Consequently, we developed and piloted Juntos - a participatory support programme which aims to improve knowledge, capacities and build support networks for caregivers of children with CZS.

Methods: Six caregiver groups received the programme between August 2017 and June 2018: three in Rio de Janeiro and three in Bahia. We assessed the feasibility of Juntos against six of the eight areas of a feasibility framework described by Bowen *et al.* to consider whether Juntos 'could work'. These areas were: acceptability, demand, implementation, practicality, adaptation and limited efficacy. We used mixed methods including: 1) baseline and end-line questionnaires completed by all group participants; 2) in-depth interviews with 18 participants, seven facilitators and three key stakeholders; 3) participant focus group discussions after each session; 4) researchers session observation; and 5) recording programme costs.

Results: 37/48 (77%) enrolled families completed both questionnaires. Acceptability and demand were noted as high, based on participant responses to interview questions, focus group feedback and satisfaction scores. Potential for implementation and practicality were also demonstrated through interviews with facilitators and key stakeholders and analysis of project documents. Two groups included caregivers of children with non-Zika related developmental disabilities, showing potential for adaptability. Self-reported quality of life scores increased in caregivers between baseline and end-line, as did the dimensions of family relationships and daily activities in the Pediatric Quality of Life Inventory (PEDS QL) Family Impact Module,

showing limited efficacy.
Conclusions: The programme showed feasibility according to Bowen's framework. However, further research of scale up, particularly in the areas of integration, expansion and limited efficacy are needed to ascertain if the programme is effective.

Keywords
Zika, disability, microcephaly, early intervention, Congenital Zika Syndrome, family, caregiver, Brazil

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Author roles: Duttine A: Conceptualization, Data Curation, Formal Analysis, Investigation, Project Administration, Software, Supervision, Validation, Writing – Original Draft Preparation; **Smythe T**: Methodology, Project Administration, Resources, Supervision, Writing – Review & Editing; **Calheiros de Sá MR**: Supervision, Validation, Writing – Review & Editing; **Ferrite S**: Supervision, Validation, Writing – Review & Editing; **Moreira ME**: Funding Acquisition, Project Administration, Supervision, Writing – Review & Editing; **Kuper H**: Conceptualization, Funding Acquisition, Methodology, Supervision, Writing – Review & Editing

Competing interests: One of the researchers (AD) joined the Pan American Health Organisation (PAHO) during the research period. Work on the research study was undertaken outside and separate to his PAHO duties.

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Introduction

Brazil was the most heavily impacted country in the 2015–16 Zika outbreak, which triggered the World Health Organization to announce a Public Health Emergency of International Concern (PHEIC)[1,2]. The causal link between Zika and birth impairments and developmental delays has now been demonstrated[3]. Congenital Zika syndrome (CZS) is the collective term used to describe the pattern of structural anomalies and functional impairments seen after Zika infection[4]. This condition includes features such as microcephaly, which was the most associated sign of neurodevelopmental disability during the Zika outbreak. However, not all children display microcephaly at birth and many children may be presenting with Zika-related neurodevelopmental delays who were not detected at birth[5,6]. Common conditions and impairments include physical and motor difficulties, intellectual impairments, vision loss and epilepsy. Between November 2015 and March 2020, 18,828 cases of suspected CZS or other aetiologies have been reported to the Ministry of health in Brazil, with 3,523 confirmed cases of CZS[7]. The states of Pernambuco and Bahia in the North East of the country and Rio de Janeiro in the South East have seen the most cases[7].

Children born with CZS are likely to have long-term impairments and disabilities leading to social and economic impacts on families and caregivers, such as depression, anxiety and stress[8]. Similar trends have been observed in families raising children with other neurodevelopmental disabilities, such as cerebral palsy (CP)[9–13]. Children with long-term neurodevelopmental disabilities are likely to need medical, rehabilitative and social support in order to optimise functioning and participation[14–16]. In addition, parents and families themselves may have support needs, including education and psychological support[17,18].

The health response in Brazil was mainly focussed on addressing the clinical needs of the children, including provision of medical and therapy services. However, a needs analysis conducted in April-August 2017 showed that there were gaps in service provision and that the needs of children with CZS and their caregivers were not being met though the Brazilian health system alone[19]. Several formal support groups had been established, but they did not follow a curriculum of training and support. Informal parent support networks existed, such as WhatsApp groups, which were important for parents. However, they did not provide structured guidance and support. The needs assessment highlighted that provision of a structured support programme was an emerging priority within the context of the Zika outbreak[19].

In response to these unmet needs, we adapted an existing programme that had been developed for children with cerebral palsy in low- and middle-income contexts. *Getting to Know Cerebral Palsy* (GTKCP) was developed in South Asia for settings where parents had little or no access to formal health or social services[20,21]. The programme involves a series of structured sessions around different aspects of caring for children with CP. It has been implemented in many countries and settings across the world. A later adaptation – the Early Intervention Programme (EIP) - has been developed for families of children under two years[22]. We hypothesised that adapting GTKCP to the Zika context in Brazil would be a feasible and efficient approach, given the likely similarities in experiences of caregivers of children with CP to those with CZS[19].

The intervention

Juntos (meaning 'together') was developed from GTKCP and EIP using an evidence-based approach[23]. The programme aims to improve knowledge, capacities and build networks of support for caregivers of children with CZS. Each week focusses on a different topic and covers the basics of child development and developmental delays, practical sessions such as facilitating play, feeding and communication, and social sessions such as knowing rights and living in the community. The groups are facilitated by a partnership between a therapist (physiotherapist, occupational therapist or speech and language therapist) and a mother of a child with CZS ("expert mother"). The combination of a therapist/mother was chosen to bring a balance of different expertise to the sessions. Facilitators were trained over a one-week period and received support from supervisors during the programme. A coordinator, at each location, oversees the planning and implementation of the programme including recruitment of facilitators and participants, identification of training locations and coordinating logistics for hosting the sessions. Children attend the sessions but were cared for in another space to allow fuller engagement of the caregivers. A full description of the development of the intervention and the Juntos programme is available in a separate paper[23].

The aim of this paper is to describe the feasibility of implementing the pilot Juntos programme in two settings in Brazil.

Methods

The feasibility study was undertaken in partnership with the Instituto Fernandes Figueira in Rio and the Federal University of Bahia in Salvador, with each site nominating a site coordinator. Ethics approval was obtained in both Brazil (IFF/FIOCRUZ - RJ/MS 2.183.547) and the UK (LSHTM Ethics number 13608). A protocol for the project, including the feasibility assessment, was established[24].

Intervention implementation

Juntos was piloted across six groups in two geographical locations (Rio de Janeiro and Greater Salvador) between August 2017 and June 2018. Groups of six to 10 caregivers of children with CZS were formed, who met weekly in the local community over a period of 10 weeks[23].

Feasibility assessment. The holistic evaluation of the programme's feasibility was structured based on a model proposed by Bowen *et al.* (2010)[25] for assessing the feasibility of public health interventions. Bowen *et al.* proposed eight areas of focus to measure, giving an overall picture of the feasibility of an intervention (Table 1).

Table 1. Eight areas of focus from Bowen *et al.*[25].

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?"*

Bowen *et al.* also proposed that these eight areas can be measured at three different stages of an intervention[25] to answer the following questions:

1. "Can it work?" Is asked at the stage when the intervention is being developed and piloted.
2. "Does it work?" Is asked when positive preliminary results have been shown and the intervention has been formally tested.
3. "Will it work?" Is asked at the stage when an intervention has shown to work and aims to be adapted or scaled up.

While this intervention is derived from an existing intervention, Juntos contained new elements and was being delivered to a pilot group in Brazil for the first time[26]. Therefore, this research project aimed to primarily answer the question of 'Can it work?', but with some attention given to explore 'Does it work?' and 'Will it work?'. For our feasibility assessment, we focused on six of the eight areas of Bowen *et al.*'s framework: acceptability, demand, implementation, practicality, adaptation and limited efficacy. The other two areas (integration and expansion) were difficult to measure reliably given the small scale of the pilot project.

The data for the feasibility assessment was collected between August 2017 and June 2018 within the context of six pilot groups implemented in two locations in Brazil – Rio de Janeiro and Greater Salvador, which were both heavily impacted by the Zika outbreak[7,27]. Four researchers were recruited by the site coordinators in collaboration with the London School of Hygiene and Tropical Medicine (LSHTM) team. The researchers' role was twofold: to observe sessions and conduct rapid feedback with participants and facilitators in order to inform fast-track learning for adjusting and honing the programme; and to facilitate baseline and end-line questionnaires, and participant and facilitator semi-structured interviews. The researchers were all female and all had a background in psychology, though this was not a pre-requirement for the position. A two-day training workshop was held for the researchers before the first group, followed by a one day updating session between the first and second groups of each location.

All participants who took part in the programme completed a consent form, relevant to their involvement in the study (e.g., questionnaires, interview). Participants were also requested to provide consent for photographs or other media to be recorded during the group sessions, after explaining that non-agreement to the media consent form would not impact their position in the groups.

We collected a range of data and utilized various methods to assess the six selected areas of Bowen *et al.* This allowed us to analyse the data and acquire a richer understanding of the different facets of each area:

Participant data

Baseline and end-line programme semi-structured quantitative questionnaires were completed by all programme participants before the first session and after the last session. If caregivers from the same family came together (e.g., mother and father, mother and grandmother), they would complete one questionnaire per pair with the primary caregiver (usually the mother) as the lead responder. Questionnaires included the following items (*Extended data*[28]):

- Socio-demographic characteristics of the child and caregivers (usually mother and father) (baseline only).

- Understanding and knowledge about the child's condition by the caregiver.
- Knowledge and confidence to care for child, assessed as a five-point Likert scale.
- Health status of child (including questions on general health, serious health issues, seizures and sleep)
- Eating and drinking status of child (including questions around difficulties feeding, level of support, weight gain etc)
- The PedsQL Family Impact Questionnaire Module (Brazilian Portuguese version)[29,30].
- A Cantril scale for assessing quality of life of caregiver and of child[31], implemented as a 10 point ladder.
- Review of goals achieved (end-line only).
- Satisfaction (scored out of five for satisfaction of content, organisation and facilitators) and qualitative reflections on the programme (end-line only).

Questionnaires were developed in English, then translated into Portuguese.

The baseline and end-line programme questionnaires were particularly useful in assessing acceptability, demand and limited efficacy areas of feasibility.

In-depth, semi-structured interviews were undertaken face to face by the researchers with 18 participants (16 female, two male) after each group's final session. Participants were selected purposively at the researchers' discretion to reflect a broad a range of perspectives (e.g., caregivers of children with different disability severities, mothers and fathers). Since the researchers had been observing all the sessions, they were all known to the participants before the interview. Interviews were undertaken in Portuguese by the local researchers and focussed on satisfaction with and perceived impact of the groups using an interview guide (*Extended data*[28]) developed by the lead author. The guide was not piloted before the first interviews. The interviewers recorded and transcribed the interviews in Portuguese and they generally lasted 30–45 minutes. Participants were usually with their child.

Participant data was used to evaluate the acceptability, demand, implementation and limited efficacy areas of feasibility.

Facilitator data

In-depth interviews were undertaken with each of the facilitators (total = 7) at the final session of the final group. These were undertaken face to face in Portuguese by the local researchers, who recorded and transcribed the interviews. The interviews focused on the facilitators' reflections and lessons learned, including perception of participant engagement and the impact of using an interview guide (*Extended data*[28]).

Facilitator interviews were used to evaluate the implementation, practicality and adaptation areas of feasibility.

Key stakeholder data

In-depth interviews were conducted with the two site coordinators and a senior medical provider. The interviews focussed on the practical components of implementing the sessions, reflections on lessons learned and potential future expansion using an interview guide (*Extended data*[28]). Interviews were undertaken in English by the study lead (AD) and were transcribed but none returned to the participants.

Key stakeholder data was used to evaluate the practicality and adaptation areas of the feasibility.

Other data

Session costs were assessed by analysing the budget, establishing an overall cost for delivery of the programme, and the cost per participant. Facilitator training costs were calculated and presented separately, as they may not reflect the true costs if the programme was to be scaled up (e.g., the number of facilitators, international travel, etc.). These are variable costs that would need to be estimated at the start of each new programme.

The researchers observed the sessions and used an observation framework to record notes on the: logistics (e.g., timeliness), environment created by facilitators (e.g., room set up), response of caregivers and parents (e.g., contributions of participants) and response of children (e.g., volunteer caregivers' presence)

Costs and researcher observations were used to evaluate the implementation and practicality areas of feasibility.

Data analysis

Analysis of the interviews and session notes/focus groups was undertaken by a social scientist fluent in English and Portuguese who coded the interview responses in NVIVO 12 (Taguette is an alternative open-sourced software). Thematic analysis was structured in advance and centred around the six areas of feasibility being used for this assessment[25]. The lead author then reviewed all interviews in English and the Portuguese translated elements.

Quantitative data on participant demographics, PEDSQL Family Impact Questionnaire, Cantril Scale and satisfaction were tabulated into Microsoft Excel. Two tail T-testing was performed for the PEDSQL and Cantril Scale. Only questionnaires which had both base- and end-line questionnaires completed (n=37) were used for analysis of PEDSQL and Cantril Scale. All baseline questionnaires (n=48) were used for analysis of the participant demographics.

Results

A total of 48 families enrolled in the Juntos programme across the six groups (Salvador n=25, Rio n=23) and completed a baseline assessment. 37 families (77%) completed the programme and undertook the end-line assessment, with a slightly higher completion rate in Salvador (84%) than Rio (70%). The number of family units enrolled in each group ranged from seven to 10 (average = eight). The average age of the children of the caregivers at the time of the first session was 23 months (range 13–58 months[1]). Table 2, below, summarizes the six groups.

Caregiver demographics

The mother was the stated primary caregiver in 46/48 (96%) of the baseline assessments. Most mothers (75%) self-reported that they were married, with 6% divorced and 19% single. A 69% portion of the fathers were living with the mother and 83% of the fathers saw their child on a daily basis. Only three fathers (6%) reported that they had not seen their child in the past six months. The ages of the mothers and fathers are shown in Table 3 below:

Only 13% of mothers reported being employed compared to 70% of fathers. 92% of the mothers said they were not working because they needed to care for their child.

The participant, facilitator, key stakeholder and other data that was collected are grouped below following the six selected areas of Bowen *et al.*'s framework: acceptability, demand, implementation, practicality, adaptation and limited efficacy.

Acceptability

Satisfaction with the programme, as assessed in the end-line questionnaires, was scored highly by the participants (n=35), with an average score of 4.6 (out of 5) for content, 4.8 for organization and 4.7 for facilitators. Group attendance had an average of 6.3 participants per session (based on data from four out of the six groups) ranging from 1 to 17.

Table 2. groups, dates and participant numbers.

Group	Dates	Number of families who enrolled	Number of families who completed the end-line questionnaire
Salvador 1	11th August – 17th November 2017	8	6 (75%)
Salvador 2	20th January – 19th May 2018	10	8 (80%)
Salvador 3	13th March - 13th June 2018	7	7 (100%)
Rio 1	17th August – 21st November 2017	7	7 (100%)
Rio 2	11th January – 26th April 2018	7	3 (43%)
Rio 3	26th February – 6th June 2018	9	6 (67%)
		48	**37 (77%)**

Table 3. Ages of mothers and fathers.

Age (years)	Mother (n=48)	Father (n=37)[2]
15–20	5 (10%)	3 (8%)
21–25	17 (35%)	8 (22%)
26–30	5 (10%)	8 (22%)
30–40	18 (38%)	12 (32%)
40–50	3 (6%)	6 (16%)

During the interviews, a number of caregivers spoke about how being able to engage and share with other caregivers in similar situations was an attraction:

> *"I could notice that there were mums in the same situation as me, mums with bigger weaknesses than me, others stronger than me, so I saw it all, and this programme was very important."* (Participant, Rio)
>
> *"I think that the main thing - the most important thing - is the sharing of experiences, because although there is microcephaly, every case is different, right? So, my daughter has her characteristics, the daughter of another has other characteristics, but I think that sharing experiences is a big plus, right? "Look, I did this, and it worked out, I do it this way". The sharing of experiences is very valid.* (Participant, Rio)

There also appeared to be recognition that the programme focused on areas that the caregivers felt they needed guidance and support with and provided psychological comfort:

> *"The programme is interesting…the purpose of this programme is to make the mother be able to administer the issue of microcephaly in a good way...it's not easy, not only for the parents, but for the family, and daily life is complicated...the logistics of everything. There's also the emotional side. So, I think the idea of this programme contributing to helping administer things better is great."* (Participant, Rio)
>
> *"I think it supported us a lot, totally supported us. It's like the group was a huge hug. It was a hug, it was what we needed, the support of someone, greater than even ourselves. So I guess support was everything.*

---

[1] The 58-month-old was a non-Zika child.

[2] Although we collected information on 37 fathers, not all of them participated in the sessions. The mother/primary caregiver was asked to provide information about the father even if they did not attend.

> *It changed everything for us. It pulled us up again because everyone was already down. It helped us a lot." (*Participant, Salvador)

Having a mother expert as a facilitator was also deemed as important/advantageous by other caregivers, as well as by the therapist facilitators:

> *"I think the dynamics are great! Very good! I think it's essential that you have a mother, or an aunt, or a grandmother, that a caregiver is one of the facilitators."* (Therapist facilitator)

However, it was more challenging to get fathers to commit and stay for all the sessions of the programme. When fathers did come, they would not always come to every session or be able to stay for the whole session. Part of this low attendance was related to sessions being offered during the week, when many fathers were working. Furthermore, one father who did attend, and was interviewed, suggested that the format may not be the most natural environment for fathers, who he felt tend to be more timid in interacting in group settings.

Demand

Out of the forty-eight families who were enrolled in the groups, thirty-seven (77%) attended the final session and provided end-line questionnaires (Salvador n=21, Rio n=16), Table 2. In Salvador, fifteen (60%) families attended at least seven sessions, but only four (16%) came to all 10 sessions. Reasons for dropping out or non-attendance included sickness or death of the child, difficulties with work schedules and transportation, and having too many other appointments and commitments. Recruiting families was a challenge for two of the groups and one, in Greater Salvador, needed to restart due to low numbers.

Despite these challenges, the interviews revealed that the Juntos programme potentially filled an important gap that was not currently being provided though the formal health system or other services. This was reflected by participants, facilitators and key stakeholders:

> *"And I still hadn't had personal contact with mothers in the same situation as me, I had seen them on TV or here in the corridors, but I didn't have the opportunity to sit and talk, "How is your life, is it similar to mine?"* (Participant, Rio)
>
> *"There were [other] mothers who wanted to participate, but they couldn't and wanted to join on the fourth or fifth module, and we said they couldn't, but that when a new one opened we would let them know."* (Parent facilitator, Rio)

Implementation

The six-pilot group programmes were successfully completed in two major cities of Brazil, suggesting that the programme can be effectively delivered within urban areas. The direct observation revealed that sessions flowed well and that there were good levels of interaction. Facilitators reported that the structure of the sessions and the facilitator guide made the programme logical and easy to lead:

> *I think that 'Our Child'* [session 2] *went well and easy to lead, the 'Play and Early Stimulation' as well [...] and diet, was easy, and the module I think that worked the best, and answered most doubts for everyone was 'Daily Activities'. Nobody included their children in activities, apparently, and there, well, it opened a horizon of options for them"* (Facilitator)

A second room was used as a creche/play area for the children of the caregivers, which allowed the caregivers to be fully engaged in the process. A team of volunteers looked after the children during the sessions. This was positively reflected in the facilitators' interviews:

> *The fact that there were two rooms, I think that it was a positive point, because it's a moment that … not that they stop being mothers, but they leave aside a bit the "I have to live for my child, everything is for my child" and look at themselves as people.* (Facilitator)

Practicality

The site coordinators spent time in identifying a suitable community location that was accessible for participants. These varied from primary health facilities, from the office of an NGO to a school. This consideration was appreciated and deemed as important:

> *"I liked it, I think it was an easy place to get to, near metros, trains, you know?"* (Participant, Rio)

During their interviews, the coordinators also highlighted key features that they identified for a space to make the group sessions work: sufficient space to arrange participants in a circle, space to have the children close to parents (ideally in another room, close by), in a safe location, on the ground floor (or elevator available), accessible by public transportation, and no more than a 30-minute travel away.

Observation notes do suggest that at times the logistics could be challenging, with sessions often starting behind schedule and some not being able to finish all of the content on the same day. The time management aspect did seem to improve in later groups as facilitators became more confident and familiar with the content.

We calculated the average cost to run a 10-session group to be 37,300 Brazilian Reais (7,460 GBP[3]). This includes the salaries of two facilitators for four months, a coordinator for five months and all the group activities (materials, refreshments and transportation). With an average of eight families per session, this amounts to 4,662 Brazilian Reais (932.50 GBP) per family for the entire 10 sessions. It is important to note that

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[3] GBP/BRL exchange rate of 4.96 taken from xe.com for the 13th June 2018 (last day of the last Juntos session)

the facilities we used for the groups were all made available at no cost, but this may be an added expense to consider if the programme scales up. This cost does not include the cost to run a facilitator training session and assumes facilitators are trained.

Adaptation

Adaptations were made to the structure of the programme from the first two groups to the last four, as fast track learning and adjustments were made. In terms of location, different settings were used. In Salvador, all the settings were local primary health facilities, but in different parts of the city. In Rio, two sessions occurred at the same location, a central office space used by a local NGO. The other session took place in a school.

The fast-track learning approach, tailoring content as feedback was received, showed that adaptation was able to happen in 'real time.' Coordinators and facilitators conducted rapid analysis of feedback and discussion results, which allowed them to make micro adjustments and improve the structure and flow of the sessions. For example, in one of the Salvador groups, the facilitators identified an issue ahead of the session that discussed toileting (none of the children in the group were yet able to use a toilet). They quickly adapted the session so that they could spend more time on practical aspects for that group (e.g., diaper changing).

The final groups in Rio and Salvador both included a number of children with CP. The programme showed that it was adaptable for caregivers of children with Zika and non-Zika-related neurodevelopmental disabilities.

Limited efficacy

The study was not powered to show impact. Efficacy limitations were assessed in order to identify domains of potential impact for future studies.

Data from the PEDSQL showed improvement in parent-reported outcomes from baseline to end-line across all dimensions except cognitive functioning (see Table 4 below). Baseline scores of the Juntos participants were similar to those recorded using the same scale in another study which focused on the social and economic impacts for caregivers of children with CZS[8]. Only two of the PEDSQL were statistically significant: Daily activities (p=<0.001) and the Family Functioning Summary Score (<0.001).

In the Cantril Scale measurement, there was an increase from 5.6 to 6.5 (out of 10) for the self-reflection on happiness of the caregivers between baseline and end-line (n=37, p=0.007). There was no change in the perceived happiness of the child (n=35, 7.3 at baseline, 7.4 at end-line, p=0.48).

Qualitatively, there were several positive reflections which emerged from the interviews relating to different focus areas of the programme. One mother, for example explained how Juntos helped her with feeding and caring for her child outside of the home:

> *"Today she eats very well. The programme helped me a lot with that. When I arrived, I was lost on how to deal with [child] outside the house, at home I had her under control, completely, but out, how would it be?"(Participant, Rio)*

Table 4. Changes from baseline to end-line scores across the dimensions of the PEDS QL.

Dimensions of PedsQL	Baseline (n=36) mean (SD)	End-line (n=36)	p-value (t-test)
Physical functioning	50.1 (37.5)	51.8 (37.2)	0.46
Emotional functioning	57.3 (35.1)	60.2 (33.4)	0.29
Social functioning	59.1 (39.9)	59.3 (38.2)	0.95
Cognitive functioning	66.5 (34.1)	61.7 (32.7)	0.06
Communication	60.2 (41.4)	64.1 (36.9)	0.40
Worry	36.1 (41.7)	38.0 (42.8)	0.50
Daily activities	33.5 (36.8)	48.3 (38.5)	<0.001*
Family relationships	60.6 (37.3)	66.4 (32.8)	0.07
Total score	**53.4 (16.9)**	**56.0 (14.0)**	**0.36**
Parent HRQL summary	**57.8 (37.0)**	**57.9 (35.6)**	**0.96**
Family functioning summary	**50.6 (39.3)**	**59.7 (36.0)**	**<0.001***

Another potential impact is the increased confidence and trust for mothers to be able to leave their children for periods of time with relatives or even in daycare. One mother explained how seeing her child doing so well with the volunteers has given her confidence to pursue job opportunities:

> *“I already have a clearer idea of the fact that I want to put my daughter in daycare if I need to, especially if I get accepted into the job I applied to. Before I would say, "I don't want to take the job because I don't have the courage to leave [child] but now, no, I will already look for a place because I saw that it is a good thing”* (Participant, Rio)

Another mother described how being able to leave her daughter with someone else was a goal that she had achieved:

> *“[The goal] was to be able to spend a little time without her, because we are always together, yes... I think it was good for her as well as for me, for me it was great. Because we need to have these moments, you know? Because it's me and her the whole day, - twenty-four hours a day, like glue, I think it was good for her because she learnt to be a bit less attached to me and learnt that other people can also treat her the same way, to caress her, and it doesn't need to be me.”* (Participant, Rio)

A third mother shared how the programme had allowed her to take the pressure off herself and accept others to provide support.

> *I learnt that we can count on other people, that it doesn't mean that it can only be the dad or the mum, or her little brother, but that we can look for support in other family members like her uncle, godmother, godfather. That we need to trust to leave her with these people. So it was great that I learnt that here in the group too. That we have family members, so why don't we seek their help? Not only these family members but the community as a whole.”* (Participant, Salvador)

Discussion

The study generated evidence supporting the feasibility of the Juntos programme in several core domains. Our findings have similarities to studies on the Getting to Know Cerebral Palsy programme in Ghana[21] and on the Early Intervention Programme in Uganda[32].

Juntos has demonstrated acceptability. Participants valued being given an opportunity and an outlet to process their emotions and share day-to-day life experiences in a safe place with their peers. The potential for demand was also demonstrated. Participants, facilitators and key stakeholder interviews all suggested that parents of children with CZS and other neurodevelopmental disabilities would likely welcome the programme across Brazil. This echoed the key findings from the needs analysis that was conducted ahead of the study[19]. However, recruitment of participants was somewhat of a challenge and the completion rates and attendance of some sessions could have ideally been higher. Further exploration is needed to understand why some participants discontinued the programme. Implementation and practicality were demonstrated to a certain extent. The combination of a mother expert and therapist facilitator seems to work well, and the two differing profiles brought a complementarity to the approach. The Juntos manual was reported to be easy to follow by the facilitators and sessions took place in a range of settings. Location was an important factor for the participants and the provision of transport and refreshment options, although adding to cost, seems to have been another positive factor for the group participation. As a pilot initiative, costs at 7,460 GBP per group were relatively high, but we believe they could be potentially reduced if implemented at scale. A level of adaptability has been demonstrated by implementing the programme in two different contexts and in different settings, and the ability of groups to encompass children with non-Zika-related neurodevelopmental disabilities.

The suggestion of positive caregiver outcomes in the PED-SQL, in the dimensions of daily activities and in the Family Functioning Summary Score, is encouraging. It is logical to observe improvements in these areas, given the nature of the programme and its focus on strengthening the networks providing support to the immediate caregivers. Improvements in the Cantril Scale for perception of quality of life among caregivers also points to potentially promising efficacy of the programme. Some of these sentiments were echoed in the participant interviews, such as the mother who had begun to recognise that raising her child was everyone’s responsibility in the family. The reflections from some caregivers on increased confidence to leave their child with others is encouraging. Some mentioned that this may allow them space to look for work opportunities, and given that only 13% of mothers worked (compared to 70% of fathers) this demonstrates potential impact for the mother and the family’s economic security.

It is important to note that the sample numbers were too small to make any broad conclusions/generalizations on efficacy limitations. Given that the focus of the intervention was on the caregiver rather than directly on the children, and the relatively short (10 week) duration, we did not hypothesize that there would be any significant change in the functional status of the child and this was shown by our findings. The reasons for the decline in the cognitive functioning dimension of the PEDSQL, which includes aspects such as attention, remembering and thinking quickly, are not clear. As with the positive results, few conclusions can be drawn with such a limited sample size. However, if Juntos is scaled up, further scrutiny may be warranted. Baselines of the PEDSQL scores were broadly similar to those taken from a different cohort of caregivers for a social and economic impact study[19].

Scaling up the Juntos programme to other areas of Brazil will take important considerations. However, there is potentially a strong case for doing so: a high need has been demonstrated; it can be extended for families of children with other neurodevelopmental disabilities; it fills an existing gap at community level for families and, with a wide network of community services through healthcare or other sectors, there is potential for its integration into existing structures. However, this will require investment and it would be pertinent to continue to research any future programmes, particularly looking at the areas of integration, expansion and limited efficacy.

Strengths and limitations

The study had a number of strengths and limitations, which need to be taken into account when considering the results. Strengths included the use of a mixed methods approach (e.g., questionnaires, interviews, focus groups, data analysis) to assess the six areas of feasibility. This allowed, to a certain extent, for triangulation of these different sources of data. For example, acceptability from participants could be assessed through satisfaction scores in the questionnaires, through interviews and focus groups. We collected data from two different settings and across six groups, which allowed for comparisons between groups and locations. Having two researchers per group was also a strength. Observations could be validated, and different viewpoints reflected.

In terms of limitations, the small sample size means it is not possible to draw any firm conclusions with regards to limited efficacy of the programme, despite the promising results seen in the PEDSQL and Cantril ladder. Furthermore, the selection of interview participants, although done to gather a range of perspectives, may have introduced an inherent bias to the qualitative data that was analysed from the participants. Reasons for participants dropping out of the group were not fully explored in this pilot test, which limits any conclusions made on acceptability, demand and/or practicality. Finally, since the focus of the research was on people already in the programme, we were unable to ascertain reasons why those who never enrolled, chose not to partake in Juntos.

Conclusions

Juntos has shown that it potentially ‘can work’ according to six of Bowen *et al.*’s eight areas of feasibility. Nevertheless, more research is needed before conclusions can be drawn about whether it ‘will’ or it ‘does’ work. This will require scale-up of the programme to capture data from a wider number of participants. A scaled-up programme would also allow measurement of integration and expansion, which were two areas of feasibility that this pilot did not explore.

Data availability

Underlying data

Data associated with this study will not be made freely available, owing to the small number of children with CZS, making data potentially identifiable, and the sensitive nature of the subjects discussed in the interviews and from the questionnaires. However, we are committed to collaborating with other researchers in the analysis of our data (full questionnaire available online). Applications for access to the raw data for this study should be made by contacting Professor Hannah Kuper (hannah.kuper@lshtm.ac.uk), or Mr Antony Duttine (antony.duttine@lshtm.ac.uk) and outlining the purpose of the proposed analyses and the variables requested. These applications will be reviewed by the three researchers, and if accepted, the requested variables will be shared.

Extended data

Open Science Framework: Assessment of the feasibility of Juntos: A support programme for families of children affected by Congenital Zika Syndrome, https://doi.org/10.17605/OSF.IO/AFYBS[28]

This project contains the following extended data:

- 3. pre and post questionnaires.xlsx
- Qualitative interview questions - participants.docx
- Qualitative interview questions - facilitators.docx
- Qualitative interview questions - key informants.docx

Data are available under the terms of the Creative Commons Attribution 4.0 International license (CC-BY 4.0).

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CHAPTER 7: DISCUSSION

Figure 14: Rio facilitators and researcher team

Summary of key results

The aim of this DrPH research was to assess whether a community intervention aimed at families of children with CZS in Brazil is needed, acceptable and feasible. As described in chapter one, the **specific objectives** of the research project were:

1. To undertake a needs assessment for the intervention.
2. To adapt *Getting to know Cerebral Palsy* for the Zika context in Brazil.
3. To pilot test the intervention with two parent groups in Rio de Janeiro and Salvador.
4. To assess feasibility of the intervention with four parent groups in Rio de Janeiro and Salvador.

These objectives are expanded on below:

To undertake a needs assessment for the intervention

A need for a community support intervention in the context of the 2015-16 Zika epidemic in Brazil was demonstrated and described in the second paper [2]. A mixed methods approach, combining literature searching; analysis of early results from a sister study exploring the social and economic impact of Zika [124]; and a scoping visit undertaken in April 2017 were incorporated.

As the literature review took place in April-July 2017, few papers had yet been published specifically on caregiver needs. The few papers that had been published identified similar trends to contained in the literature about caregiver needs of children with other NDDs – mental health, access to healthcare and quality of life. Emerging themes from the social and economic study [124] seemed to mirror many of the literature search findings – higher levels of anxiety, depression and stress, and healthcare access challenges and costs.

The scoping visit, which included visits to three sites (Rio de Janeiro, Salvador, and Recife) helped to identify some of the specific gaps in services and needs of the caregivers. In particular it identified that while many children with CZS were accessing individual medical or therapy services (generally at secondary or tertiary health facilities rather than nearby in their community), parents still had gaps in their knowledge, and they generally lacked supportive peer-to-peer networks. It was concluded that caregivers of children with CZS were likely to have (and, to a degree, were already demonstrating that they did have) specific and unmet needs such as educational gaps, psychosocial difficulties and a lack of peer-to-peer support that could benefit from a structured programme such as *Getting to Know Cerebral Palsy*. This provided the rationale for adapting *GTKCP* and the *EIP* for the Zika context in Brazil.

To adapt Getting to know Cerebral Palsy for the Zika context in Brazil

As described in the third paper [3], GTKCP and EIP were successfully adapted for the context of CZS in Brazil. The adaptation process involved working with specialists in both the UK and Brazil to restructure the programme based on emerging needs ascertained in the needs analysis and with extensive feedback from the *GTKCP and EIP* team leads. The resulting programme was a ten-session course that bore several of the key hallmarks of the *GTKCP and EIP* programmes, but with some important changes such as the regular end of session mental health 'check ins', the use of a mother as a facilitator expert and reimagined chapters on *'Our Child' and 'Raising Our Voices'*. Furthermore, a module was added on 'Our Community', where caregivers invited 1-2 members of their close community to join for part of the session as a culmination of the programme and to encourage ways they can be more involved in supporting the caregivers to meet the needs of the child. Figure 15, below, shows the final structure of *Juntos* (after all phases were completed) [3].

Module	Topics
1. Introduction	• About the program • Information about Zika and Congenital Zika Syndrome • How to find information • Personal stories
2. Our child	• Introducing your close family and friends • Development milestones for young children • Determining your child's progress • Managing irritability and crying
3. Positioning and moving	• How to position children who need assistance • How to assist children to learn to move
4. Eating and drinking	• Feeding challenges • Practical skills to address challenges for your child
5. Communication	• Importance of communication • Practical advice to help your child communicate
6. Play and early stimulation	• Importance of play for children to develop and learn • Early stimulation • Making simple toys • Inclusion of play in the family and broader community
7. Everyday activities	• How to use everyday activities to help your child develop • Managing seizures
8. Uniting our voices	• Understand the context of disability rights • Education • Communicating with your health team • Advocating
9. Our community	• Who is in your community? • Common barriers to inclusion • Addressing negative attitudes and exclusion • Social activity
10. Next steps	• Summing up • Planning next steps for yourself and the group

Figure 15: Finalized Module Topics Included in Juntos, A Community-Support Group for Caregivers of Children with Congenital Zika Syndrome in Brazil taken from Duttine et al (2021)

The programme was designed so that groups meet weekly, and sessions are led by two facilitators – an expert mother (who is a parent of a child with CZS) and a rehabilitation professional. They follow a guidebook that takes them through the specific activities for that day. Activities are often highly participatory including icebreakers, quizzes, small group activities and practical sessions, which aim to optimally engage the participants. Each session ends with a half hour psychosocial 'debrief' where parents are asked to share any concerns or worries that they have experienced that past week or any particular highlights that they have had.

To pilot test the intervention with two parent groups in Rio de Janeiro and Salvador

The piloting of *Juntos* is also described in the third paper [3] where one group in each of Rio de Janeiro and Salvador became the first to successfully complete the *Juntos* programme. Sessions were observed by researcher teams. In addition, participants were interviewed via focus groups after each session which provided real time feedback on the content and structure of the programme. This allowed us to make timely, positive changes for the subsequent groups.

Before the sessions could commence, facilitators were identified and trained at each site. This comprised of two therapists and two expert mothers for each location (n=8). All eight facilitators were trained in Rio in July/August 2017. An international expert, who had been involved in facilitator training for *GTKCP,* led the training for the facilitators. One expert mother and one therapist from each site were then selected to lead the first pilot groups, with the second mother/facilitator pairing acting as an observer and backup in case of emergency. Overall, 15 families (seven in Rio and eight in Salvador) were enrolled and undertook the programme. Sites were selected in downtown Rio and in Camaçari, a town just north of Salvador. Sessions ran from August to November 2017. The first pilot group tested the first iteration of the programme and real time feedback allowed for adjustments to be made for the four programmes delivered in the second phase of the project.

It should be noted, as described in paper 3, that we did not set strict criteria for the severity of disability that the child with CZS had. The main criteria related to whether the child had received a CZS diagnosis, the caregivers were willing to participate in the programme and did not live a great distance from the programme location. We did also include the fact that the child should be at home (rather than in hospital) since it would not be feasible to bring a child with ongoing acute medical care needs. We did not collect Gross Motor Functioning Classification System (GMFCS) or other similar functioning scores for the children, as discussed later in the limitations section. Anecdotally many of the children had more severe levels of impairment that tended to be enrolled, in part since they were already identified and being supported in the health system.

To assess feasibility of the intervention with four additional parent groups in Rio de Janeiro and Salvador

The four additional groups ran from January until June 2018 and a further 33 families (16 in Rio and 17 in Salvador) were enrolled in the programme. This followed adjustments to the structure of *Juntos* and an 'upgrade training' for the facilitators.

Feasibility was assessed using the framework proposed by Bowen et al [5] and is described in the fourth paper [4]. The mixed-methods approach for data collection involved in-depth interviews with purposefully selected participants, all facilitators and three key stakeholders. Baseline and endline questionnaires were completed by participants and post-session group discussions, researcher session observations and cost analysis were also used to evaluate the different areas of feasibility.

Of note, improvements in the PEDS QL Family Impact module scores, particularly in the domains of *family relationships* and *daily activities*, are encouraging. Similar hallmarks were seen in studies of *Getting to Know Cerebral Palsy* in Ghana [17] and the *Early Intervention Programme* in Uganda [18]. Qualitatively, there was a myriad of positive remarks from all three of the interview groups (participants, facilitators, and key stakeholders) about the impact of the programme. Many caregivers (particularly mothers) described how this programme had filled a social, emotional, and practical support gap that they needed. In the words of one of the participants, *Juntos* was like "a huge hug".

Since the project was a pilot, it was easier to gather useful data and draw conclusions on some of the areas of Bowen's feasibility framework than others. *Acceptability, Demand* and, to a certain degree, *Implementation* contained a stronger evidence base than *Practicality or Adaptation.* However, *Practicality and Adaptation* these also had more evidence than *Integration* and *Expansion,* which we eventually did not include in the feasibility analysis. *Limited Efficacy*, as discussed in the fourth paper, could be assessed, but relatively few conclusions could be drawn due to the small sample size.

Other findings to emerge from *Juntos* evaluation.

Outside of the feasibility analysis, we were also able to gather some useful data on the *Juntos* programme which could inform future programme planning. These dimensions particularly relate to the engagement of fathers in the programme [125] and the role of mothers as facilitators [126]. Peer-reviewed articles were written and published in relation to each of these two areas, and I was a co-author of both (full texts included as Annexes 4 and 5).

We found that fathers did not participate in the programme in large numbers [125]. This may have been in part since sessions took place during the working day, midweek. However, there was a sense from an interview with one of the fathers that perhaps this type of format was not best suited for or the most conducive to male participation. Perhaps more surprisingly in the focus groups, some of the mothers were uncertain whether they would like to have their partners present. Some felt the programme offered them a space to share and open up about familial dynamics and they would feel more reserved if their partners or other fathers were present. This issue was not explored a lot further but could be an important consideration in terms of how to provide support to fathers as well as mothers in future programmes.

The decision to have a mother as a co-facilitator was an intentional one, and contrasts with the previous GTKCP programme, which was delivered by a therapist only [127]. The co-facilitation by mothers certainly seems to have only had positive effects, such as greater acceptance and connection to the programme from participants and building of confidence and independence of the expert mothers (they were paid at an equal rate to the therapist facilitators). An important aspect to this initiative was to ensure that the “professional” (therapist) was fully bought into the idea that the two facilitators were equal partners in the endeavour, and each brought their own expertise that the other could not.

The findings from this study are also consistent with literature from other global health and disability movements. Firstly, there is a strong emphasis on the importance of Early Childhood Development (ECD) [128]and the important role that parents play [129]. Secondly there is a growing awareness and promotion of the need to include children with disabilities within ECD actions [130] and to connect this to wider coordinated efforts for inclusion, including education [131, 132].

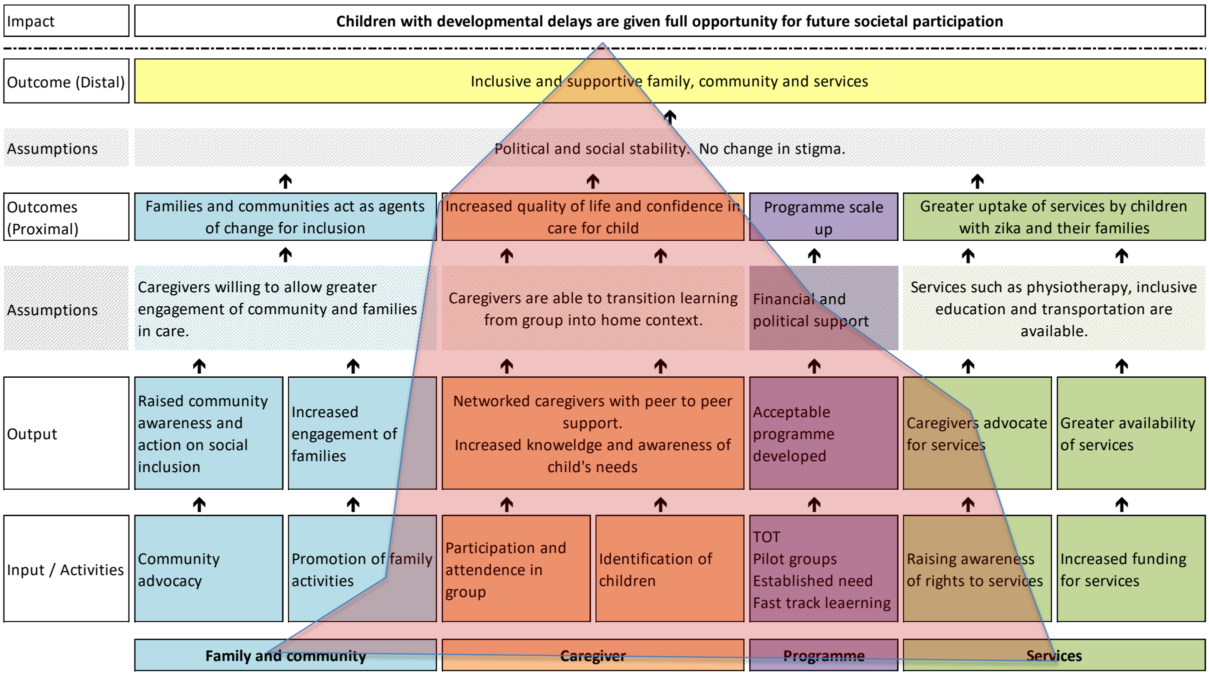
Findings in the context of Bowen's framework to assess feasibility.

The feasibility framework proposed by Bowen et al [5] has been a valuable guide for the design and analysis of this intervention. Taking such an in-depth dive into a tool and applying it to an intervention has also given me a chance to reflect on where the tool itself may be most beneficially used for future feasibility endeavours. Bowen suggests that all areas of focus can be applicable and relevant at all development phases. While I agree that this is true, I feel it could be also beneficial to highlight which areas may lend themselves more naturally at different stages of an intervention's development. *Acceptability, Demand and Implementation* may be most valuable when measured at the ”can it work?” stage; *Practicality, Adaptation, Integration and Expansion* at the “does it work?” stage; and *Integration, Expansion and Limited Efficacy* at the “will it work?” stage. These largely go with the chronological sequence of the list, so it may well be placed in that order deliberately.

Acceptability, Demand, and Implementation are extremely important in ascertaining if an intervention ‘can work’ and should inform new pilot initiatives. It can be argued that while the other five areas can (and perhaps should) be improved through adjustments to the intervention over the course of its development, *Acceptability, Demand, and Implementation* are the building blocks upon which a good intervention must be based. Having an unacceptable, unneeded, or unimplementable intervention requires a radical rethink and redesign and these issues are best identified at the “can it work?” stage. *Practicality, Adaptation, Integration* and *Expansion* are most helpful at the “does it work?” stage as they provide valuable insights into potential adjustments that may be required before the intervention is scaled up too much to have optimum impact. *Integration, Expansion and Limited Efficacy* is most relevant when demonstrating if an intervention “will work?” as these are the key questions that may help decision makers make evidence based decisions as to whether the intervention warrants major scale up.[1] Another study, looking at a very different area of work, but also using Bowen et al's framework came to similar conclusions about its potential applications: Wilbur et al focussed on the first four of the eight areas and suggested that the latter four would be better assessed in the next phase of scale up [133].

Adjustment of theory of change

Bringing this work together, allowed the group to reflect on previous work and develop the Theory of Change. Figure 16, below, shows a slight adjustment to the ToC with the addition of an 'iceberg' which shows where *Juntos* really appears to be affecting. As can be seen, the middle two columns were the areas most directly impacted by *Juntos*.



Impact	**Children with developmental delays are given full opportunity for future societal participation**						
Outcome (Distal)	Inclusive and supportive family, community and services						
Assumptions	Political and social stability. No change in stigma.						
Outcomes (Proximal)	Families and communities act as agents of change for inclusion		Increased quality of life and confidence in care for child		Programme scale up	Greater uptake of services by children with zika and their families	
Assumptions	Caregivers willing to allow greater engagement of community and families in care.		Caregivers are able to transition learning from group into home context.		Financial and political support	Services such as physiotherapy, inclusive education and transportation are available.	
Output	Raised community awareness and action on social inclusion	Increased engagement of families	Networked caregivers with peer to peer support. Increased knoweldge and awareness of child's needs		Acceptable programme developed	Caregivers advocate for services	Greater availability of services
Input / Activities	Community advocacy	Promotion of family activities	Participation and attendence in group	Identification of children	TOT Pilot groups Established need Fast track leaerning	Raising awareness of rights to services	Increased funding for services
	Family and community		**Caregiver**		**Programme**	**Services**	

Figure 16: ToC with 'iceberg' showing the areas where Juntos' impact is most likely.

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[1] expansion and integration could be swapped depending on the nature of the intervention and context. Oftentimes, it is the integration of an intervention into existing programmes or services that are the final scale up solution that is necessary and showing feasibility of expansion may be the way to demonstrate ease or possibility of integration

To have the fullest impact, *Juntos* needs to be implemented in coordination with other actions that are particularly focussed on the outer two columns of the ToC: Family and Community and Services. *Juntos* is not designed to replace specific services such as rehabilitation but need to complement and coordinate with these services so that participants get the full support that they need. Anecdotally, during the piloting of *Juntos*, facilitators provided feedback for participants to discuss with their therapists or medical team about the individual care that their child was receiving. This created a link between *Juntos* and their medical care. Similarly, *Juntos* does not go into wider aspects of community change and awareness raising, beyond the two sessions later in the programme that focus on "raising our voices" and "our community". Activities undertaken as part of these sessions may empower caregivers to initiate societal change, but this would also depend on the creation of broader community groups.

Key learnings from *Juntos*

Juntos is an adaptation of existing programmes which have been tested and deemed positive for their audience. As an intervention it displays several strengths:

- The approach of *Juntos* appears to be its biggest strength. It was intentionally designed using participatory approaches. It had a structure/set course, but it was also built to be flexible which allowed the caregivers to tread their own path and make the programme personal.
- Having mothers as facilitators, in equal partnership with a trained rehabilitation professional, provided a good balance and optimised group sessions. Certain modules or sessions lent themselves to being led by a therapist, with a background and expertise in these areas. However, the expert mother was in a better position to lead on other areas and draw from her own personal experience. She was able to broach topics and subjects that might feel uncomfortable or inappropriate for others.
- Having the children of caregivers on-site, but out-of-sight, was another strength. Mothers remarked frequently that they felt that they were the only person responsible or capable of

caring for their child. With the ethos of *Juntos* being about how caregivers can better empower themselves and draw from their support networks, the opportunity to directly engage with them, without a focus on the child, was important. it also showed a caregiver that others could be responsible for the care needs of their child.

Limitations of *Juntos* included:

- *Juntos* does not replace the individualised rehabilitation, psychosocial or other medical and social interventions which a child with CZS and her caregiver may require on an individual basis. Given the gaps in availability of these services, however, it is inevitable that facilitators will be asked questions about the specifics of a particular child or caregiver. Therefore, it is Important that facilitators (both expert mothers and therapists) understand the scope and limitations of their role.

Strengths and limitations of study

The study provided valuable initial insights into the feasibility of running *Juntos* as a standalone programme. There were, however, several limitations to this feasibility assessment. As discussed in the fourth paper, the sample size was small and cannot be well generalised for a broader population within Brazil or in other contexts. Despite encouraging feedback on the acceptability and limited efficacy of the programme, these were not powered to show efficacy and a full Randomised Control Trial would be needed to do so. Furthermore, feasibility was only partially assessed. While there were useful assessments of six out of eight of Bowen's framework. Two important areas - *expansion* and *integration* - were not deeply explored. These are two key areas to explore when it comes to scale up of an intervention. The in-depth interviews with the key stakeholders (site coordinators) did flag some concerns and considerations with regards these two areas, but they were more speculative than based on concrete findings from the pilot. There is a potential of bias in that not all participants were selected for in depth interviews, nor was it a random selection but rather an intentional selection based on the subjective assessment of the researchers. The researchers were briefed to try to identify interviewees who would bring a diversity of opinions and had a diversity of backgrounds. For the area of *practicality*, the calculation of the financial costs was also somewhat limited by only having limited access to the Brazilian side of the budget. The design of the project meant that each group (LSHTM and FIOCRUZ) handled their own budget and project costs (design, training, research coordinator and travel costs) were integrated into the LSHTM budget and the implementation of the interventions, Brazil coordinator and researcher salaries were integrated into the Brazil budget. As previously mentioned, I also did not collect specific data on the children are particularly around their level of functioning (for example through collection of GMFCS). Therefore, I did not assess whether caregivers of children with different severities of disability may benefit more than others. This may be beneficial to consider in future studies. That said, one of the important considerations, when designing the programme, was to include caregivers of children whose impairments may have been more mild or moderate since there was a strong sense during the needs assessment that this group may be at higher risk of not having access to rehabilitative or other care services.

The qualitative analysis of the participant interviews was largely undertaken by a Portuguese speaking researcher under instruction from me. I established a framework for identifying and selecting quotes that fit into one of the areas of Bowen's feasibility framework. The researcher then reviewed the Portuguese transcription and selected quotes at her discretion before translating them into English. I validated the English translation to ensure that the quotes fit within the specific area of feasibility but as a non-Portuguese speaker was unable to confirm if other parts of the interviews contained additional information that could have been useful or relevant. Quotes were not revalidated with the participants.

Despite the limitations listed above, the study had several strengths. *Juntos* was had an excellent, cross-functional team with mixed skills who contributed towards the development and delivery of *Juntos* and ensured its success. There was also a strong link with GTKCP which facilitated development of Juntos. There was good support from therapists and parents which ensured delivery of the intervention. The mixed-methods approach and range of data collected allowed a relatively comprehensive analysis for six of the eight areas of feasibility. Bowen et al suggest that a different approach may be needed for each area and a study may only be able to explore one to two areas, so to achieve detail on six is positive. Although it was not intentional, all the researchers who collected the data all had profiles in psychology. This proved to be an invaluable resource, especially when adjusting the programme between the first and second phases to ensure that the mental health needs of participants were raised and addressed in an appropriate way.

Scaling up the intervention: policy and organisational considerations

The initial results from the feasibility analysis are encouraging in that the *Juntos* programme was clearly welcomed and accepted by most of the caregivers who participated. This is reflective of similar findings of studies for GTKCP and EIP [114, 116, 127, 134]*,* as well as a recent implementation of *Juntos* in Colombia [135]. Being a relatively small-scale project and with only six groups of 48 participants, it's difficult to draw any strong conclusions with regards generalizability of the feasibility analysis. We have determined that according to Bowen et al's feasibility framework that *Juntos* "can work" but cannot say with any statistical confidence whether it "does" or "will work". While some aspects of feasibility have been shown from the pilot intervention, most notably *acceptability, demand* and *implementation*, *Juntos* was piloted as a standalone project with external funding. If the ultimate aim of *Juntos* is to reach as many families as possible, and have the widest potential benefit, it is essential to consider the next steps for scaling up. Two of the areas of Bowen et al's feasibility framework: *expansion* and *integration* are likely to be key in ascertaining whether *Juntos* has longer term sustainability and/or can become an integrated initiative within a national or regional programme.

Drawing from knowledge acquired from other elements of the DrPH programme, there are several important considerations that need to be considered for *Juntos* to be effectively scaled up in Brazil and beyond:

Firstly, as described in the introductory chapter, the health sector in Brazil is divided into Federal, Provincial and District departments, each with their own budget and different levels of autonomy [71]. It seems unlikely that a small pilot project would be immediately considered at the Federal level for national uptake as it has not yet been demonstrated that it “does” or “will work”. Much more likely are Provincial or District health departments. Secondly, there is an assumption that the health sector would be the most appropriate to take the mantle of scaling up *Juntos.* This is due to the somewhat medicalised approach and needs of children in early years, the recommendation of a rehabilitation professional to be a co-facilitator of the programme (who would be potentially easier to identify and utilise if already working in the same sector) and the fact that the health sector usually has facilities reaching out into community level though primary health systems (SUS). There is n reason, however, why *Juntos* cannot be housed outside of the health sector such as the non-governmental sector or in another governmental agency (e.g. the Ministry of Education (Ministério da Educação) or the Ministry of Social Welfare and Security (Ministério de Previdência e Assistência Social)). Lastly, scaling up will require this programme to be a priority for the Ministry of Health (or other Ministries or Agencies as suggested above), and for it to be included in project plans and budgets. The fact that *Juntos* has been applied to children with other neurodevelopmental disabilities, and not just CZS, could be a major positive since the cumulative burden of these conditions creates a major need and would support a wider range of families.

CHAPTER 8: CONCLUSIONS

Figure 17: The Juntos logo

Juntos is an adaptation of an existing programme for caregivers of children with neurodevelopmental disabilities that was developed rapidly out of necessity in the context of an acute epidemic in Brazil. It is a 10-session program for parents and caregivers of children with congenital Zika syndrome, and is based on a similar program for lower resource contexts for caregivers of children with cerebral palsy.

Juntos was piloted in Salvador and Rio de Janeiro in 2017 and 2018. Six groups were successfully run with 48 families taking part in some or all of the program. Topics covered in the course include sessions around positioning, play, feeding and drinking, engaging in their community and raising their voices. As such, the program is designed to both provide a framework of practical tools that caregivers can use in supporting them to raise their children, but also to empower them to ensure they have their needs met and stand a better chance at being included in and participating fully within society.

Despite this project being a pilot, and on a relatively small scale, *Juntos* has shown an encouraging level of success and has demonstrated feasibility when considering Bowen's question of "can it work?". The areas of acceptability and demand, in particular, have proven successful. The caregivers reported that before the programme they often felt isolated and, despite often having access to specific services such as rehabilitation, they still felt under-supported and that they had significant gaps in their knowledge that this program helped to fill.

The project has shown certain innovative approaches, including collecting real time feedback from participants, facilitators and researcher observers during the first two pilot groups helped adjust, inform and improve the subsequent groups. By adopting this approach, we showed that there was a degree of flexibility that can apply to the Zika context, and we could repurpose content to meet the needs of a specific group. In the future, the same approach could be used for adapting Juntos from its current form to new contexts or with new groups.

The use of an expert mother as a co- facilitator to a therapist brought a valuable dynamic to the group, and perhaps allowed for exploration of areas that otherwise would not have been possible. For example, therapists were often able to bring a clinical background and knowledge into discussions around positioning and answer specific questions that parents may have on symptoms or clinical presentations. Meanwhile, expert mothers often led the discussions on mental health, and they led the end of session check-ins as they clearly had a greater level of connection and trust could since they could empathise with the participants.

Juntos has already successfully been tested outside of Brazil, but the Bowen's questions of 'will it work?' and 'does it work' remain largely unaddressed. In particular, more research exploration is needed in the area of *expansion*, *integration* and *efficacy*. Ultimately research should be aiming for a large-scale randomized trial to truly show effect and impact.

Moreover, it is important to scale availability of the programme. Over 5000 children have been born with congenital Zika syndrome who are likely to have lifelong care needs and many more children are born each year in Brazil with other neurodevelopmental disabilities who could benefit from such a programme.

Scaling up of the programme will require policy, programmatic and research support. From a policy perspective, there will be a need to consider how this programme fits into more broad public health (and perhaps in the future education) strategy and actions within Brazil. For example, how it fits into primary health programming or child healthcare agendas. From a programmatic perspective, this will necessitate a deeper dive into the costs and practicalities for running such a programme at larger scale including considerations around training a network of facilitators, identifying lead actors (whether governmental or non-governmental) who will take the initiative forwards and of course acquiring funding to implement a greater number of groups.

Although the threat of Zika has lessened, there is still a risk of the virus mutating and a new outbreak beginning. Even if many fewer children with CZS are being born, those with the condition, and their caregivers, are likely to have ongoing support needs. Further, there will always be children born with other neurodevelopmental disabilities and establishing such a program is a crucial step to ensure that both the caregivers and the children have the best possible opportunity for success.

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ANNEX 1: ETHICS APPROVALS

London School of Hygiene & Tropical Medicine
Keppel Street, London WC1E 7HT
United Kingdom
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Observational / Interventions Research Ethics Committee

Mr Antony Duttine
LSHTM

3 August 2017

Dear Mr Antony Duttine

Study Title: Development of an early intervention to support babies with Congenital Zika Syndrome and their families

LSHTM Ethics Ref: 13608

Thank you for responding to the Interventions Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

Approval is dependent on local ethical approval having been received, where relevant.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document Type	File Name	Date	Version
Protocol / Proposal	Concept note Jan 12	12/01/2017	1
Investigator CV	ADuttine CV Jan 2017	23/02/2017	1
Information Sheet	Media ethics consent form	15/03/2017	1
Protocol / Proposal	Data management plan	30/05/2017	1
Protocol / Proposal	Qualitative interview questions - facilitators	30/05/2017	1
Protocol / Proposal	Qualitative interview questions - participants	30/05/2017	1
Protocol / Proposal	Qualitative interview questions - key informants	30/05/2017	1
Information Sheet	Consent forms (interviews and questionnaires)	30/05/2017	2
Protocol / Proposal	Questionnaire 31.5.17	30/05/2017	1
Protocol / Proposal	Protocol for ethics	30/05/2017	1
Covering Letter	Ethics response letter 25.7.17	25/07/2017	1
Protocol / Proposal	Consent forms v2 (interviews and questionnaires)	27/07/2017	2

After ethical review

The Chief Investigator (CI) or delegate is responsible for informing the ethics committee of any subsequent changes to the application. These must be submitted to the Committee for review using an Amendment form. Amendments must not be initiated before receipt of written favourable opinion from the committee.

The CI or delegate is also required to notify the ethics committee of any protocol violations and/or Suspected Unexpected Serious Adverse Reactions (SUSARs) which occur during the project by submitting a Serious Adverse Event form.

An annual report should be submitted to the committee using an Annual Report form on the anniversary of the approval of the study during the lifetime of the study.

At the end of the study, the CI or delegate must notify the committee using an End of Study form.

All aforementioned forms are available on the ethics online applications website and can only be submitted to the committee via the website at: http://leo.lshtm.ac.uk

Additional information is available at: www.lshtm.ac.uk/ethics

Yours sincerely,

Professor John DH Porter
Chair

ethics@lshtm.ac.uk
http://www.lshtm.ac.uk/ethics/

PARECER CONSUBSTANCIADO DO CEP

DADOS DO PROJETO DE PESQUISA

Título da Pesquisa: Análise de um Programa de Intervenção precoce para lactentes com síndrome congênita do vírus Zika e suas famílias: pertinência e viabilidade do programa

Pesquisador: maria elisabeth lopes moreira

Área Temática:

Versão: 4

CAAE: 70525217.5.1001.5269

Instituição Proponente: Instituto Fernandes Figueira - IFF/ FIOCRUZ - RJ/ MS

Patrocinador Principal: The Wellcome Trust

DADOS DO PARECER

Número do Parecer: 2.183.547

Apresentação do Projeto:

Esta pesquisa, parte de um projeto financiado pelo Wellcome, visa desenvolver e testar um projeto piloto para a adoção de um programa existente, "Conhecendo a paralisia cerebral" (Getting to know cerebral palsy) no contexto da SCVZ. O programa consta de uma série de10 sessões de grupo com os familiares, que serão facilitadas por especialistas locais, que busca orientar as famílias e fornecer apoio e aconselhamento sobre como estimular as crianças com SCVZ. O objetivo desta pesquisa é determinar a pertinência e viabilidade (inclusive a aceitabilidade) de uma intervenção comunitária direcionada às famílias de crianças com SCVZ. A intervenção piloto ocorrerá em dois locais: Rio de Janeiro e Salvador. Um grupo piloto inicial de aproximadamente 8-10 famílias de crianças com SCVZ será estabelecido em cada local, com um programa de aprendizagem acelerada. Posteriormente, dois grupos serão testados em cada local, somando um total de 48-60 famílias participantes (16-20 piloto, 32-40 (teste). A viabilidade da intervenção será descrita através de 8 áreas centrais propostas por Bowen et al (2009):
aceitabilidade, demanda, implementação, praticidade, adaptação, integração e eficácia limitada. Estes fatores serão medidos através de métodos qualitativos e quantitativos variados, incluindo

questionários, entrevistas semi-estruturadas e descrição de custo. Uma demonstração positiva de demanda, viabilidade e aceitabilidade levará a uma maior divulgação do programa dentro e fora do Brasil, além de pesquisa adicional sobre o impacto do programa na qualidade de vida das crianças com SCVZ e suas famílias.

Objetivo da Pesquisa:

Objetivo Primário:

-Analisar se uma intervenção comunitária em famílias de crianças com SCVZ no Brasil é efetiva, aceitável e viável.

Objetivo Secundário:

- Adaptar o programa "Conhecendo a Paralisia Cerebral" ao contexto da Zika no Brasil
- Realizar o diagnóstico situacional das famílias para análise da efetividade da intervenção
- Testar o piloto da intervenção em dois grupos de pais/mães no Rio de Janeiro e em Salvador
- Avaliar a viabilidade e aceitabilidade da intervenção em 4 grupos de pais/mães no Rio de Janeiro e em Salvador

Avaliação dos Riscos e Benefícios:

Riscos:

Os riscos oferecidos pela pesquisa estarão vinculados à mobilização de narrativas que compõem a memória dos sujeitos entrevistados sobre sua experiência relacionada ao tema.

Benefícios:

Fornecer intervenção precoce para maximizar o progresso no desenvolvimento de crianças afetadas. Isto é importante porque há agora clara evidência de que a intervenção precoce é crucial para maximizar o potencial das crianças com deficiência de desenvolvimento neurológico.

-Apoiar as famílias, tanto em termos de apoio psicossocial, quanto de melhorar sua capacidade para cuidar de suas crianças o melhor possível.

Comentários e Considerações sobre a Pesquisa:

Trata-se de uma pesquisa de extrema relevancia no sentido de investigar se estratégias de intervenção precoce em crianças com SZC é efetiva, aceitável e viável no contexto de 2 centros urbanos brasileiros.

Considerações sobre os Termos de apresentação obrigatória:

Termos de apresentação obrigatória adequados. O CEP-IFF anexou carta da SCTIE e portanto o projeto não necessita ser encaminhado à CONEP.

Recomendações:

TCLEs de acordo com Resolução 466/12.

Conclusões ou Pendências e Lista de Inadequações:

Sem pendências. Liberar parecer aprovado.

O projeto não deve ser encaminhado à CONEP uma vez que apresentou a carta da SCTIE.

Considerações Finais a critério do CEP:

Este parecer foi elaborado baseado nos documentos abaixo relacionados:

Tipo Documento	Arquivo	Postagem	Autor	Situação
Informações Básicas do Projeto	PB_INFORMAÇÕES_BÁSICAS_DO_PROJETO_946246.pdf	23/07/2017 21:57:53		Aceito
TCLE / Termos de Assentimento / Justificativa de Ausência	TCLEparaoutrosparticipantesversao2.doc	23/07/2017 21:51:10	maria elisabeth lopes moreira	Aceito
Projeto Detalhado / Brochura Investigador	TCLEparaoutrosparticpantesversao2.doc	21/07/2017 10:34:12	maria elisabeth lopes moreira	Aceito
TCLE / Termos de Assentimento / Justificativa de Ausência	TCLEparacuidadoresversao2.doc	21/07/2017 10:04:41	maria elisabeth lopes moreira	Aceito
Outros	cartasctie.pdf	29/06/2017 00:10:41	maria elisabeth lopes moreira	Aceito
Outros	autorizacaopesquisa.pdf	29/06/2017 00:10:07	maria elisabeth lopes moreira	Aceito
Folha de Rosto	folhaderosto.pdf	29/06/2017 00:08:08	maria elisabeth lopes moreira	Aceito
Projeto Detalhado / Brochura Investigador	brochura.doc	21/06/2017 13:52:35	Míriam Ribeiro Calheiros de Sá	Aceito
Orçamento	OrCamento.doc	21/06/2017 13:51:12	Míriam Ribeiro Calheiros de Sá	Aceito
Outros	APENDICE4.doc	21/06/2017 13:48:04	Míriam Ribeiro Calheiros de Sá	Aceito
Outros	APENDICE3.doc	21/06/2017 13:47:13	Míriam Ribeiro Calheiros de Sá	Aceito

Outros	APENDICE.doc	21/06/2017 13:45:48	Míriam Ribeiro Calheiros de Sá	Aceito
Outros	apendice1.xlsx	21/06/2017 13:43:26	Míriam Ribeiro Calheiros de Sá	Aceito

Situação do Parecer:
Aprovado

Necessita Apreciação da CONEP:
Não

RIO DE JANEIRO, 24 de Julho de 2017

Assinado por:
Ana Maria Aranha Magalhães Costa
(Coordenador)

ANNEX 2: CONSENT FORMS

CAREGIVERS OF INFANTS AND CHILDREN WITH ZIKA

Participant information sheet: Semi-structured Interviews

You are being invited to take part in a research study. I will read information to you about this study again. Please ask me if there is anything that is not clear or if you would like more information.

What is the purpose of the interview? We want to know to what you have felt about the training you have been received. We will ask you some questions about your whether the training has been relevant for you, whether it has been useful and any recommendations you might have to change this training. This information will be useful to help plan and improve future training for families caring for a baby affected by Zika. An interview helps us get a little more information that the answers from the questionnaire.

What is involved in the interview? We will ask you some questions. The interview will take about 30 minutes. This interview will be recorded, unless you would prefer that a recording is not made.

Which groups are organising the survey? This study is being organised by [insert local research partner] which is a national research organisation. It is supported by the London School of Hygiene and Tropical Medicine, a university based in London. Funding for the study comes from Wellcome, a UK based organisation.

Confidentiality All information which is collected about you/your child(ren) during the course of the research will be kept strictly confidential. The information will be made publicly available on a scientific database on the internet, but this will not include any personal information such as your name, address or other information that would make it possible to identify you.

What are the benefits? The information collected in this survey can help to plan and improve training packages that are available to families caring for a baby affected by Zika.

What are the risks? There are no risks of physical or psychological harm associated with this survey. The questions will take up a bit of your time – about 30 minutes. You will not receive a financial or other type of reimbursement for taking part in the study.

Do I have to take part? No. It is up to you to decide whether or not to take part. If you decide not to take part it will not have an effect on any of the services that you receive. If you/your child(ren) agree to take part you are still free to withdraw at any time and without giving a reason.

How will the information be used? We will analyse the information that you tell use to help better understand how the programme has or has not be useful for you. This information may be used in documents which are written about the programme. We may quote some things that you say in our scientific papers, reports, website or other materials, but this will not include your name or any information that would make it possible to identify you.

If you have any further questions about that are not answered here or have require any further information or explanation please contact:

Local Research Lead:

Rio de Janeiro: [Insert named individual and contact details for local research partner]

Salvador: [Insert named individual and contact details for local research partners]

CONSENT FORM

SEMI-STRUCTURED INTERVIEWS

Participant: (First & Last Name)_______________________________________

1. The information sheet concerning this study has been read to me and I understand what is required of me if I take part in it
2. I have been given the opportunity to ask questions and a reply was given for all the questions to my satisfaction.
3. I understand that participation is voluntary and that I may withdraw at any time without giving a reason
4. I understand that some things that I say during the interview may be used and included documents that are written about the project, but this would be anonymous.

Consent for Adult to Participate

Name	Date	Signature/Thumbprint
Witness	Date	Signature

TRAINING FACILITATORS

Participant information sheet: Semi-Structured Interviews

You are being invited to take part in an interview for a research study. I will read information to you about this study again. Please ask me if there is anything that is not clear or if you would like more information.

What is the purpose of the interview? We want to know more about your views on the training package related to families looking after a child with Zika. We will ask you some questions about how you felt when delivering the training, what went well, what didn't go well and things that you might change about the training.

What is involved in the interview? We will ask you some questions. The interview will take about 30 minutes. This interview will be recorded, unless you would prefer that a recording is not made.

Which groups are organising the survey? This study is being organised by [insert local research partner] which is a national research organisation. It is supported by the London School of Hygiene and Tropical Medicine, a university based in London. Funding for the study comes from Wellcome, a UK based organisation.

Confidentiality All information which is collected about you during the course of the research will be kept strictly confidential. The information will be made publicly available on a scientific database on the internet, but this will not include any personal information such as your name, address or other information that would make it possible to identify you.

What are the benefits? The information collected in this survey can help to plan and improve training packages that are available for people affected by Zika.

What are the risks? There are no risks of physical or psychological harm associated with this survey. The questions will take up a bit of your time – about 30 minutes. You will not receive a financial or other type of reimbursement for taking part in the study.

Do I have to take part? No. It is up to you to decide whether or not to take part. If you decide not to take part it will not have an effect on any of the services that you receive. If you agree to take part you are still free to withdraw at any time and without giving a reason.

How will the information be used? We will analyse the information that you tell use to help better understand how the programme has or has not be useful for you. This information may be used in documents which are written about the programme. We may quote some things that you say in our scientific papers, reports, website or other materials, but this will not include your name or any information that would make it possible to identify you.

If you have any further questions about that are not answered here or have require any further information or explanation please contact:

Local Research Lead:

Rio de Janeiro: [Insert named individual and contact details for local research partner]

Salvador: [Insert named individual and contact details for local research partners]

CONSENT FORM

SEMI-STRUCTURED INTERVIEWS

Participant: (First & Last Name) ____

1. The information sheet concerning this study has been read to me and I understand what is required of me if I take part in it
2. I have been given the opportunity to ask questions and a reply was given for all the questions to my satisfaction.
3. I understand that participation is voluntary and that I may withdraw at any time without giving a reason.
4. I understand that some things that I say during the interview may be used and included documents that are written about the project, but this would be anonymous.

Consent for Adult to Participate

Name	Date	Signature/Thumbprint
Witness	Date	Signature

KEY INFORMANTS

Participant information sheet: Semi-Structured Interviews

You are being invited to take part in an interview for a research study. Please read this information sheet about the study and ask me if there is anything that is not clear or if you would like more information.

What is the purpose of the interview? We want to know more about your views on a training package related to families looking after a child with Zika. We will ask you some questions about your opinions on the training programme in the Brazilian context

What is involved in the interview? We will ask you some questions. The interview will take about 30 minutes. This interview will be recorded, unless you would prefer that a recording is not made.

Which groups are organising the survey? This study is being organised by [insert local research partner] which is a national research organisation. It is supported by the London School of Hygiene and Tropical Medicine, a university based in London. Funding for the study comes from Wellcome, a UK based organisation.

Confidentiality All information which is collected about you during the course of the research will be kept strictly confidential. The information will be made publicly available on a scientific database on the internet, but this will not include any personal information such as your name, address or other information that would make it possible to identify you.

What are the benefits? The information collected in this survey can help to plan and improve training packages that are available for people affected by Zika.

What are the risks? There are no risks of physical or psychological harm associated with this survey. The questions will take up a bit of your time – about 30 minutes. You will not receive a financial or other type of reimbursement for taking part in the study.

Do I have to take part? No. It is up to you to decide whether or not to take part. If you decide not to take part it will not have an effect on your further involvement with the project, if you have any. If you agree to take part you are still free to withdraw at any time and without giving a reason.

How will the information be used? We will analyse the information that you tell use to help better understand how the programme has or has not be useful for you. This information may be used in documents which are written about the programme. We may quote some things that you say in our scientific papers, reports, website or other materials, but this will not include your name or any information that would make it possible to identify you.

If you have any further questions about that are not answered here or have require any further information or explanation please contact:

Project Lead:

Antony Duttine, London School of Hygiene and Tropical Medicine,
antony.duttine@lshtm.ac.uk

CONSENT FORM

SEMI-STRUCTURED INTERVIEWS

Participant: (First & Last Name) ____

1. The information sheet concerning this study has been read to me and I understand what is required of me if I take part in it
2. I have been given the opportunity to ask questions and a reply was given for all the questions to my satisfaction.
3. I understand that participation is voluntary and that I may withdraw at any time without giving a reason.
4. I understand that some things that I say during the interview may be used and included documents that are written about the project, but this would be anonymous.

Consent for Adult to Participate

Name	Date	Signature/Thumbprint
Witness	Date	Signature

Participant information sheet: questionnaires

You are being invited to take part in a questionnaire for research study. I will read information to you about this study again. Please ask me if there is anything that is not clear or if you would like more information.

What is the purpose of the questionnaire? We want to understand certain aspects of your day to day life and how they have changed over the course of the programme. Know to what you have felt about the training you have been received. We also want to find out whether you have found the programme useful and if it something that you feel might be needed for others. This information will be useful to help us understand whether the training is useful, needed and improve future training for families caring for a baby affected by Zika.

What is involved in the questionnaire? We will give you a questionnaire to complete at the beginning and end of the training. The questionnaires should take about 30 minutes to complete. Most of the questions at the beginning and end are the same, but some may appear in one and not the other.

Which groups are organising the survey? This study is being organised by [insert local research partner] which is a national research organisation. It is supported by the London School of Hygiene and Tropical Medicine, a university based in London. Funding for the study comes from Wellcome, a UK based organisation.

Confidentiality All information which is collected about you/your child(ren) during the course of the research will be kept strictly confidential. The information will be made publicly available on a scientific database on the internet, but this will not include any personal information such as your name, address or other information that would make it possible to identify you.

What are the benefits? The information collected in this questionnaire can help to plan and improve training packages that are available to families caring for a baby affected by Zika.

What are the risks? There are no risks of physical or psychological harm associated with this survey. The questions will take up a bit of your time – about 30 minutes. You will not receive a financial or other type of reimbursement for taking part in the study.

Do I have to take part? No. It is up to you to decide whether or not to take part. If you decide not to take part it will not have an effect on any of the services that you receive. If you/your child(ren) agree to take part you are still free to withdraw at any time and without giving a reason.

If you have any further questions about that are not answered here or have require any further information or explanation please contact:

Local Research Lead:

Rio de Janeiro: [Insert named individual and contact details for local research partner]

Salvador: [Insert named individual and contact details for local research partners]

CONSENT FORM

QUESTIONNAIRE

Participant: (First & Last Name) ______________________________________

1. The information sheet concerning this study has been read to me and I understand what is required of me if I take part in it
2. I have been given the opportunity to ask questions and a reply was given for all the questions to my satisfaction.
3. I understand that participation is voluntary and that I may withdraw at any time without giving a reason

---

Consent for Adult to Participate

Name	Date	Signature/Thumbprint
Witness	Date	Signature

ANNEX 3: INTERVIEW SCHEDULES

Endline Qualitative:
Zika programme participant interviews

Materials & preparation:

- Preparation: print our interviews and summarise key issues to further explore for each individual family based on the last visit.
- Copy of the 'ladder'

Overall aims:

To explore if here have been any changes over the course of the programme

To find out what the families felt about the programme

Present the information sheet and consent form.

Introductions and outlining purpose of today's interview

General health and well-being of the child

How has (insert child's name) been since the programme started?

Has anything changed? Explore in concrete terms e.g. Functional changes, what brought that about, change in general health of the child, child feeling happier, child able to go to school

For each change – <u>explore further what the caregiver thinks has brought about that change</u>

> **Individual factors** e.g., change in knowledge/understanding, a change in practice- what was it?
>
> **Assistive devices e.g.**, new chair, different equipment for feeding etc- can I see – how has this helped
>
> **Family level changes**: more support in the family, change in attitudes of family- more help in the house?
>
> **Wider changes: Referrals** and links to other services e.g. access to nutritional support/schools. What did that support look like – can you take me through the last time e.g. last visit from the community nutritionist.
>
> **Other?**
>
>> **Livelihoods –** last time I saw you explained that you were not able to pay for X ……….you were not able to work- has this changed? In what way?

How do you feel about these changes?

Insert specific follow up probes for each family which will vary:

Use of the assistive device- can I see the assistive device-what has it allowed the child to do?

Support around nutrition- what did that mean – can they take me through the last visit to the nutritionist, changes in food, feeding practice

Most Significant Change. Looking over the changes, which do you think is the *Most Significant Change* for your child? Why? **For You what has been the most important change**

Changes for you as the caregiver

We have been talking a lot about X, now I want to ask you how you are?

Has life changed for you over the course of the programme? If so, in what way?

Prompts around changes Individual: changes in how you understand things, Family: changes in support, wide support group or community support)

Prompts – now able to work more/less, do household chores, less exhausted? emotionally – understand more, more worried/less worried? Communication – with other families members, get more help from other family members)

- To get a more concrete example – ask about something that has happened to the child recently – such as taking to a family event – how did they feel about that? When the child started to feed better/attend school?

Support network? Who do you go to support? So if you have a problem now with child X (for example refer back to an example from the ladder), who would you go and discuss this with?

In your role as caregiver, what is the **Most Significant Change** for you over the last year?

Wider learning in the family/community

(Exploring here how is the learning in the parent groups being translated into changes at home and engagement with other family members. If this has been covered in part 2, you do not need to repeat questions, but try to probe about how the family AND community may have changed)

- Do you feel there is any change in how other family members view your child's condition? Why? Why not? What does that mean in practice? Who looks after the child when you need to work? How do they know this – perhaps a concrete example
- Greater understanding in the community? In what way? (huge levels of stigma, beliefs that child a spirit child- what can you say to people? Have you shared any of this information with other neighbours etc.) How did this come about? Do you think it's really making any difference? (group activities, role of the facilitator)

The role of the support group

What did the group represent in your life?
How did you know about the group?
How did you find the group?

Good things? Any sessions you have particularly liked? Didn't like?

Can you tell if there is anything that you value about the group? Probe around not just learning new things and general comments but on the value of being a member with other caregivers. Try to ask if the found the group appropriate/acceptable in terms of venue, mix of participants, content of sessions etc.

- Prompts: Meeting other mums, Being able to share your problems?
- Do you find other mums share their experiences? In what way? Is it just focussing on their child?
- Learning from others?
- Language?
- How easy/difficult did you find putting what you learned in the sessions into action at home?

Did you manage to come to each session? If you missed a session, why was that? Did you find it difficult to carry on after missing a session?

Joint group activities: Is your group planning to organise something after this week? How has that come about? How do you feel about that? How will you keep in touch?

If you were to say ONE thing that was of most importance to you from the support group, what would it be?

If the family was not able to attend regularly? And if not, why not (will have the attendance data so probe on families who missed sessions

Now that the group has stopped are you in touch with anyone? Plans to stay in touch? If not, why not?

The role of the home visit (if applicable)

Reflect back on the time we came to visit you at home.

What are your views on the home visits – how have they worked? Do they help? In what way? Are other family members involved at all, and if so, in what way? Has that made any difference in terms of caring for child X?

Need of the programme

Do you feel that the programme would be useful for other families in similar situations to you? Have you met other parents who might make use of the programme?

Other

Thank you for your time and for sharing your experience of the training with us. As we think about conducting this training and home visits with other parents, is there anything else important which I haven't asked you which you want to share with us? Other recommendations?

Endline Qualitative:
Zika programme facilitator interviews

Materials & preparation:

- Preparation: Print out this interview guide

Overall aims:

To explore how the programme ran

To explore how the programme may integrate into existing services

Present the information sheet and consent form.

Introductions and outlining purpose of today's interview

Reflection on the programme

Overall, how do you feel the programme has gone? What areas did you feel worked the best? What areas do you feel worked the least?

Why do you feel these areas worked well/not so well

Participant factors e.g., content not relevant to needs of group

Facilitator factors e.g. did not feel confident in a particular subject.

Organisation factors e.g., venue location, facilities

Other?

How did you feel that the training course prepared you for being a facilitator?

What would you do differently if you were to run the programme again?

How did you feel about the other facilitator? Did the dynamic of having one parent/one health worker work? What was good? What was not good?

How has the programme impacted on you personally? (if a therapist) how has it made you consider and reflect on your practice? (if a mother) how has it affected you – have there been any changes? Did you find it easy, difficult? Are you happy to have been part of the programme?

Content of the programme

Which were the best sessions of the programme? What made them so good?

Are there subjects that you feel were missing in the training programme? Were there areas that you feel were not needed in the training programme? Are there sessions that need to be improved?

Impact of the programme

How do you feel the caregivers responded to the programme? Did you notice any changes in any of the participants? What were they?

How did you find the group dynamic? Good things? Prompts: Meeting other mums, being able to share your problems?

Do you think that the group will stay in touch now that the programme has finished?

Did you manage to come to each session? If you missed a session, why was that? Did you find it difficult to carry on after missing a session?

Joint group activities: Is your group planning to organise something after this week? How has that come about? How do you feel about that? How will you keep in touch?

The role of the home visit (if applicable)

Reflect back on the home visit

What are your views on the home visits – were they easy to arrange? Did you find it useful or not?

Need of the programme

Do you feel that the programme would be useful for other families in similar situations? Have you met other parents who might make use of the programme? Which type of children should be the target of the intervention?

Integration and expansion

DO you think there is a need for this programme to expand across other parts of Brazil? If so who do you think should be the organisers of such a programme? **Prompts** ask about health clinics, hospitals or parent groups, community centres

Other

Thank you for your time and for sharing your experience of the training with us. As we think about conducting this training and home visits with other parents, is there anything else important which I haven't asked you which you want to share with us? Other recommendations?

Endline Qualitative:
Zika programme key informant interviews

Materials & preparation:

- Preparation: Print out this interview guide, provide a copy of the project overview

Overall aims: To ask key informants about need, integration and expansion of a programme

Present the information sheet and consent form.

Introductions and outlining purpose of today's interview

Need/demand of the programme

Based on what you have seen in your role, do you feel a programme such as this is needed in Brazil? Why?

Integration of the programme

Having looked at the programme, to what extent do you feel that this could be integrated into the work of your organisation? How would this best be undertaken?

Expansion of the programme (if applicable)

To what degree do you feel a programme like this could be expanded to other areas of the country? How would this be done?

Other

Thank you for your time and for sharing your experience with us. As we think about conducting this programme with other parents, is there anything else important which I haven't asked you which you want to share with us? Other recommendations?

ANNEX 4: FIFTH RESEARCH PAPER (published)

Reflections on Health Promotion and Disability in Low and Middle-Income Countries: Case Study of Parent-Support Programmes for Children with Congenital Zika Syndrome

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RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	International Journal of Environmental Research and Public Health		
When was the work published?	14 March 2018		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion			
Have you retained the copyright for the work?*	**No**	Was the work subject to academic peer review?	**Yes**

*If yes, please attach evidence of retention. If no, or if the work is being included in its published format, please attach evidence of permission from the copyright holder (publisher or other author) to include this work.

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SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	
Please list the paper's authors in the intended authorship order:	

Stage of publication	Choose an item.

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I was a co-author of this paper, providing input into the structure and design and feedback and edits on the various versions before submission and after reviewer feedback.

SECTION E

Student Signature	
Date	19th September 2022

Supervisor Signature	
Date	20 September 2022

International Journal of
*Environmental Research
and Public Health*

MDPI

Article

Reflections on Health Promotion and Disability in Low and Middle-Income Countries: Case Study of Parent-Support Programmes for Children with Congenital Zika Syndrome

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Abstract: Universal health coverage (UHC) has been adopted by many countries as a national target for 2030. People with disabilities need to be included within efforts towards UHC, as they are a large group making up 15% of the world's population and are more vulnerable to poor health. UHC focuses both on covering the whole population as well as providing all the services needed and must include an emphasis on health promotion, as well as disease treatment and cure. Health promotion often focusses on tackling individual behaviours, such as encouraging exercise or good nutrition. However, these activities are insufficient to improve health without additional efforts to address poverty and inequality, which are the underlying drivers of poor health. In this article, we identify common challenges, opportunities and examples for health promotion for people with disabilities, looking at both individual behaviour change as well as addressing the drivers of poor health. We present a case study of a carer support programme for parents of children with Congenital Zika Syndrome in Brazil as an example of a holistic programme for health promotion. This programme operates both through improving skills of caregivers to address the health needs of their child and tackling poverty and exclusion.

Keywords: disability; health promotion; Zika; parent-support; low and middle income

1. Introduction

People with disabilities include those who have long-term physical, mental, intellectual or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others (Table 1). The WHO estimates that there are one billion people living with disabilities globally, of whom 80% live in low and middle-income countries (LMICs) [1]. People with disabilities are more likely to be poor [2] and also face a broad range of exclusions, including from school, employment and social engagement [1]. These exclusions are inter-linked and reinforcing, for instance, a lack of money can reduce inclusion in school and lack of education can make it more difficult to find a job, which leads to deepening poverty. Disability has therefore increasingly become a focus in international development, because the number of people with disabilities is large and they are being left behind while progress is made for other groups. An example is the specific reference to disability within the Sustainable Development Goals (SDGs) adopted internationally in 2015, including with respect to Goals on "Quality education" and "Decent work" [3].

2. Health, Universal Health Coverage and Disability

Disability is not mentioned within the SDG for "Good Health and Well-Being", which includes the target to "Achieve Universal Health Coverage" (UHC). UHC focuses on covering the whole population with the health services they need, without suffering financial hardship. These health services cover the full spectrum from promotion, treatment and rehabilitation to palliative care. There tis a clear rationale for focusing on people with disabilities in this Goal and in efforts towards UHC, as there is growing evidence that people with disabilities are more at risk of experiencing poor health (e.g., non-communicable diseases, hypertension and mental health conditions) [4–13] and to face great barriers and higher costs in accessing healthcare [1]. Consideration therefore needs to be given as to how to include the large number of people with disabilities within ambitions to achieve UHC.

Considering the relationship between health and disability is complex as these constructs are overlapping, inter-twined and reinforcing (Figure 1). Furthermore, people with disabilities are a highly diverse group, including people with a range of impairment types, age, gender and environments. The relationship between health and disability will therefore not be the same for all.

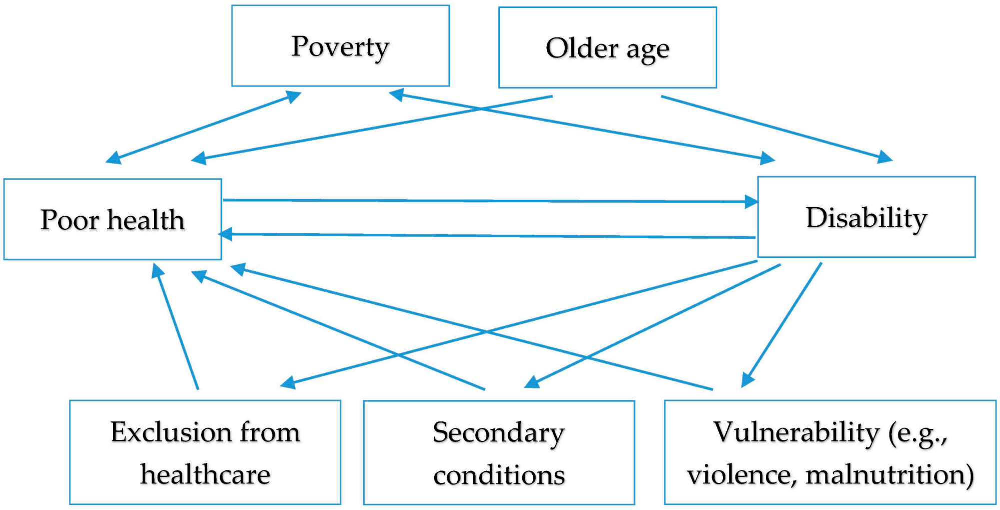

Figure 1. Complex relationship between disability and poor health.

On the one side, poor health may lead to disability. People with disabilities, by definition, have an impairment as a result of an underlying health condition (Table 1) [1,14]. Disability is not the inevitable consequence of poor health or impairment but occurs in the context of unfavourable personal or environmental factors that hinder the person's full and effective participation in society on an equal basis with others. A health condition or impairment is therefore a necessary condition for disability, but is not a sufficient cause on its own.

Table 1. Definitions of Disability.

UN Convention on the Rights of Persons with Disabilities	Persons with disabilities include those who have long-term physical, mental, intellectual or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others [14].
World Report on Disability	[Disability is] an umbrella term for impairments, activity limitations and participation restrictions, denoting the negative aspects of the interaction between an individual (with a health condition) and that individual's contextual factors (environmental and personal factors) [1].

On the other side, disability may contribute to worsening health. The underlying health condition causing the disability may have other negative impacts, for instance diabetes may cause visual impairment but also kidney or nerve damage. The impairment may itself be associated with an increase in risk of poor health outcomes, for instance people with spinal cord injuries are at risk of developing pressure sores, or people with limited mobility are prone to osteoporosis. People with disabilities are also on average older [1] and therefore more likely to experience multiple health conditions at the same time. The unfavourable structural condition of people with disabilities, in terms of their poverty and exclusion, will also make them more vulnerable to ill health and injuries [15,16]. For instance, poverty is associated with malnutrition, inadequate access to public health services (e.g., immunisation), poor living conditions (e.g., lack of safe water) and environmental exposures (e.g., unsafe work environments) and so these issues will disproportionately affect people with disabilities [16]. People with disabilities may also experience barriers to accessing health care services, or inadequate quality of health care, which also potentially results in poorer health [1]. These links are not the same for all people with disabilities. For instance, people with intellectual impairments may face more exclusions from health promotion activities, while people with physical impairments may be particularly vulnerable to difficulties caused by physical accessibility.

A clear consequence of the vulnerability of people with disabilities to poor health is the need to ensure their good access to healthcare, and without their inclusion UHC is unlikely to be achieved. Access to health is also the fundamental right of people with disabilities, as set out in the UN Convention on the Rights of Persons with Disabilities [14]. Access should be to the full spectrum of services, addressing both general healthcare needs (e.g., sexual and reproductive health services) as well as impairment-focussed care (e.g., rehabilitation services). These services are needed to treat conditions as they arise, to prevent further morbidity and reduce mortality. Provision of curative and rehabilitation services alone is insufficient. Health promotion is also a priority to maintain health and avoid long-term health effects associated with disability. Health promotion should tackle both general and specialist health issues—acknowledging that people with disabilities require the same health promotion as people without disabilities (e.g., sexual health [17–19], oral health [20]) but may also require other specific messaging (e.g., prevention of secondary complications such as contractures) [3].

3. Health Promotion for People with Disabilities

Health promotion is defined by WHO as "*the process of enabling people to increase control over and to improve, their health*". At the individual level, health promotion aims to promote health and healthy lifestyles through personal behaviour change. These interventions include promoting good nutrition, undertaking regular physical activity and engaging in vaccine and preventative health initiatives (e.g., vitamin A supplementation). There is strong evidence that people with disabilities may fall behind in terms of individual behaviours related to a healthy lifestyle, for instance, they are on average more likely to be physically inactive [6,10,21], smoke [6,10] and use illegal drugs [22,23]. Low engagement in preventative health behaviours is also of concern among people with disabilities [11,24–26]. As examples, people with disabilities are less likely to attend regular visits to the dentist [27] or take part in cancer screening [28,29], or be reached by messaging about HIV [30,31].

There is therefore an urgent need to focus on people with disabilities in health promotion activities, because they are at higher risk of poor health and are falling behind in individual healthy behaviours. This focus is currently not happening well for a variety of reasons. Information may not be transmitted in accessible formats, such as braille, sign-language or easy read. There may be misconceptions that people with disabilities do not need certain services, such as information about sexual health and they are therefore not targeted with these messages. People with disabilities may also be excluded from the health promotion campaigns for other reasons. For instance, children with disabilities, who are less likely to attend school, may not be reached with school-based health promotion activities. Pragmatic evidenced-based solutions are needed to overcome these barriers and better meet the needs of people with disabilities in health promotion [32,33].

A twin-track approach has been long recognised and advocated for within the disability community. This approach aims to ensure that people with disabilities are included in mainstream programming but also have specific targeted interventions to meet any additional needs (Examples given in Table 2). If we consider this approach with respect to health promotion for people with disabilities, it means first ensuring that they are included within mainstream health promotion activities (e.g., including examples of people with disabilities in health promotion material, providing material in braille, ensuring meetings are held at accessible locations) but also targeting people with disabilities with specific interventions around health promotion (e.g., providing people with mobility impairments information about how to prevent bed sores) [34–39]. People with disabilities have unique insights about their disability and situation, but are often excluded from the decision-making process about issues that directly affect their lives, ref. [40] and this must be addressed in planning health promotion activities, whether mainstream or targeted.

Table 2. Examples of the twin-track approach.

Issue	Mainstream Intervention	Targeted Intervention
Children with disabilities are excluded from school	Ensure that policies reinforce the rights of children with disabilities to education	Provide financial subsidies to children with disabilities to facilitate their school attendance
People with disabilities are more likely to be poor	Ensure that people with disabilities are eligible for social protection programmes	Offer vocational training for people with disabilities
Women with disabilities receive inadequate sexual health services	Train doctors on the needs of women with disabilities for these services	Provide information to women with disabilities about their right to healthcare and how they can realise these rights

Health promotion also has a broader aim, however, beyond changing individual behaviours. This broader aim is to tackle the determinants of health, for instance through improving income, housing, food security, employment and quality working conditions. We have already described how people with disabilities are more likely to experience structural inequalities, such as exclusion from jobs and schooling and higher levels of poverty [1,2] and these factors will make them vulnerable to poor health. The broader ambition of health promotion is therefore to improve the inclusion and living conditions of people with disabilities in order to promote and preserve their health. This more holistic and far-reaching approach to health promotion for people with disabilities therefore has the same ambitions as the SDGs including: less poverty, better living standards and equitable inclusion in employment and education [3]. Again, a twin-track strategy can be used to address these inequalities, ensuring both that people with disabilities are included in mainstream programmes and that they are additionally targeted with specific interventions.

Targeting the drivers of poor health can be challenging, though ultimately is likely to have the biggest impact. As an example, people with disabilities often have difficulties in accessing Water, Sanitation and Hygiene (WASH) services [41], which will make them vulnerable to poor health. Providing accessible facilities alone will likely reap some benefits towards overcoming these barriers. However, making efforts to address stigma and discrimination and improving the policy framework around disability will likely improve not only access to WASH but will also have more wide-reaching impacts for people with disabilities, such as encouraging inclusion in school, jobs and society at large [41,42].

The focus of health promotion therefore needs to be expanded, if UHC is to be achieved. First, it needs to be inclusive of people with disabilities as they are a large and vulnerable group. Second, it needs to tackle the underlying drivers of poor health as well as immediate behaviours. Health promotion policies and activities should therefore address deeper causes of poor health, as well as health behaviours.

4. A Case Study on Health Promotion and Disability: Children with Congenital Zika Syndrome

We can make this discussion on health promotion and disability more concrete by considering the example of Congenital Zika Syndrome. Following the Zika Virus epidemic in South America in 2015, there was a massive increase in the number of babies born with microcephaly and other abnormalities, now collectively called "Congenital Zika Syndrome". These children experience a range of health conditions, including severe developmental delay, intellectual and visual impairment and musculoskeletal abnormalities and epilepsy [43]. The affected children are also vulnerable to a range of secondary health conditions arising from the syndrome, such as respiratory illness, malnutrition and pressure sores. These health concerns will persist as the children transition into adulthood. Children with Congenital Zika Syndrome will therefore have a lifetime greater vulnerability to poor health and an increased need for a range of healthcare services as a result of their physical and intellectual impairments.

Promoting the health of these children is imperative. Health promotion messaging is mostly directed at parents, as they are usually the main carers for children with Congenital Zika Syndrome. Health promotion efforts focus on improving knowledge, skills and behaviours of parents so that they can (1) better address their child's existing health issues (e.g., control of seizures) to prevent them causing further concerns; (2) avoid the occurrence of health problems in the future (e.g., malnutrition through better feeding, contractures through improved positioning) and (3) maximize the development of their child (e.g., through early stimulation). These efforts aim to preserve good health as far as possible, maximise development and quality of life and reduce the need of these children for healthcare services.

There is an important concern with this approach. Looking after a child with severe developmental disabilities and complex needs places an enormous emotional strain on families, with resultant high risks of paternal abandonment and maternal mental health concerns. Families of children with Congenital Zika Syndrome are disproportionately more likely to be poor and also often experience further financial strain through medical costs and lost income. Parents are therefore being expected to take on caring tasks for their child with complex needs, with little training or support, while they are also experiencing emotional distress, poverty and disadvantage. Developing interventions to improve the knowledge and skills of parents to change behaviours are unlikely to be effective and sustainable long-term without addressing these other difficulties that the parents may face. As a consequence, health promotion efforts need to go beyond improving the knowledge and behaviour of parents to also addressing other underlying determinants of poor health for these children: poverty, exclusion and carer distress.

One potentially effective strategy to achieve more holistic and sustainable health promotion is through caregiver support programmes [44]. We used this approach to develop a carer-support intervention targeting parents of children with Congenital Zika Syndrome in Brazil. This programme aims to support the families, both in terms of providing psychosocial support and in improving their skills, to be able to care for their child optimally and to connect to available services. The programme also focusses on the underlying drivers of poor health: poverty, stigma and exclusion. Ultimately, this programme aims to promote the health and functional status of the child and eventually the child's (and the parents') participation in society. Using the twin-track framework described above, this is an example of a targeted intervention and complements mainstream programmes that are inclusive of children with disabilities.

The health promotion intervention is offered through groups of parents and their children with Congenital Zika Syndrome. It consists of 10–11 sessions offered over a period of 3 months. The content of the programme includes information about essential care practices, such as feeding, positioning, communication, play and early stimulation, in order to promote health and maximise development. This focus takes learning from some of the health concerns that often affect children with cerebral palsy (a similar condition to Congenital Zika Syndrome) such as contractures and malnutrition and aims to preserve health and avoid the occurrence of these secondary health conditions. It also encourages effective health seeking behaviour and the sharing of experiences between parents as to how this can be achieved. For instance, in the food and nutrition module, there is a focus on what constitutes a balanced diet—important for all children. This module also covers positioning for feeding for children with disabilities and looking out for signs of aspiration—a potential risk for some children with developmental delays, which can lead to severe respiratory problems.

The programme goes beyond a focus on changing individual behaviours and promoting the child's physical condition and health; It also addresses disability rights and how parents can advocate for their child's inclusion in school and health care and receipt of disability benefits. As an example, navigating the health care system is an important concern raised by parents. A session in the programme helps parents to understand how they can get the most out of health visits, which may encourage them to attend appointments regularly and engage more effectively in their child's care plan. The ambition is that the parent groups will become self-sustaining and a focus in the latter part of the programme is on how to run parent groups and engage effectively with the community to overcome stigma and discrimination and promote inclusion and acceptance. The emotional support activity, as part of every session, has helped to stimulate open and supportive discussion between parents about their successes and difficulties. This component fosters an atmosphere of empathy and solidarity, contributing towards mental health promotion for carers and their improved capacity to look after their child. The programme therefore also aims to empower parents so that they can address the drivers of poor health among their children with disabilities, which include stigma and discrimination, exclusion from health care services and poverty.

The parent groups are led by two facilitators—one therapist and one parent of child with Congenital Zika Syndrome ("expert mother"). The role of the expert mother is crucial in order to facilitate a participatory and egalitarian atmosphere and to encourage sharing of learning between parents. The facilitators attended a one-week training course before the start of the programme and are given on-going support through a set of materials, regular supervisory visits and access to mentors. Groups are held in the local community (e.g., local health centre or church) so that strong networks can be built between carers who live close to each other. The sessions are participatory and include activities, open discussions, explanations, demonstrations and light-hearted ice breakers (Table 3). This approach is informed by adult learning theory and aims to minimise issuing further instructions to over-loaded parents [45].

Table 3. Example of part of a facilitated group session on eating and drinking.

Example	Discussion	Aim
Ice-breaker In pairs: One person tries to give the other a drink of water in different positions (e.g., head leaning back, turned to one side, or flopping forwards).	*How easy or difficult is it to swallow in each position? How does it feel to be fed?*	To understand a range of issues that your child may experience with eating and drinking.
Discussion As a large group to share experiences	*What is a nutritious or "balanced" diet?*	To know what a balanced diet is and how to maximise your child's nutritional intake and prevent malnutrition.
Activity Show a banana and a biscuit and other common foods.	*Discuss—Are the items hard or soft? Can they be made into a smooth puree? How?*	To learn ways to feed your child safely

The intervention is currently undergoing pilot testing with 6 parent groups spread across Rio de Janeiro and Salvador to assess the feasibility and acceptability of the intervention. The next stage is to evaluate the effectiveness of the programme in improving the health (e.g., nutritional status, utilization of health care services) and functional status of children (e.g., measured using Bayley Scales of Infant Development) and the mental health (e.g., using PHQ-9) and quality of life of parents (e.g., using WHO Quality of Life scale), ideally through a randomized controlled trial.

5. Conclusions

The focus of health promotion needs to be expanded if UHC is to be achieved. First, health promotion activities must be inclusive of people with disabilities as they are a large group who are more vulnerable to poor health. In addition, health promotion must also address poverty and inequality as key drivers of poor health, since tackling individual behaviours alone is insufficient. Achieving inclusion of people with disabilities in health promotion will be helped if this is supported by appropriate policies and if evidence is generated as to how inclusion can be achieved. One potential approach is through parent support programmes which offer innovative and sustainable ways to improve the skills and knowledge of parents and thereby the health of their children with disabilities and the drivers of poor health (e.g., poverty, stigma) as part of a wider strategy. Including people with disabilities in health promotion will ultimately help to make sure that we Leave No-one Behind as we move towards Universal Health Coverage.

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ANNEX 5: SIXTH RESEARCH PAPER (published)

Engagement of Fathers in Parent Group Interventions for Children with Congenital Zika Syndrome: A Qualitative Study

RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

SECTION B – Paper already published

Where was the work published?	International Journal of Environmental Research and Public Health		
When was the work published?	12 October 2019		
If the work was published prior to registration for your research degree, give a brief rationale for its inclusion			
Have you retained the copyright for the work?*	**No**	Was the work subject to academic peer review?	**Yes**

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SECTION C – Prepared for publication, but not yet published

Where is the work intended to be published?	
Please list the paper's authors in the intended authorship order:	

Stage of publication	Choose an item.

SECTION D – Multi-authored work

For multi-authored work, give full details of your role in the research included in the paper and in the preparation of the paper. (Attach a further sheet if necessary)	I was a co-author of this paper, providing input into the structure and design and feedback and edits on the various versions before submission and after reviewer feedback.

SECTION E

Student Signature	
Date	19th September 2022

Supervisor Signature	
Date	20 September 2022

International Journal of
Environmental Research and Public Health

Article

Engagement of Fathers in Parent Group Interventions for Children with Congenital Zika Syndrome: A Qualitative Study

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Abstract: We aimed to explore the engagement of fathers in a community-based group intervention (Juntos) for children with congenital Zika syndrome (CZS) and their caregivers in Brazil. Six Juntos groups were facilitated from August 2017 to May 2018. We conducted a qualitative study to evaluate the feasibility and acceptability of the intervention for fathers of children with CZS. Methods included participant observation, focus group discussions, and semi-structured interviews of fathers with a child enrolled in the program. Data collected were transcribed, coded and thematically analyzed to explore father preference for, and beliefs about the intervention and to assess potential barriers and enablers to their involvement. Forty-nine families (61 participants) enrolled, of whom 20% (12/61) were fathers. Seven (58%) fathers attended more than 7 out of 10 sessions. The content of Juntos was found to be acceptable to those fathers who attended. Participation in the group offered fathers the opportunity to share experiences of caring for their child and demonstrate their importance as care agents. Work commitments, and the view of mothers as primary caregivers were barriers to engagement of fathers. Facilitators to engagement included a presentation of clear objectives for fathers' involvement and the opportunity to learn a practical skill related to caring for their child. A better understanding of the perspectives of fathers is crucial to help increase their involvement in parenting interventions.

Keywords: Zika; disability; parent support; low and middle income; father engagement

1. Introduction

There are an estimated 95 million children with disabilities, constituting one in 20 children globally (5.1%) [1]. Caregivers of children with disabilities experience pressure to meet the emotional and physical needs of their child, while at the same time maintaining family functioning [2]. Consequently, families of children with disabilities report social isolation, stigma, mental health challenges and increased financial and emotional strain [3–5]. The risk of fathers abandoning their child with disabilities is high [6], making families even more vulnerable.

Child development programs focus mostly on the role of the mother, as women are traditionally socially constructed as primary carers, and this minimizes the caring role of fathers in co-parenting [7]. Yet there is strong evidence that well-designed health interventions that include fathers positively impact on child development, wellbeing, and family functioning [8–10], particularly since social support has emerged as an important factor in influencing both parent and child functioning [11].

Despite this evidence, engaging with fathers is one of the least well-explored and articulated aspects of parenting interventions [12], and attendance at parent groups is much lower for men than for women [13]. Fathers are often recruited via women [14], and evaluation studies have been conducted mostly with mothers [15,16]. Insufficient attention is, therefore, given to reporting fathers' participation and impact on child or family outcomes. There is little robust evidence as to "what works" [14,17] to engage fathers with parenting interventions, mostly restricted to families of children without disabilities [18]. Group interventions that successfully integrate fathers of children with developmental disabilities are rare.

One domain in which the engagement of fathers is relevant, but has not yet been explored is the care of children with Congenital Zika Syndrome (CZS). More than three thousand children have been born with CZS since the start of the Zika epidemic in Brazil, and children with CZS present with a range of neurological conditions and sensory and musculoskeletal impairments [19,20], secondary to central and perhaps peripheral nervous system damage. In describing CZS, Moore et al. [21] suggest five unique features:

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

Nevertheless, a wider spectrum of developmental impairments may yet manifest in children who were exposed to Zika in utero as they continue their development [22].

Children with CZS are provided formal medical support services, which in Brazil are primarily delivered through tertiary care centers. However, innovative ways to provide support and education to families of children with CZS are required, given the wide-ranging and complex needs of children with CZS, and consequent strain on their families [23–26]. One such intervention is 'Juntos' ('together' in Portuguese), which is a facilitated participatory group intervention for caregivers of children with CZS that runs over 10 sessions [4,6]. Each Juntos group is facilitated by one 'expert carer', a mother of a child with CZS, and one allied health professional (physiotherapist, occupational therapist or speech therapist). Each session includes ice-breaker activities, practical sessions and group discussions, and lasts approximately 4 h. Topics covered include positioning and moving, eating and drinking, communication, play and early stimulation, everyday activities, community inclusion and disability rights (Table 1). An emotional support activity, as part of every session, is included to stimulate open and supportive discussion between caregivers about their successes and difficulties. The intervention aims to improve the quality of life of caregivers and children with CZS [27]. Program material is available from www.ubuntu-hub.org.

Within the context of the Juntos intervention we aimed to (i) explore fathers' views about the program, and (ii) assess potential barriers and facilitators to fathers' involvement in Juntos to support care for their child with CZS.

Table 1. Juntos intervention module topics.

Module Number and Title	Topics Covered
1: Introduction	About the program Information about Zika and Congenital Zika syndrome How to find information Personal stories
2: Our child	Introducing your close family and friends Development milestones for young children Determining your child's progress Managing irritability and crying
3: Positioning and moving	How to position children who need assistance How to assist children to learn to move
4: Eating and drinking	Feeding challenges Practical skills to address challenges for your child
5: Communication	Importance of communication Practical advice to help your child communicate
6: Play and early stimulation	Importance of play for children to develop and learn Early stimulation Making simple toys Inclusion of play in the family and the broader community
7: Everyday activities	How to use everyday activities to help your child develop Managing seizures
8: Uniting our voices	Understand the context of disability rights Education Communicating with your health team Advocating
9: Our community	Who is in your community Common barriers to inclusion Addressing negative attitudes and exclusion Social Activity
10: Next steps	Summing up Planning next steps for yourself and the group

2. Materials and Methods

Ethical approval was gained from Instituto Fernandes Figueira—IFF/FIOCRUZ—RJ/MS 2.183.547 and LSHTM Ethics Ref: 13608. Informed consent was acquired from all participants. The study protocol was published in May 2019 [28].

Participants of the Juntos intervention were caregivers (mothers, fathers, grandparents or aunts) of children with neurologist-confirmed CZS in Rio de Janeiro and Salvador (Bahia), identified through clinical and therapy networks at the two sites. For the purposes of this study, we used CZS to describe any child with impairments that can be directly attributed to Zika. The impairments in children included either mild cognitive, communication or functional skill delay and ranged to severe delay in all three developmental categories. Identified participants were contacted by site coordinators about joining the groups, and a total of 49 children were recruited with a broad range of impairments. Six Juntos groups, each of 10 sessions, were run in Rio de Janeiro and Salvador (August 2017–May 2018) to pilot test and assess the feasibility of the intervention. All participants attending the intervention had been invited to participate in the feasibility study by the study site coordinator.

We conducted a qualitative study of the Juntos intervention between August 2017 and June 2018. The reasons for doing the research were explained, and data were collected by four female Brazil-based research assistants (psychologists). None of the psychologists worked in the clinical area or had prior knowledge of the participants. The number of program sessions attended between baseline and program completion were recorded for each participant, and three techniques were used for collecting qualitative data. First, participant observation by a psychologist was undertaken during the delivery of all the sessions (n = 10 sessions) of the Juntos intervention in the six groups. An observation checklist was used to assess the fidelity of delivery of the intervention by the facilitators. In addition, observation of the ice-breaker activities, practical sessions and group discussions provided an opportunity to examine the interaction of the fathers with their child and their partners during each session. The psychologists observing the sessions wrote detailed field-notes on individual men's engagement. Second, the psychologists (one per site) facilitated focus group discussions related to the content of the session at the end of each session. Focus group discussions were approximately forty-five minutes in duration. The psychologists took comprehensive notes of the discussion with the participants. For conversations which took place with facilitators outside of the sessions, pertinent comments were recorded in an excel file. Third, one-to-one semi-structured interviews were undertaken by the psychologists with caregivers post-intervention. All participants who had attended the Juntos groups were eligible for selection, and then a subsample of caregivers (n = 13) out of those who expressed an interest in being interviewed were purposively selected to include male and female caregivers and participants of different ages to provide a diverse sample. The 13 participants had completed between 7 and 10 sessions. The interview guide was adapted from a previous study [6], and piloted for understanding. Interviews were designed to explore participants' motivation for attending the intervention, their views around how they were approached to participate in 'Juntos', how they felt about the program itself, and aspects around engagement were explored. The interview guide is provided as Supplementary 1. Interviews lasted on average 1 h, ranging between 50 min and 1 h 20 min and were recorded through audio techniques.

Audio-recorded data were transcribed verbatim and translated into English. The responses to the direct observation data and open-ended questions were collated into a word document by one lead UK based researcher, a specialized pediatric physiotherapist trained in qualitative and quantitative techniques (TS). The transcribed text was analyzed using an inductive thematic analysis [29]. This included three steps. First, two psychologists (A.C.D.V. and B.S.M.) who collected and transcribed the data, and who were, therefore, familiar with the content, read the text several times to form an impression of the overall content. Second, words and phrases that described fathers' views of the intervention and that identified barriers and facilitators in the transcripts by both psychologists and were checked and verified by TS. These words provided an initial coding framework, and the preliminary codes were assigned to the data to describe the content. Third, the codes were collated into potential themes that were reviewed and refined. We undertook a narrative description of themes raised in the participant questionnaires, focus group discussions, interviews and conversations with facilitators. Consensus on emergent themes was reached through regular discussions. The themes were defined and named following agreement. Illustrative quotes are presented in the findings. We reported the results according to the consolidated criteria for reporting qualitative research (COREQ) [30], which is a 32-item checklist for interviews and focus group (Supplementary 2). Findings were not presented to the participants, and they were not invited to give feedback on the findings.

3. Results

Altogether, 49 families (61 participants) enrolled and attended at least one session of a Juntos group. Of these, 20% (12/61) were fathers, and 58% (7/12) fathers attended more than seven sessions. One father was parenting his child on his own, and all other fathers attended the groups with their partners. The four psychologists spent over 240 h in the facilitated group sessions, observing, and discussing with the participants. Sixty-two focus group discussions were held.

Through observation, the psychologists perceived that fathers initially participated in the group as a guide for their child and female caregiver.

> *"The fathers arrive to accompany the mothers in the group and it does not seem easy for them to feel comfortable talking about their feelings and difficulties with parenting."* (Psychologist 01, Rio de Janeiro)

The ice-breakers and practical activities, such as practicing feeding each other, helped the fathers to engage with the group. The facilitators of the group had an important role in encouraging fathers to contribute and providing space for the fathers to talk and the psychologists observed:

> *"When they [fathers] are encouraged and comfortable in the group, they bring up very important topics and are able to show great sensitivity to parenting (...) they demonstrate to mothers that they [fathers] can be good caregivers and that mothers need not be the only specialists in caring for their children."* (Psychologist 01, Rio de Janeiro)

Themes that emerged from the analysis were organized into: (1) Fathers' preference for, and belief about, the Juntos group intervention; and (2) Barriers and enablers to fathers' engagement in the Juntos intervention.

3.1. Fathers' Preference and Beliefs

3.1.1. Advantages of Group Format

Participation in the group offered fathers the opportunity to share similar experiences in caring for their child with complex multiple needs and fathers talked about the benefits of this:

> *"I see the power of this group, this is one of the most important things. It is good to know that there are others in the same situation as you; it gives you a greater strength."* Father 03, Rio de Janeiro

> *"I am moved to hear someone else talking about difficult moments at her child's birthday, when she felt people looking at her child with prejudice (...) She cried, and I felt the same feeling I've already felt so many times with my own child."* Father 05, Salvador

The group setting also provided an environment in which to speak and listen to others in similar situations. The face-to-face format was viewed by fathers as acceptable, due to the benefits of social relationships and learning from others. Fathers were motivated to participate when they were offered speaking space within the parent group.

> *"It is important to be able to speak and know that someone else is available to listen. It is good to express some feelings that are almost physiological."* Father 01, Salvador.

When fathers were offered speaking space, they voiced knowledge of their children, concerns about the reality in which they live and social challenges.

3.1.2. Importance of Clear Objectives and Goals

Fathers attending the Juntos group suggested that including clear goals for their involvement was useful, but had not always been provided before the start of the group. Fathers reported not being aware of the aim of the program and what topics would be covered.

> *"I [initially] thought the aim of the program was to help mothers manage in a better way, because it really is not easy. But the day to day logistics are complicated for both parents, and family members."* Father 02, Rio de Janeiro

> *"I thought this was going to be an academic study like the others, which didn't influence anything in our lives (...) but when we came to the group, my first impression was the welcome we received from the technical team. They were very kind, affectionate, very polite and they explained the project (...) and that encouraged us to attend and to participate."* Father 05, Salvador

3.1.3. Practical Learning through the Intervention

The content of Juntos was found to be acceptable to fathers who attended the groups. For example, both mothers and fathers expressed appreciation of the group format, but there was a difference between parents in the perceived value of the information taught. Fathers saw themselves as being engaged predominantly in their child's care through practical and resource support. Fathers, in comparison to mothers, requested pictures and information that could be applied, such as how to brush their child's teeth in a practical way. They also suggested the use of more stories and repeatedly asked for case studies, photographs and videos of children that were older:

> *"Sometimes the positions that are suggested to be better for my child are not comfortable for her. So, what do I do? I would like to see how older children progress."* Father 05, Salvador

This feedback from fathers during the first phase was used to reform the content of the program; case studies, videos of fathers helping their child with everyday activities (such as brushing teeth and playing in a park) and images of fathers building supportive equipment were developed for the intervention.

Fathers demonstrated initial hesitation with participating in some activities in the program, perhaps to a greater extent than mothers, although these were often overcome:

> *"At the beginning it was a bit weird, funny, and I was shy (...) then I saw the activity and I was involved, and I had fun."* Father 05, Salvador

> *"Even if you have more difficulty, if you are shy (...) the activities are good so that everyone does not just sit and be quiet."* Father 02, Rio de Janeiro

3.1.4. Improved Knowledge and Skills

Fathers viewed improved knowledge and skills as a way to demonstrate their importance as care agents for their child and family. Positive outcomes of participating in the group reported from the fathers' perspective included increased communication with their child, and learning a practical skill that was helpful for the care of their child, such as creating supportive equipment:

> *"Right after the communication module I started to talk more with my daughter. I talk with her when we are playing together, and she answers me laughing."* Father 07, Salvador.

> *"After the positioning module I adapted a shower chair. We take this little chair everywhere we go (...) because she opens her eyes more when she is seated. That it is why I take the chair. She keeps looking, it increases her curiosity."* Father 06, Salvador.

All of the interviewed fathers reported that the parent group helped them to understand their child's development and improved their confidence in caring for their child. Fathers viewed several topics relevant to child development as important for understanding their role as a father in caring for a child with complex needs, including Session 3: Positioning and Moving, Session 5: Communication, and Session 6: Play and Early Stimulation.

> *"I liked the positioning and moving session. We learned with each other and shared tips, and now I know how to help my child to progress and develop through play."* Father 01, Salvador.

> *"We learned about equipment. I understand now that she needs a tray to support her arms in a standing frame. I can help her to do this."* Father 06, Salvador.

3.2. Barriers and Facilitators to Engagement

3.2.1. Time of Delivery

Groups in Salvador were held on Saturdays and groups in Rio de Janeiro were held during the week. The timing and place of program delivery proved a barrier to fathers' attendance, as they were often at work, which was less of a concern for mothers' attendance.

> *"But maybe on the weekend, Saturday, Sunday, could be a solution because during the week the schedule is hard. I try to be present."* Father 02, Rio de Janeiro.

3.2.2. Cultural Norms about Fatherhood and Marital/Partner Relationship

In general, mothers identified with the role of primary carer for everyday need. Fathers that participated in the group were observed to demonstrate affection for their child and voiced desire for a better quality of life for their child. However, women attending the groups perceived that the contribution of men in childcare needs to be increased. This issue was not raised by any of the fathers, and the barrier may be driven by the marital/partner relationship.

> *"And there it is: [T]he mother leads all the activities with the child, who comes to the group? The mother. The person that practices with the child is the mother. I think it is a cultural thing. But if we had some way (. . .) to bring the father (. . .) some formal invitation or something written for him so he can see that he is important too."* Mother 12, Rio de Janeiro

> *"I learned that I have to talk to him [husband] (. . .) and I sat down with him to talk about a consultation (. . .) and he said: I like to know things, but you do not like to speak. In my head it was he [husband] who was not interested."* Mother 14, Rio de Janeiro

The invitation to join the group was administered to the mother, who then became the gate-keeper as to whether the father was invited and encouraged to attend. Whilst women in the group also stated that having fathers there was a positive experience, this was contradicted by other mothers who wanted space to be able to raise issues about the burden of caregiving in a protected space without the father:

> *"We want some space without our husbands, so we can relax, we can talk about them, about a husband who pushes his wife all the time. My husband says 'our son is not getting any better, because you don't do the exercises properly.' When I go home I have to do so many things, prepare meals, cleaning (. . .) how could I do more exercises?"* Mother 11, Rio de Janeiro.

4. Discussion

We explored fathers' preferences for, and perceived barriers to, a group intervention to support care for children with CZS. The group format and content of Juntos was found to be acceptable to fathers who attended the groups. During the development of the content for the Juntos intervention, feedback from fathers during the first phase was directly utilized in the reforming of the program (for example providing case studies, images and films of fathers undertaking practical tasks), which may have made the overall content more useful and acceptable for the later groups. Involving men in program design and implementation is an important factor for promoting male involvement in programs [26]. The fathers who attended over seven of the sessions reported changes in behavior and confidence in caring for their child. The main perceived benefits of the program by fathers were the opportunity to share experiences of caring for their child, and the chance to demonstrate their importance as care agents. Fathers saw themselves as being predominantly engaged in their child's care through practical and resource support.

There remain fewer fathers engaged than mothers as only one in five of the participants of the groups were fathers. However, children with CZS in Brazil may have had more fathers involved than with other similar related conditions, such as cerebral palsy, as expressed by several members of medical and rehabilitation teams [28] from the initial scoping visit for Juntos in April 2017. One possible reason identified was that the high level of discussion and awareness-raising through the media on Zika has meant that there was much less shame related to having a child with CZS than with other similar conditions. Fathers were limited by availability as they saw themselves as the provider of financial support; taking time from work was a barrier to attendance. Women attending the groups perceived that the contribution of men in childcare was not equal, and saw the need for this to change. However, not all women were supportive of fathers being included in parenting interventions.

The difficulty in engaging fathers with parenting programs needs to be addressed given the positive impact that fathers have on their child's behavior and development (e.g., school readiness, cognitive development and pro-social behaviors) [14], and the frequency of paternal abandonment of disabled children [5]. In addition, when both parents engage in parenting programs, the outcomes for children are more positive [31]. A meta-analysis found that, compared to mothers, fathers have a greater ability to influence a child's misbehaviors [32], and promotion of effective co-parenting enhances family functioning and child outcomes [33], but evidence on these father-related outcomes for children with disabilities is lacking [34,35].

These findings have important implications for the targeting and tailoring of parenting interventions in order to increase father engagement, including the need to (i) identify and support strategies to involve fathers from the beginning of interventions, (ii) clearly communicate the goals of the group and what can be expected to be achieved, (iii) provide opportunities for both fathers and mothers to participate, (iv) develop media that shares the men's point of view and includes illustrations of fathers, and (v) include practical information as requested by fathers. It must be ensured that perceptions of childcare as "women's work" are not reinforced. In addition, strengthening fathers' involvement also requires increased awareness from mothers about the need and importance of care offered by fathers to their children.

Alternatives to regular participation in group interventions are also required, since men may be unable to attend the group, do not see its importance, or do not want to engage in group activities or discussions, or mothers may want to have a group consisting solely of women. Alternatives include the development of more printed material, a detailed website, and a greater focus on practical information.

The design of this study has strengths and limitations that need to be considered. This study used qualitative methods to evaluate the engagement of fathers in a group intervention to improve the quality of life of children with CZS and their caregivers. Data presented provides multiple perspectives, from both fathers and mothers, which is a strength of this study. Limitations include the inability to interview any fathers that did not take part in the group intervention about reasons for non-attendance, and the small number of fathers included and interviewed. In addition, participants included some fathers who had not previously participated in group interventions, which may increase the applicability of the findings to fathers who have little experience of parenting interventions. However, we did not specifically measure how many fathers had previously participated in parenting interventions, so we are unable to quantify this. Generalizability of the findings may be limited. Factors that differentiated those that completed the program compared to those that did not were, therefore, not explored. The use of psychologists (non-clinicians) as researchers has implications for assessment of clinical data; however, the use of a specialized pediatric physiotherapist as the third researcher reduces the likelihood of inaccuracies in interpretation of these data. With regards range of impairment in children, we could not compare the severity of impairment of those who participated versus those who did not.

5. Conclusions

The group format and content of Juntos was found to be acceptable to those fathers who attended. Alternatives to regular participation in parent group interventions may be required to provide support and education to fathers of children with CZS. Perspectives, needs and preferences of fathers within their context should be considered in the design and delivery of parent group interventions.

Supplementary Materials: The following are available online at http://www.mdpi.com/1660-4601/16/20/3862/s1, Supplementary 1: COREQ checklist, Supplementary 2: Topic guide: Post-intervention participant interviews.

Author Contributions: Conceptualization, T.S. and H.K.; methodology, T.S., A.D. and H.K.; validation, T.S., A.C.D.V. and B.d.S.M.d.C.; formal analysis, T.S., A.C.D.V. and B.d.S.M.d.C.; investigation, T.S., A.C.D.V. and B.d.S.M.d.C.; data curation, T.S., A.C.D.V. and B.d.S.M.d.C.; writing—original draft preparation, T.S.;

writing—review and editing, T.S., A.D., A.C.D.V., B.d.S.M.d.C. and H.K.; supervision, H.K.; project administration, T.S. and H.K.; funding acquisition, H.K.

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ANNEX 6: SEVENTH RESEARCH PAPER (Published)

Mothers as facilitators for a parent group intervention for children with Congenital Zika Syndrome: Qualitative findings from a feasibility study in Brazil

RESEARCH PAPER COVER SHEET

Please note that a cover sheet must be completed for each research paper included within a thesis.

SECTION A – Student Details

Student ID Number	1405794	**Title**	Mr
First Name(s)	Antony		
Surname/Family Name	Duttine		
Thesis Title	Assessing the feasibility for Juntos: a support programme for caregivers of children with congential Zika syndrome in Brazil during the 2015-16 Zika epidemic		
Primary Supervisor	Hannah Kuper		

If the Research Paper has previously been published please complete Section B, if not please move to Section C.

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Where was the work published?	PLOS ONE		
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RESEARCH ARTICLE

Mothers as facilitators for a parent group intervention for children with Congenital Zika Syndrome: Qualitative findings from a feasibility study in Brazil

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Abstract

Background

The Zika virus outbreak in Brazil (2015–2016) affected thousands of children who were born with Congenital Zika Syndrome (CZS). Families play an important role in their care of children with complex needs, yet their knowledge, experience and skills are rarely harnessed in existing interventions to best support these families.

Objective

This study explores the use of mothers as facilitators for a community-based group intervention for children with CZS and their caregivers in Brazil.

Methods

Four facilitators were trained to deliver the 10-week intervention called "Juntos". Two were mothers of a child with CZS ("expert mothers") and two were therapists (speech therapist and physiotherapist). The intervention was delivered to three groups, generally including 8–10 caregivers. Two researchers, who were psychologists, observed the groups and held focus group discussions at the end of each session. They undertook semi-structured interviews post intervention with a purposive sample of caregivers, and with the facilitators. Observation notes were collated and summarised. Transcripts were transcribed and thematically analysed using five elements to assess feasibility: acceptability, demand, implementation, practicality and adaptation.

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Results

The use of expert mothers as facilitators was considered to be acceptable and there was demand for their role. Their experiential knowledge was viewed as important for sharing and learning, and supporting and encouraging the group. The intervention was delivered with fidelity by the expert mothers. The practicality of the intervention was facilitated by holding the group sessions in the community, providing transport costs to facilitators and participants, paying expert mothers and therapist facilitators equally and supporting the expert mothers through a mentorship programme. Equal payment with the therapist enabled the expert mothers to better facilitate the groups, through increased confidence in the value of their role. Adaptation of the intervention included development of video resources and mentoring guidelines.

Conclusion

The use of expert mothers as facilitators of caregiver groups provides a unique approach to harness the knowledge, experience, and skills of families to provide care, and is likely to be feasible in similar contexts.

Introduction

Developmental disabilities affect at least 50 million children under the age of 5 years globally, and are a major contributor to child and adult morbidity in low and middle-income countries (LMICs) [1]. Children with developmental disabilities have multiple impairments (e.g. cognitive, physical, visual) that have a long-term influence on their health and development [2]. Families play an important role in care for these children, and caregivers of children with disabilities often experience high levels of stress, anxiety, depression, physical exhaustion, and discrimination [3–8]. These experiences contribute to decreased quality of life compared to caregivers of non-disabled children, and may result in reduced effectiveness of parenting [9, 10]. Evidence is lacking on how best to support these families, particularly those living in resource limited settings where healthcare providers with the appropriate expertise may be lacking. Support programmes in partnership with health professionals are increasingly being used to try to fill these gaps, but they often have a top-down approach, rather than drawing on the knowledge, experience and skills of family members.

The World Health Organisation (WHO) recommends a range of interventions for children with or at risk of developmental disabilities. These generally include a focus on empowerment of caregivers and a shift from child-centred to family-centred care, to provide optimal stimulation for development in a safe, stable and nurturing environment [11]. Family-centred care is an approach in which families are recognised as the experts on their child, and work with service providers to make informed decisions about their child's care. There is a growing body of evidence on how best to offer family-centred care in lower resourced settings, including through participatory peer learning [12]. Peer support interventions are hypothesised to work by increasing the amount of social support available to parents and caregivers, and providing that support in a form which is most useful and acceptable to participants [13]. As an example, women's groups that practice participatory learning and action have been shown to improve maternal and child health and empower women [14], and are recommended by the WHO to reduce newborn mortality and improve health in low resource settings [16]. With respect to caregivers of children with developmental disabilities, participatory peer learning in groups provides caregivers opportunities to practice activities with their child and receive feedback, and has greater benefits compared with providing parenting information only [11]. Support groups provide positive benefits to child development and wellbeing, and family functioning [14], improve caregiver understanding, confidence and self-esteem, and reduce self-blame [15]. Additionally, support groups may offer an important social safety net for caregivers who are excluded in their communities [15].

One context in which peer support group interventions could be relevant is for the care of children affected by the Zika Virus (ZIKV). Since late 2015, there have been more than three thousand cases of microcephaly suspected of being related to ZIKV in Brazil [16–18]. Congenital infection with ZIKV is linked to other abnormalities besides microcephaly, including neurological conditions, ophthalmic abnormalities, hearing loss and bone and joint disorders [19–21], now collectively called Congenital Zika Syndrome (CZS). Children with CZS are likely to have complex intellectual, physical and sensory impairments over their lifetime [22]. Additionally, a wider spectrum of developmental impairments may yet manifest in children who were exposed to ZIKV in utero, but do not have CZS, as they continue their development [23]. Provision of family and supportive services is essential to meet the broader needs of these children and caregivers, and to complement clinical and other services available in Brazil.

We therefore developed a group intervention ("Juntos", meaning 'together' in Portuguese) to provide psychosocial support and improve the skills of caregivers of children with CZS in Brazil, to optimally care for their child [24, 25]. The Juntos programme consists of ten sessions offered over a period of 3 months held in the local community. Each session includes icebreaker activities, practical sessions and group discussions, and a psychological support component, and lasts approximately 4 hours. The content of the programme takes learning from health concerns that affect children with cerebral palsy (CP) [26], which is a similar developmental disability to CZS. The programme includes participatory learning about care practices, such as feeding, positioning, communication, everyday activities, play and early stimulation, in addition to disability rights and inclusion (Table 1). Every session includes an emotional support activity to provide a safe environment in which to stimulate open and supportive discussion between caregivers about their successes and difficulties. Programme material is available from www.ubuntu-hub.org

The existing caregiver support programmes for children with CP relied on therapists as facilitators, but within Juntos we pilot-tested the use of a parent of child with CZS ("expert mother") working alongside a therapist facilitator. This approach was used to encourage a more participatory process and atmosphere of sharing between the caregivers. There were concerns about the acceptability and practicality of this approach, and we therefore aimed to explore the feasibility of the use of mothers as facilitators for the community-based group intervention for children with CZS and their caregivers in Greater Salvador, Bahia.

Materials and methods

Ethical approval was gained from Instituto de Saúde Coletiva—ISC/UFBA Ethics Ref 2.369.348, Instituto Fernandes Figueira—IFF/ FIOCRUZ—RJ/MS Ethics Ref 2.183.547 and LSHTM Ethics Ref 13608. Written informed consent was acquired from all participants.

Study design

This study is part of a larger pre post intervention design in Greater Salvador and Rio de Janeiro, which has been described in detail previously [24]. This qualitative study was undertaken between August 2017 and May 2018 in three municipalities of Greater Salvador, Bahia.

Table 1. Juntos intervention module topics.

Module number and title	Topics covered
1: Introduction	About the programme
	Information about Zika and Congenital Zika syndrome
	How to find information
	Personal stories
2: Our child	Introducing your close family and friends
	Development milestones for young children
	Determining your child's progress
	Managing irritability and crying
3: Positioning and moving	How to position children who need assistance
	How to assist children to learn to move
4: Eating and drinking	Feeding challenges
	Practical skills to address challenges for your child
5: Communication	Importance of communication
	Practical advice to help your child communicate
6: Play and early stimulation	Importance of play for children to develop and learn
	Early stimulation
	Making simple toys
	Inclusion of play in the family and broader community
7. Everyday activities	How to use everyday activities to help your child develop
	Managing seizures
8. Uniting our voices	Understand the context of disability rights
	Education
	Communicating with your health team
	Advocating
9. Our community	Who is in your community
	Common barriers to inclusion
	Addressing negative attitudes and exclusion
	Social Activity
10. Next steps	Summing up
	Planning next steps for yourself and the group

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Data were collected from observation of the caregiver groups (n = 30), focus group discussions with caregivers (n = 30), and semi-structured interviews with group facilitators (n = 4) and purposively selected caregivers (n = 9).

Participants and setting

Participants were caregivers of children with neurologist-confirmed CZS in Great Salvador, Bahia. For the purposes of this study, we used CZS to describe any child with impairments that can be directly attributed to Zika. The impairments in children ranged from mild cognitive, communication or functional skill delay to severe delay in all three developmental categories. The average group size was 8–10 caregivers. Each group met weekly for approximately four hours for ten sessions.

The groups were facilitated by one expert mother paired with one therapist (speech therapist or physiotherapist); in total two mothers and two therapists were included. The facilitators were identified and selected by the study site co-ordinator. Selection characteristics included (i) a similar socioeconomic level to participants, (ii) having a child with a pattern of severity of

CZS, (iii) a willingness to help other families, (iv) an active participant in disability rights, and (v) tolerance to personal differences and perspectives (e.g. religion). Both expert mothers were purposively selected by the site co-ordinator, as they had a similar socio-economic level as the group participants, were literate and had completed high school at a minimum as they were required to follow a facilitator manual to deliver the intervention. The hypothesised role of the expert mother was to facilitate a participatory and equal atmosphere, and encourage the sharing of learning between caregivers. For example, the expert mother led the emotional support activity to help to stimulate open and supportive discussion between caregivers about their successes and difficulties in the previous week. The Juntos materials were also developed to include videos of expert mothers and fathers demonstrating techniques such as brushing teeth and play. The role of the therapist was to facilitate the technical aspects of skill acquisition and practice of techniques, such as feeding positions. The therapists and expert mothers completed a joint standardised 5-day facilitators training programme to prepare them for the delivery of Juntos. The expert mother was paid the same as the facilitator therapist (2,100BRL/month, approximately USD500). Transport costs were reimbursed all facilitators and all participants.

Data collection

The reasons for undertaking the research were explained, and data were collected by two female Brazil based research assistants (psychologists). Neither of the psychologists worked in the clinical area or had prior knowledge of the participants. Three techniques were used for the collection of qualitative data. First, participant observations of all ten sessions for each of the three groups were made during each session by the research assistants. An observation checklist was used to assess fidelity of delivery of the intervention by the facilitators. In addition, observation of the ice-breaker activities, practical sessions and group discussions provided an opportunity to examine the interaction of the expert mothers with the participants and facilitator therapist during each session. The psychologists observing the sessions wrote detailed field-notes, and an Excel file was completed for each session. Second, the research assistant (one per site) facilitated focus group discussions related to the content and processes of the session (approximately forty-five minutes) at the end of each session. The research assistant took comprehensive notes of the discussion with the participants. Third, semi-structured interviews were undertaken by the research assistants with both facilitators of the group and a purposively selected sample of participants, to allow for triangulation of data.

Three participants per group were selected after the completion of the ten intervention sessions. Participants were selected at the discretion of the researchers to reflect a broad range of perspectives (e.g. caregivers of children with different severities of disabilities, different caregivers that included mothers, fathers and grandmothers, caregivers of different ages and reflecting a geographic spread). The interview guide was piloted for understanding (S1 and S2 Tables) and interviews lasted on average 1 hour, ranging between 50 minutes and 1 hour 20 minutes. Interviews were designed to explore participants' motivation for attending the intervention, their views around how they were approached to participate in 'Juntos', how they viewed the programme itself, and aspects around engagement were explored. The interviews were audio recorded by digital sound recorder. Focus groups and interviews were conducted in Portuguese.

Data management and analysis

Observational data and comments that related to the role of facilitators from focus group discussions were collated in an Excel sheet (Microsoft Excel 2000 (Microsoft Inc., Redmond, Washington) after each group intervention by one lead UK based researcher, a specialized paediatric physiotherapist trained in qualitative and quantitative techniques (TS). The two research assistants (MM and JR) transcribed and translated the audio-recorded data from the semi-structured interviews.

We assessed the feasibility of the participatory group intervention using a framework based on a model proposed by Bowen et al (2010) [27]. Specifically, we focussed on the facets of acceptability, demand, implementation, practicality and adaptation of the delivery of the programme by expert mothers, in collaboration with a therapist. Acceptability considers how the recipients react to the inclusion of expert mothers. Demand for the intervention is indicated by the documenting of activities by the expert mother in the caregiver groups. Implementation concerns the extent, likelihood, and manner in which facilitation by the expert mother can be fully implemented as planned and proposed, often in an uncontrolled design. Practicality considers the extent to which facilitation by the expert mother can be implemented when resources, time, commitment, or some combination thereof are constrained in some way. Adaptation focusses on how the role of the expert mother may need to be adapted for a new situation. The remaining three areas outlined in the Bowen model of limited efficacy, expansion and integration are to be explored in future studies.

Words and phrases relating to the effect of having an expert mother facilitate the intervention were identified and coded to the feasibility framework. They were discussed between the two research assistants for agreement and checked and verified by TS. Consensus on coded phrases was gained through discussion. We undertook a narrative synthesis of the findings. Illustrative quotes are presented in the findings. We reported the results according to the consolidated criteria for reporting qualitative research (COREQ) [28], which is a 32-item checklist.

Results

Three Juntos groups were convened in Greater Salvador (in Simões Filho, Lauro de Freitas, Camaçari) between August 2017 and May 2018. A total of 25 families and their children enrolled across the three groups. Thirty-eight participants (mother, father or other caregiver) attended at least one session. The two research assistants undertook 120 hours of observation.

Acceptability

Support from expert mothers was considered to be acceptable and highly valued by the participants in this study. Both participants and facilitators talked about the benefits of including the expert mother and the role of the expert mother was viewed as acceptable in two ways; (1) promoting sharing and learning from each other, and (2) supporting and encouraging each other.

(1) Sharing and learning from each other. The research assistants observed that inclusion of the expert mothers contributed to an environment in which participants shared their own experiences and listened to others facing the same challenges. Expert mothers' knowledge and expertise from first-hand experiences was shared with others in similar situations, and an example of sharing from the perspective of the expert mother includes:

> "*I experienced something really incredible when a mother did not believe that her daughter was capable of doing something and, at that moment, I remembered that my son had had the same difficulty. I knew that her child was capable, so I shared strategies that I learnt for my child and she was able to do it.*" (Expert mother 01)

Expert mothers led by example to increase the inclusion of other family members and their communities to assist with care of their child. One expert mother required support from her husband to facilitate a group and she explained to the group:

> "*to be here [facilitating the group intervention] I had to train my husband*" (Expert mother 02)

This example demonstrated to other caregivers what may be possible when seeking support from other family and community members.

Caregivers and expert mothers reported finding a shared social identify. This was evident through the shared experience of their children's unpredictable medical needs, which was faced by the expert mothers and the participants. For example, an expert mother was unable to attend one session due to her child being ill:

> "*I cannot be here today because I have to be in hospital with my child*" (Expert mother 01)

Fostering a shared sense of identity led caregivers to meet outside of the support programme after the three months concluded, expanding their social and support network.

(2) Supporting and encouraging each other. The expert mothers were regularly observed to encourage parents that "you know better than you believe". Through the process of participatory peer learning, one expert mother told us:

> "*By learning and acting as an expert mother, Juntos enabled me to find out about so many new things and people, who will forever remain part of my story. We had so many questions, and together we mothers were heard and we listened to each other. It was a very important change because normally families believe that they should just listen to professional therapists.*" (Expert mother 01)

The expert mothers appreciated how difficult it could be to meet the needs of a child with a disability. Participants reported that acceptance from the expert mothers helped to increase their self-belief and confidence in their own ability to care for their child. This was facilitated through being seen as on an 'equal level':

> "*At the first sessions I felt like a mere spectator. I was only there to "learn" to handle my daughter. But, what does that mean, "learn"? Does it mean that I, as her carer, who is with her every second of the day, has nothing to offer? . . .we ran the sessions so that the families had equally important roles.*" (Expert mother 02)

The benefits of including the expert mother and reducing the traditional medical hierarchy were also articulated by the therapist facilitator. One facilitator therapist reported change in how she provided support to families:

> "*Being a facilitator is not easy, you need to be available to care. . .sharing the facilitator space with a mother enabled me to get closer to a reality that I had only experienced from a distance. Hearing from other carers and sharing so much knowledge, which only they have, was transforming.*" (Facilitator therapist 02)

The expert mothers were viewed as acceptable to participants as they encouraged the provision of mutual support. This provided a strong motivation for participants to offer the same support to other caregivers, which they had benefitted from themselves:

> *"No one has asked before about my hopes or dreams. Today I see if I am well then I can take better care of my child. Others should know this too. We must take care of ourselves. . .it is important to help us understand this. I now tell others about this often."* (Participant 04)

There was no reported objection from other mothers about payment of expert mothers as facilitators. Limited acceptability may occur when caregivers are concerned with the comparison between their own and the child of the expert mother. For example, if participants consider the child of expert mothers much less affected by CZS compared with their own child. However, this was not experienced within these groups.

Demand

There was a demand for expert mothers expressed by the other caregivers, and participants reported valuing the opportunity to share personal experiences and problem solve together with someone who understood their circumstances. The expert mothers provided an example of what may be possible through their lived experience, which indicates the way the intervention was delivered was appropriate and accessible to caregivers. This experience was underpinned by the importance of building hope and confidence. One participant acknowledged:

> "*if she is telling me, then I can do it.*" (Participant 07)

Additionally, as the facilitator pairs worked together over time, the value of the expert mothers became more clear to the therapist facilitator. The therapist facilitators voiced wanting to continue facilitating groups of parents alongside an expert parent:

> *"Working together with a mother facilitator, my senses are much more attentive to each family's story, and these stories have acted on me and changed me, both personally and professionally."* (Facilitator Therapist 02)

Expert mothers were viewed to improve the understanding of the need of families and the professional abilities of the therapist facilitator to address these needs.

Implementation

The intervention content and processes were delivered with fidelity by the expert mothers; the content (e.g. topics covered from module one to ten, the emotional support activity) and processes (e.g. participatory learning, adult learning techniques) of the intervention were used to a large extent by the expert mothers. The expert mothers enacted their expected role within the group. The research assistants observed that they facilitated a participatory and equal atmosphere, and encouraged the sharing of learning between caregivers. The expert mothers filled their planned role (described above) and this differed from the role of the therapist, who facilitated the technical aspects of skill acquisition, such as feeding positions. However, expert mothers described a tension in their role as a facilitator:

> *"There are some moments where I freeze. It is more difficult that I thought it would be, to be a mother and a facilitator, because I keep pushing myself to act like a therapist during the theoretical moments."* (Expert mother 01)

Both facilitators contributed to practice of techniques. The relationship between the facilitator therapist and the expert mother developed over time through the delivery of the ten session programme and this relationship was sustained by enthusiastic therapists who advocated for family centred services.

Interviewer: *"What would you like to do differently in the next session?"*

> *"We would like to have a meeting, before the session, to prepare our work together. This will help us to feel more confident to lead the next session."* (Facilitator therapist 01)

However, changes to the on-going support through increased access to mentors was required. For example, the expert mothers led the emotional support activity as part of every session, but they did not have a mental health training. The observing research assistants, who were trained psychologists, identified the need to provide regular psychological support to the expert mothers after delivering each session. The expert mothers were therefore provided with weekly mentoring by the psychologists. This input deviated from the limited support that was initially planned.

Practicality

The practicality of the intervention was facilitated by holding the group sessions in the community, providing transport costs to the expert mothers, paying expert mothers and therapist facilitators equally and supporting the mothers through a mentorship programme. However, certain challenges were identified in the practicality of including mothers as facilitators. There were two occasions when an expert mother was unable to attend. The weekly sessions were held on different week days and the other expert mother was able to assist with one session. In the other session, it was run only by the facilitator therapist. The expert mother in these cases was unable to attend due to her child being ill. Over the three-month period, the majority of caregivers missed at least one session due to illness of their child. Fifteen (60%) families attended seven or more modules. Four families participated in all sessions. Module nine, titled 'Our Community', which includes a community day and celebration, was the best attended (group one n = 9; group two n = 7; group three n = 6). The 'Play and Early Stimulation' module delivered in week six was the least attended (group one n = 4, group two n = 4, group three n = 3).

The group sessions were designed to be held over two to three hours, and typically took approximately four hours. Observation notes suggest that the logistics of delivering the intervention could be challenging, with sessions often starting behind schedule and all the content was not covered in some sessions. As the group facilitators became more familiar with the content, the delivery of the intervention did not exceed the allocated time.

With regards finance, it appeared that equal payment enabled the expert mothers to better facilitate the groups, being more confident of the value of their role in a pair with the therapist. However, the decision to pay expert mothers and therapist facilitators the same amount was initially viewed as unfavourable by the administration team due to precise costing guidelines provided for therapists and relevant experience, with the suggested payment for expert mothers as 1,500BRL/month, approximately USD360. The estimate of salary cost for one therapist to deliver 10 group sessions over three months is 6,300BRL (approximately USD1,500) and the incremental cost of including an expert mother would be 6,300BRL (approximately USD1,500) per programme.

The success of the facilitator partnership is likely dependent on both personality and perspective of the pair. Selection characteristics of expert mothers included a similar socioeconomic level to participants and having a child with a severe pattern CZS, with personal characteristics of consideration and understanding. These characteristics may link to a sense of belonging and creation of common ground between the participants.

Adaptation

Few adaptations were considered to be needed to the role of the expert mother, following the feasibility study, excepting the increased mentoring on providing mental health support, already described. Weekly mentoring guidelines were developed by the research team, and in partnership with the expert mothers and facilitator therapists, after the first pilot. These guidelines were used to assist the research assistants (psychologists) to provide targeted mentoring for the facilitators to support emotional wellbeing of participants. Resources and processes were refined through the piloting of the 'Juntos' intervention and sections of the paper based manual were transformed to video format. The videos were reported as helpful to expert mothers to further demonstrate techniques to participants. Additional resources were requested by the participants and these included:

> *'We could bring pictures of our shower chairs, or maybe you could show us pictures with the different phases of communication and development of eating, so that we can identify where our children are now, and what the next steps will be–just like we did for the development of moving."* (Participant 08)

There was a need for the study co-ordinator to provide support to facilitators in organisation of logistics e.g. travel, food and intervention materials. The study co-ordinator also provided instruction, in addition to the comprehensive intervention manual, on how to run practical sessions in order to maximise their impact. Therefore, more practical support may be needed in organizing the sessions in future programmes.

When considering future scale up in Brazil, it is likely that adaptation of the training to include a greater number of expert mothers compared to the number of facilitator therapists may be required to account for contexts with fewer number of therapists and as mothers may not have the capacity to travel as much or facilitate as many groups as the therapists. The 'Juntos' intervention performed in a similar way with all three groups, which consisted of a different population in each group. The co-ordinator selected different sites for the groups and it was reported as important for the expert mother to be local to the community in which the community-based group intervention is run, whilst it is acceptable for the facilitator therapist to travel further and be involved in a greater number of groups.

Discussion

The theory of change guiding the 'Juntos' intervention is that a sense of belonging and creation of common ground would provide an environment to improve the knowledge and skills of caregivers through a social support network. This approach, in turn, would improve the quality of life of children with developmental disabilities and their caregivers, as indeed has shown to be the case for a similar intervention [26]. We introduced the expert mothers as facilitators in order to reinforce the participatory and peer learning aspects. Our findings show that the role of mothers as facilitators in community group interventions is likely to be a feasible approach to participatory peer learning to improve care for children with developmental disabilities. The use of expert mothers was considered to be acceptable for participants and facilitator therapists and there was demand for their role.

Comparison to other studies

Our findings contribute to the growing body of evidence of the importance of participatory peer learning to improve child care and support caregivers' psychological and emotional wellbeing [29, 26]. A systematic review and meta-analysis of randomised controlled trials undertaken in Bangladesh, India, Malawi, and Nepal of women's groups practicing participatory learning and action show that these practices improve maternal and neonatal survival [30], by increasing appropriate care-seeking, home prevention and care practices for mothers and newborns [31]. The women's groups drew on principles of Paulo Freire's work [32], namely: (i) health challenges are often rooted in powerlessness, and can be addressed by social empowerment; (ii) including dialogue and problem solving in health education is more empowering than information giving; and (iii) communities can develop critical consciousness to recognise and address the underlying social and political determinants of health. While there is no single recognised theory of how women's groups practising participatory learning and action achieve their health impacts [33], group participation and membership offers a valuable social support network to navigate medical hierarchies, and may contribute to change in care practices through increasing confidence of caregivers.

Comparison of our findings is also possible with other interventions for children with developmental disabilities in different low resourced settings, such as: (i) a Caregiver Skills Training (CST) for caregivers of children with intellectual disabilities [34, 35], developed by the World Health Organisation, (ii) 'Titukulane', an eight module community group intervention that aims provide contextualised psychological support to caregivers of children with intellectual disabilities [36], (iii) Learning through Everyday Activities with Parents (LEAP-CP), which aims to improve the mobility of children with cerebral palsy over 30 weekly peer-to-peer home visits [37], and (iv) PASS, a parent-mediated intervention for autism spectrum disorder in India and Pakistan [38] that was adapted for delivery by non-specialist workers and uses video feedback methods to address parent–child interaction. The focus on caregiver involvement is a common thread in all of these interventions, which is critical, particularly where there are few health services. These interventions demonstrate that reaching family-centred care goals can be facilitated through having mothers as facilitators. However, formal evaluation of their effectiveness and cost-effectiveness is lacking.

Value added from expert mothers

A scoping visit by the research team to Salvador, Bahia, prior to piloting the intervention, demonstrated that the majority of support that was provided to families was medically orientated and that informal support networks that were established varied in focus and structure [39]. For example, some groups focussed on advocacy and promoting children's rights, while many mothers reported being part of WhatsApp groups with other caregivers, which provided some social and emotional support on an ad hoc basis. The addition of an expert mother to the 'Juntos' intervention therefore supports the provision of family-centred services through including the sharing of lived experience of caring for a child with CZS. In this study, the role of expert mothers was seen specifically as being important to share and learn together, and to provide support and encouragement.

Limitations

This study has limitations. We explored feasibility of use of two expert mothers in only one setting. We did not compare different strategies of delivery. Nor did we explore in detail what aspects of expert mother were critical to success. Consequently, it is difficult to identify the extent to which the perceived feasibility depended on specific personal characteristics of individual expert mothers, and how much on the use of an expert mother of any kind. The therapist and expert mothers were paid and equal amount and this may have influenced their strong commitment to the programme. Only a subset of participants was selected to participate in semi-structured interviews and this selection, although purposive to gather a range of perspectives, may have introduced a positive reporting bias in the responders. Attendance rates may have influenced participant responses although there was no evidence of better retention for earlier compared to later modules. Furthermore, the selection of interview participants, although done to gather a range of perspectives, may have brought an inherent bias to the qualitative data that was analysed from the participants. However, the triangulation of findings between participants, expert mothers and facilitator therapists gives us confidence in our results. In addition, with a small sample size it is not possible to draw any firm conclusions with regards limited efficacy of the programme or the impact on families of children with different severities of functional impairment.

Implications and future steps

We have identified research questions that have been framed by the gaps in evidence of this feasibility study. Future studies may seek to establish the cost-effectiveness and long- term benefits (such as improved survival and hospitalization rates) of the inclusion of expert mothers in the delivery of the Juntos intervention. Future research should also investigate the impact of the personal characteristics and experience of the person offering support, and the impact of peer support on caregivers' relationships with health care professionals. In addition, the role of equal payment between the two facilitators warrants further attention.

Conclusion

Caregivers with similar life experiences may provide innovate community support to families of children with CZS in resource limited settings. The use of expert mothers in a participatory group setting offers a unique approach to harness the capacity of families to provide care for their child and may be feasible in similar settings. Future consideration for scale up in Brazil includes accounting for resource-limited contexts with fewer number of therapists. It is likely that adaptation of the training to include a greater number of expert mothers compared to the number of facilitator therapists may be required.

Supporting information

S1 Table. Post intervention interview guide–facilitators.
(DOCX)

S2 Table. Post intervention interview guide–participants.
(DOCX)

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ANNEX 7:

Comments from Peer reviewers relating to Paper 1

Open Peer Review

Current Peer Review Status: ✓ ✓

Version 1

Reviewer Report 28 June 2019

https://doi.org/10.21956/wellcomeopenres.16458.r35511

✓ **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

https://doi.org/10.21956/wellcomeopenres.16458.r35510

✓ **Michel D. Landry**
Physical Therapy Division, Department of Orthopeadic Surgery, School of Medicine, Duke University, Durham, NC, USA

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, structed 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.

ANNEX 8:

Comments from peer reviewers regarding Paper 4

Open Peer Review

Current Peer Review Status: ✓ ✓

Version 1

Reviewer Report 23 May 2022

https://doi.org/10.21956/wellcomeopenres.19258.r49915

✓ **Sarah Mulkey**
[1] Prenatal Pediatrics Institute, Children's National Hospital, Washington, DC, USA
[2] George Washington University School of Medicine, Washington, DC, USA

Thank you for the opportunity to review this manuscript. It was well written and described a special support program for families of children with congenital Zika syndrome. The quotes of the participants show just how important the program was for them and it sounds that it has made a difference for the families that participated. Families often benefit so much by the support of other families going through similar situations. The program seems well structured and that it could be refined to enhance program completion and session timelines. A follow-up of how lasting the benefits of the program are for the families would be very interesting.

"Are all the source data underlying the results available to ensure full reproducibility?" - Partly, but the authors acknowledge this and provide a valid reason of confidentiality, given the small sample size. Full data can be requested, and considered upon review to be shared.

Some additional questions:

1. Can the authors please provide more detail of the PEDS QL and how this is scored. Is the total score a sum of all of the dimensions? What values are considered high and low?

2. I really loved the direct quotes! They were beautiful and very descriptive about the experiences of these families.

3. Why do the authors think that the Rio 2 cohort had the lowest number of families who completed the program? What was different about this particular group?

4. These cohorts met prior to the COVID-19 pandemic. Can the authors provide any information about any support that has continued or how support has been disrupted for these families?

5. Can the authors discuss any changes that they feel would benefit from the program based

on what they learned regarding the feasibility?

Is the work clearly and accurately presented and does it cite the current literature?
Yes

Is the study design appropriate and is the work technically sound?
Yes

Are sufficient details of methods and analysis provided to allow replication by others?
Yes

If applicable, is the statistical analysis and its interpretation appropriate?
Yes

Are all the source data underlying the results available to ensure full reproducibility?
Partly

Are the conclusions drawn adequately supported by the results?
Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Child neurology, neurodevelopment, congenital infections

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 14 April 2022

https://doi.org/10.21956/wellcomeopenres.19258.r49027

✓ **Paul Lynch**
Inclusive Education, School of Education, University of Glasgow, Glasgow, UK

Juntos programme is aimed at caregivers whose children have Congenital Zika Syndrome and other neuro-developmental disabilities in Brazil. The 10 session programme covers facilitating play, feeding and communication, knowing rights and living in the community. The paper is clearly set out, very accessible to read and refers to up-to-date literature. The mixed method design is appropriate to the type of intervention. The Bowen et al. feasibility framework is appropriate to measure the feasibility of the training programme. The data analysis is clear and succinct. The qualitative data provides useful contextual detail about the impact of the sessions on individual caregivers. All the data sources are made clear for reproducibility.

It would be helpful to know a little more about the intervention in terms of duration, length of each session and what materials were used to help facilitate the sessions. It's curious why cognitive functioning score was lower after the training. Is there a plausible reason why this was the case?

The conclusions drawn from the data are concise and not inflated, considering the small number of participants. There is reasonable amount of evidence to show that the intervention has been successful and 'can work'.

An important contribution to an under-researched field of disability.

Is the work clearly and accurately presented and does it cite the current literature?
Yes

Is the study design appropriate and is the work technically sound?
Yes

Are sufficient details of methods and analysis provided to allow replication by others?
Yes

If applicable, is the statistical analysis and its interpretation appropriate?
Yes

Are all the source data underlying the results available to ensure full reproducibility?
Yes

Are the conclusions drawn adequately supported by the results?
Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: My expertise lies in early childhood development, education and disability in low and middle income countries. I have a strong background in developing and implementing caregiver and pre-school teacher training programmes for young children with disabilities in LMICs.

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.