Setting implementation research priorities to reduce preterm births and stillbirths at community level

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Summary Points

- Preterm birth complications are the leading cause of neonatal mortality contributing 1 million deaths a year. Stillbirths account for another 3.2 million deaths. Both these causes of perinatal mortality are inextricably linked to maternal health and to conditions at birth.
- While some community based interventions have been proven to be effective in controlled settings and specific contexts, the implementation research challenge is to understand how to sustain these interventions at scale in different contexts.
- A systematic process based on the Child Health and Nutrition Research Initiative (CHNRI) methodology was used to score and rank implementation research questions regarding community based maternal newborn interventions that address prematurity and stillbirths in different contexts at scale.
- The top five questions addressed equity (e.g. reaching the poor and marginalized, reducing financial barriers), behavioral practices and skills (e.g. engaging with social norms, identifying prematurity) and quality of care provided by community health workers. The top 15 research questions encompassed issues pertaining to behavioral interventions, community health workers, referral and managing health systems.

Introduction

It is estimated that there are annually 3.2 million stillbirths globally, 1 million of which occur during birth (**1**). In addition, complications from preterm births (before 37 completed weeks of gestation) are the leading cause of death for newborns, contributing 1 million or 12% of child deaths (**2**,**3**). In 2009, more than 200 stakeholders attended the International Conference on Prematurity and Stillbirth convened by the Global Alliance to Prevent Prematurity and Stillbirth (GAPPS).[1] The community expert group convened at the conference by GAPPS included fifteen members drawn from technical and funding organizations, as well as program implementers and researchers from around the world[2]. In their discussions, the group framed efforts to address preterm and stillbirths within the broader context of maternal-newborn interventions. As most of the evidence supporting these interventions emanates from research projects in controlled settings in specific contexts, they identified the main challenge of implementing at scale in different contexts. Based on these discussions, the group began a research prioritization exercise for implementation research on community based interventions at scale in different contexts. In this paper, we present the results of this exercise.

Methods

A number of research prioritization efforts have recently been applied to various health topics and health system themes (**4-7**). The GAPPS community expert group chose the methodology proposed by the Child Health and Nutrition Initiative (CHNRI) to systematically list and score research questions. The CHNRI methodology was selected because it has a conceptual framework (**8-10**) that has been used in numerous areas by different national and international organizations (**11-16**)[3]. Guided by the CHNRI methodology, we followed three main stages to derive research priorities, as detailed in **Box (1)**:

[1] http://www.gapps.org/
[2] See acknowledgement section for specific names.
[3] Further information on CHNRI methodology, validity and potential limitations are discussed in **Supplementary Table S1**.

Respondents were fairly even in terms of gender (39% women) and diverse in terms of regional representation (26% sub-Saharan Africa, 16% Asia, 16% Latin America, 10% Europe, 32% North America). While a substantial number of respondents were based in North America, they all work full-time in developing country contexts. Half of the respondents were based in research institutions, whereas the other half were in charge of implementing programs whether through non-governmental organizations, UNICEF country offices or USAID headquarters. Non-respondents were not significantly different from respondents (see **Supplementary Table 2**).

Results

The research question that was highlighted as the most important out of the 55 reviewed was "Evaluate ways to reduce the financial barriers to facility births at the community level – e.g. user fee exemptions, emergency loans, conditional cash transfers, transportation vouchers, etc." Other research questions among the top five prioritized also addressed equity issues (reaching the poor and marginalized), but also behavioral practices and skills (engaging with social norms, identifying prematurity) and service delivery with regards to quality of care provided by community health workers. The remaining top ten research questions (**Table 1)** include other behavioral skills and practices (thermal care and feeding for preterm babies, birth planning), concerns about how to best motivate and compensate community health workers and their supervisors, as well as different dimensions of making referral more effective. Congruent with the need to measure and maintain quality of care by community health workers as a priority, rational drug use by community health workers and community engagement with regards to audits was also listed among the top 25 research questions that received an overall "research priority score" (RPS) of 0.75 or greater (**Table 2**).

Table 3 shows the 10 research questions which were assigned the lowest research priority scores. Several broad policy questions (human resource planning, gender profiles, budget flows, accountability and monitoring systems) are listed here, along with some questions related to the sequencing of community interventions and one specific question regarding private provider practice (delayed cord clamping). Questions from almost all research avenues were found among the bottom ten research questions, suggesting that no one area

was completely discriminated against by the scoring. Furthermore, even these lower ranked research questions received relatively high RPS scores compared to other CHNRI exercises. The research priority score (RPS) for all 55 questions ranged from 0.86 to 0.56, in contrast to other CHNRI exercises, which generated RPS ranges from 0.90 to 0.25 (**12-16**). This suggests that respondents collectively considered all implementation research questions as fairly important.

Research questions did vary in terms of their specificity. For example, broad questions like "evaluate community based strategies to reach the poor and marginalized" were scored alongside very specific questions like "evaluate ways to provide thermal care and feeding the preterm baby". Both broad and specific questions were ranked in the top and bottom 10 implementation research questions, suggesting that this did not lead to a bias against the kind of question asked.

The CHNRI methodology evaluates certain dimensions of each research question according to selected criteria. For example, "Evaluate methods and levels of accountability that can be ensured" was not considered easily answerable and "Evaluate ways to ensure delayed cord clamping in deliveries assisted by private providers" was not scored as likely to attract funding support or national policy attention. Among these different criteria, the most discriminative one was the one related to disease burden reduction, while the criteria that was least discriminative was the one regarding answerability in an ethical way.

The relatively high mean scores assigned to questions across all criteria (apart from disease burden reduction) indicate that most of the respondents were fairly optimistic about the value of implementation research questions. Average expert agreement ranged from 0.82 to 0.49. Similar to other CHNRI exercises, average expert agreement showed a direct positive association with research priority scores, indicating that there was more agreement among experts about what were the priority research questions. This is a property that is inherent to the way AEA is measured: very high or very low RPS scores require high levels of expert agreement, while substantial disagreement among experts will lead to RPS moving closer to a mean value. (**12-16**).

In order to explore whether there was any systematic bias against certain questions due to the profile of the respondent, we analyzed scores for researchers and implementers. We found at

least a 10% difference in the scoring assigned for 20% of the research questions as listed in **Table 4.** The 11 questions for which there was a significant difference between researchers and implementers are spread across each research avenue, suggesting that there was no one particular research area affected by this difference of opinion. In 10 out of these 11 questions, implementers ranked the implementation research question as being of higher value than researchers.

Discussion

The top 25 research questions that have been prioritized span a broad range of issues. These implementation research priorities include ways to foster and sustain specific behavioral skills and practices at the community level, engaging communities in monitoring service delivery through audits, as well as improving referral. With regards to service delivery, a host of implementation research questions regarding the management of community health workers, along with the health system supports they require to function were stressed. Lastly, issues that relate to equity, financing and referral were highlighted, reflective of how community based approaches cannot be dealt with in isolation from broader health system concerns.

While many of the implementation research priorities identified can be generalized across community based maternal, newborn and child health areas, there are a few distinctions that may be particular to this specific exercise. Issues related to referral were reflected three times within the top 25 research questions. There is little implementation research on linking families from homes to facilities or referral more broadly in low income country contexts (**17-19**). While important gains have been made with taskshifting, effective and equitable referral remains vital as the most serious cases of prematurity and other birth complications cannot be handled at the community level.

Implementation research questions related to community engagement and some other broader policy concerns central to managing health systems, like human resource planning and monitoring systems, were overall not prioritized very high by respondents. Nonetheless, it is emphasized that even the bottom 10 research questions received relatively high research priority scores. Other CHNRI exercises had lower research priority scores than those found in this exercise. This could be because they had more discriminatory criteria or because

previous exercises compared different kinds of research (basic science vs. implementation research). It may be easier for experts to discern between very different research areas (basic science vs. implementation research) than to discern between areas of implementation research, which they may consider to be of relatively similar importance.

In addition, many of the implementation research questions do not by themselves contribute to improved maternal newborn outcomes. Their value comes forth when combined with other implementation issues that together make a more comprehensive and coherent community based response with linkages to primary health care service delivery. It might therefore be difficult for respondents to think about specific implementation research questions in isolation from their broader social and health systems context.

The partiality towards some areas of implementation research could reflect the profile of respondents. Comparing scoring by implementers and researchers did find some differences, not across any particular kind of research question, but in the direction of the bias, with implementers ranking implementation research questions higher than researchers. The reasons for this difference among 20% of the questions are not known, but seem to indicate that implementers perceive the results of implementation research to be more powerful if effectively implemented than researchers do.

While the CHNRI methodology provides a systematic and transparent methodology to rank research questions that purposefully avoids biases introduced by group dynamics dominated by powerful individuals, it still is a very lengthy process to undertake. Respondents have to score 55 research questions according to 5 criteria that have 3 sub-components each. This amounts to 825 dimensions to respond to in the spreadsheet. This complexity doesn't make it easy to fill out the spreadsheet or help response rates. Eliciting participation via email alone was not successful as only 42 out of 85 experts responded to the preliminary email. The 42 experts that did express interest did reflect a group that was more familiar with the GAPPS conference and had a current working relationship with the lead authors who managed the exercise.

Despite these qualifications, this exercise still represents an important collaboration between researchers and program implementers to jointly identify the key implementation research questions vital to improving community based maternal and newborn interventions that

address preterm and stillbirths. This exercise also developed new criteria that were deemed more appropriate to implementation research, which require further testing and refinement to improve their discriminatory power.

Success in reducing stillbirth and prematurity rates, and in increasing the survival of preterm infants in low-income countries is strongly dependent on achieving high and equitable coverage with existing cost-effective interventions (**20, 21**). Yet coverage of such interventions remains unacceptably low in most countries. For example, across 68 countries with the highest mortality, only 54% of women deliver with a skilled birth attendant and 38% receive a postnatal visit (**22**). Furthermore, coverage levels are particularly low among poor and rural families in these countries. Community-based interventions are therefore essential to reach population subgroups whose current access to health facilities is severely limited. The effect of expanding coverage of family and community care to 90% can by itself lead to a 15-32% reduction in neonatal mortality (**22**). Nonetheless, the knowledge gaps regarding how to sustain these programs at scale in different contexts remain significant.

While important reviews (**23-28**) have helped to spur attention to community based maternal newborn issues, with intriguing results regarding specific interventions (**29**, **30**), the implementation research priorities identified in this article will, we hope, help to secure further research attention and financing for this important area. Priority research areas identified include equity concerns, such as removal of financial barriers and responsiveness to the poor and marginalized, specific behavioural skills and practices, and the management of community health workers including referral care. The challenge is raised: will communities, governments, donors, research institutions and international organizations respond?

(2,011 words)

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Abbreviations

Global Alliance to Prevent Prematurity and Stillbirth (GAPPS)
Child Health Nutrition Research Initiative (CHNRI)
World Health Organisation (WHO)
Millennium Development Goal (MDG)
Research Priority Score (RPS)
Average Expert Agreement (AEA)

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Supporting information

Supplemental material **Table S1 to S3**

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Table 1: The 10 research questions that received the highest overall research priority score (RPS) (with average expert agreement (AEA) shown)

Rank	Proposed Research Question	Answerable?	Burden Reduction?	Scale up?	National policy?	Ownership?	RPS	AEA
1	Evaluate ways to reduce the financial barriers to facility births at the community level (user fee exemptions, emergency loans, conditional cash transfers, transportation vouchers, etc)	0.930	0.663	0.845	0.877	0.895	0.858	0.821
2	Develop and validate strategies to identify preterm babies at community level by CHWs and family members	0.942	0.640	0.750	0.795	0.821	0.832	0.801
3	Evaluate different methods of behavior change that overcome harmful practices and promote positive cultural and social norms	0.904	0.696	0.909	0.886	0.772	0.829	0.794
4	Evaluate effective community-based strategies to reach the poor and marginalized	0.895	0.670	0.843	0.911	0.868	0.825	0.772
5	Evaluate ways to measure and maintain quality of care provided by CHWs	0.967	0.698	0.851	0.737	0.776	0.825	0.794
6	Evaluate ways to provide thermal care and feeding for the preterm baby	0.958	0.686	0.802	0.737	0.798	0.822	0.777
7	Evaluate financing measures at the community level that improve referral	0.915	0.500	0.848	0.729	0.877	0.817	0.779
8	Evaluate ways to motivate and compensate CHWs and their supervisors	0.983	0.596	0.929	0.700	0.817	0.814	0.785
9	Evaluate how to maximize referral compliance especially for the poor and marginalized	0.959	0.587	0.796	0.772	0.833	0.813	0.757
10	Evaluate ways to engage communities in birth planning for normal and at risk pregnancies	0.908	0.630	0.740	0.741	0.888	0.812	0.759

Table 2: Top 25 research questions by research area with a research priority score of 0.7 or above.

Rank	Research Area	Research Questions
12	Community Engagement	Evaluate how community audits could improve access and quality of services
14		Evaluate how community engagement improves referral and counter-referral
2	Behavioral Skills and Practices	Develop and validate strategies to identify preterm babies at community level by CHWs and family members
3		Evaluate different methods of 14ehaviou change that overcome harmful practices and promote positive cultural and social norms
6		Evaluate ways to provide thermal care and feeding for the preterm baby
10		Evaluate ways to engage communities in birth planning for normal and at risk pregnancies
13		Assess the impact of initiation and continuation of Kangaroo Mother Care at home on survival of preterm/LBW babies in setting with high home births
15		Evaluate ways to ensure the sustained use of ITNs by pregnant women and newborns
19		Evaluate ways to garner community support to ensure early and sustained breastfeeding
23		Evaluate ways to maintain CHW neonatal resuscitation skills
22	Rational Drug Use	Assess methods to ensure rational drug use among CHWs
5	Community Health Worker	Evaluate ways to measure and maintain quality of care provided by CHWs

8		Evaluate ways to motivate and compensate CHWs and their supervisors
16		Evaluate how CHWs can improve referral and counter-referral
17		Evaluate ways to assure continuous supply of essential medicines and inputs for CHWs
20		Evaluate ways to improve retention of CHWs
21		Evaluate how to measure good supervision for CHWs and different ways of providing it
24		Assess the optimal number of activities and population coverage required to maintain case load and skills of CHWs
25		Evaluate the equity impacts and effectiveness of CHW services when delivered with user fees or drug cost-recovery fees
1	Management and Health Systems	Evaluate ways to reduce the financial barriers to facility births at the community level (user fee exemptions, emergency loans, conditional cash transfers, transportation vouchers, etc)
4		Evaluate effective community-based strategies to reach the poor and marginalized
11		Evaluate demand-side financing mechanisms (e.g. insurance, demand side subsidies, vouchers)
7	Referral	Evaluate financing measures at the community level that improve referral
9		Evaluate how to maximize referral compliance especially for the poor and marginalized
18		Evaluate the barriers at the community and provider level that cause poor referral

Table 3: The 10 research questions that received the lowest overall research priority score (RPS) (with average expert agreement (AEA) shown)

Rank	Proposed Research Question	Answerable?	Burden Reduction?	Scale up?	National policy?	Ownership?	RPS	AEA
46	Assess the gender distribution of CHWs and its implications in terms of their acceptability and effectiveness	0.925	0.343	0.602	0.619	0.633	0.639	0.574
47	Assess how CHWs and other kinds of frontline health workers are represented in human resource policies, strategies and legislation	0.925	0.271	0.556	0.583	0.692	0.638	0.593
48	Evaluate methods of integrating community-based data collection into district HMIS	0.930	0.298	0.636	0.526	0.579	0.628	0.565
49	Evaluate methods and levels of accountability that can be ensured	0.650	0.345	0.565	0.510	0.608	0.618	0.540
50	Assess the methods of tracking budget allocations and flow	0.889	0.256	0.482	0.636	0.651	0.611	0.548
51	Determine the minimum set of indicators required and the most effective monitoring system	0.825	0.298	0.609	0.535	0.544	0.608	0.547
52	Evaluate the sequencing and linking of different community level interventions	0.696	0.385	0.610	0.479	0.590	0.591	0.536
53	Evaluate different stages of community engagement (consultation, cooperation, co-learning, collective action), including their phasing, cost and effectiveness	0.816	0.267	0.663	0.453	0.548	0.587	0.518
54	Evaluate ways to ensure delayed cord clamping in deliveries assisted by private providers	0.933	0.278	0.478	0.343	0.616	0.573	0.532
55	Assess the optimal number of community groups that a community engagement facilitator can support	0.923	0.208	0.471	0.365	0.611	0.562	0.497

Table 4: 11 research questions with a 10% difference in ranking between implementers and researchers

Rank	Proposed Research Question	Difference Answerable?	Difference Burden Reduction?	Difference Scale up?	Difference National policy?	Difference Ownership ?	Difference RPS	Difference AEA
37	Assess what communities consider as maternal-newborn health priorities and how communities compare maternal-newborn health with other development priorities	0.131	0.291	0.184	-0.030	0.099	0.135	0.650
22	Evaluate ways to improve retention of CHWs	-0.038	0.140	0.206	0.218	0.032	0.111	0.720
35	Evaluate different training approaches (including refresher training) for CHWs and their supervisors	-0.027	0.293	0.074	0.211	0.001	0.111	0.657
19	Evaluate ways to assure continuous supply of essential medicines and inputs for CHWs	0.083	0.265	0.162	0.135	0.028	0.135	0.751
36	Evaluate methods to prevent misuse of oxytocics	-0.101	-0.136	-0.092	-0.112	-0.112	-0.111	0.653
31	Determine culturally appropriate means to deliver skin to skin care (formative research of the cultural barriers, design of local solutions)	0.095	0.217	0.228	0.200	0.005	0.149	0.677
15	Assess the impact of initiation and continuation of Kangaroo Mother Care at home on survival of preterm/LBW babies in setting with high home births	0.022	0.105	0.237	0.154	-0.020	0.100	0.739
13	Evaluate demand-side financing mechanisms (e.g. insurance, demand side subsidies, vouchers)	0.106	0.167	0.165	0.152	-0.021	0.114	0.764
53	Determine the minimum set of indicators required and the most effective monitoring system	0.197	0.182	0.186	0.070	0.021	0.131	0.547
44	Measure the extent of household expenditures and their equity impacts	0.053	0.175	0.370	-0.010	0.137	0.145	0.609
51	Evaluate methods and levels of accountability that can be ensured	0.162	0.245	0.149	0.145	0.034	0.147	0.540

Box: CHNRI Process

Stage 1: Defining the research context, questions and criteria for priority setting

When: May-September 2009

How: Group discussions and subsequent e-mails

Results:

- Consensus on research context defined by space (developing countries), time (the next 5-10 years), the population of interest (children under five years of age) and disease burden of interest (preterm and stillbirths). Respondents were also asked to keep in mind that all research questions started with the following introduction: "*When implementing a community based maternal newborn intervention package that addresses prematurity and stillbirths in different contexts at scale…*
- Consensus around 55 implementation research questions grouped according to the following research domains: community engagement, behavioural skills and practices, community health workers, rational drug use, management health systems and referral.
- Consensus on the 5 criteria used to rank the research questions: ethical answerability, disease burden reduction, ability to support scale up, likelihood to attract financial and policy support, ownership by local actors.

Stage 2: Enlisting experts to systematically score the research questions

When: October 2009 – March 2010

How: Preliminary emails sent to 85 leading experts on community based approaches and maternal-newborn health in developing countries identified through a literature search and through snowballing of program managers. The spreadsheet was also translated into French and Spanish in order to ensure the participation of colleagues from Francophone Africa and Latin America.

Results:

- 42 experts agreed to participate
- 31 experts were able to complete the spreadsheets independently scoring the 55 research questions by each of the five criteria by answering 'Yes' (1 point), 'No' (0 points), undecided (0.5 points) or insufficiently informed to answer the question (missing input).

Stage 3: Computing and writing up results

When: March –August 2010

How: An intermediate score for each of the five criteria was calculated and the overall research priority score (RPS) computed as the mean of all five intermediate priority scores **(8-10) (Supplementary Table S3)**. Average Expert Agreement scores were computed for each research question as the average proportion of scorers that agreed on the 55 questions asked (**Supplementary Table S1)**.

Results:

- 29 correctly completed spreadsheets analysed with all 55 research questions systematically scored and ranked in order of priority and agreement.
- Draft circulated to all participants for feedback before being finalized.

Supplementary Table S1. The CHNRI methodology for setting priorities in health research investments.

STAGE 1: Defining the context and criteria for priority setting

Specifying the context a priori is a critical part of the CHNRI process, because priority scores for many research investment options may change substantially according to different contexts. According to CHNRI guidelines (**10**), the implementation research context was defined by space (developing countries), time (the next 5-10 years), the population of interest (children under five years of age) and disease burden of interest (preterm births and stillbirths). Respondents were asked to in particular keep in mind that all research questions were framed by the following introductory phrase: "*When implementing a community based maternal newborn intervention package that addresses prematurity and stillbirths in different contexts at scale…*".

The core working group also adapted the five criteria to be used to score the research questions. The standard version of CHNRI methodology uses the set of five criteria, but they can be modified or changed in different contexts – in this case, to fit the context of the implementation research. The working group agreed to retain two of the standard CHNRI criteria: (i) answerability of the research question in an ethical way; and (ii) the potential of proposed research to reduce the existing disease burden (due to prematurity and stillbirths). Three standard CHNRI criteria were discarded: (iii) likelihood of effectiveness; (iv) likelihood of deliverability; and (v) predicted impact on equity. The group felt that those three criteria, as defined in standard CHNRI framework, were not sufficiently discriminatory or appropriate for implementation research questions for community based approaches. Instead, three new criteria were developed to replace them: (iii) likelihood that the proposed research would address program gaps for scaling up; (iv) likelihood to attract funding support and national policy attention; and (v) likelihood that the research results would be owned by local actors, including political authorities and elected representatives, health workers, district managers and communities. All community CHNRI group members then validated the criteria and finalized 55 research questions.

STAGE 2: Choice of technical experts, systematic listing and scoring of research investment options

The first task of the core group of technical experts was to propose a large spectrum of research questions in a systematic way, according to the framework developed by CHNRI (supplementary **Tables S3, S4**). The conceptual framework for this process is described in detail elsewhere (**8-11**).

While a list of primarily biomedical research questions had been proposed to the group at the Seattle conference, the group felt that vital health systems concerns pertinent to how interventions are operationalised, like the management of community health workers and health systems supports for community level interventions, were not adequately reflected. We kept some of the behavioural research questions and in addition proposed new implementation research questions that fit within the following research avenues that were deemed of critical interest: (i) community engagement, (ii) community health workers, (iii) rational drug use, (iv) community level interventions (primarily behavioural), (v) referral and (vi) management/ health systems issues. The final list of 55 research questions were reviewed for potential gaps and finalized by the expert group through face to face consultations and over e-mail.

The finalized CHNRI score spreadsheet was then sent via email to 85 leading experts on community based approaches and maternal-newborn health in developing countries. They were identified through a preliminary literature search and through snowballing of program managers. Every effort was made to invite a mix of people with different backgrounds (clinicians, epidemiologists, public health experts, and programme managers) and from different countries (both developed and developing ones), so that the mix contains a diversity of views. While the exercise was on research priorities, because it focused on implementation research, special efforts were made to include the perspectives of those in charge of implementing programs. The exercise was translated into French and Spanish in order to ensure the

participation of colleagues from Francophone Africa and Latin America. Out of 85 experts engaged in research and implementation of maternal newborn programs, 42 expressed interest in contributing to this research prioritization exercise. The profile of responders and non-responders is presented in the **Supplementary Table S2**.

The co-ordinator of the project for GAPPS (AG) then invited the 42 experts with interest in implementation research to submit their scores. Every expert scored all 5 criteria, thus limiting potential impact of any single expert on overall scores. Out of 42 experts who initially agreed to take part in the exercise, 31 responded with a full list of research questions scored according to CHNRI criteria (74% response rate). Two spreadsheets had to be discarded due to errors in filling out the spreadsheet, so a total of 29 responses were analyzed. Respondents were fairly even in terms of gender (39% women) and diverse in terms of regional representation (26% sub-Saharan Africa, 16% Asia, 16% Latin America, 10% Europe, 32% North America). While a substantial number of respondents were based in North America, they were all engaged full-time on working in developing country contexts. Half of respondents were based in academic research institutions, whereas the other half represented those in charge of implementing programs whether through non-governmental organizations, UNICEF country offices or USAID headquarters. Non-respondents were not significantly different from respondents (see **Supplementary Table 2**). The process was conducted and completed via e-mail between October 2009 and March 2010. Further information on methods related to this part of the priority-setting process were presented elsewhere in greater detail (**17-19**).

STAGE 3: Computations of "research priority scores"

All the experts answered the questions listed in Box 1 by 'Yes' (1 point) or 'No' (0 points). They were also allowed to declare an informed but undecided answer (0.5 points) or declare themselves insufficiently informed to answer the question (missing input). Thus, the proposed research questions got a score for each of the five criteria as "the proportion of maximum possible points scored when an answer was given" (i.e., excluding the missing input). Each of the 55 listed research questions received five intermediate scores (each ranging between 0-100%), which were then multiplied by 100. CHNRI methodology allows for weighting of the intermediate scores based on the input from the external group of stakeholders. However, in this exercise the weights were not applied because it was not possible to appropriately define a relevant group of stakeholders for all contexts to which this exercise could potentially apply. The overall research priority score (RPS) was then computed as the unweighted mean of all five intermediate priority scores. The exact scores given to all 55 research questions from individual experts are presented in supplementary **Table S3**. The final list of priorities with intermediate and final priority scores for all 55 proposed research questions is presented in supplementary **Table S4**.

Assessment of agreement between scorers

CHNRI methodology has the ability to expose the issues of greatest agreement and controversy. This allows more focused discussion among experts following this exercise, and informs the investors and policy makers about the amount of controversy that surrounds each research question. The datasets that CHNRI methodology produces are not appropriate for application of the usual Kappa agreement statistics as discussed in detail elsewhere [**18,19**].

For each evaluated research investment option, AEA is informing us, for an average question, what proportion of scorers gave the same most frequent answer. This parameter accounts for missing answers, is unaffected by responses of 'undecided', and is also unaffected by the varying number of scorers per criterion and differences in scorer composition for the different criteria.

Advantages and limitations of the CHNRI methodology

The applied CHNRI methodology allows for the systematic listing and scoring of a large number of specific research questions. Other advantages of the CHNRI process include its well defined (a priori) context and criteria chosen for discriminating between research investment options, a highly structured way in which relevant information is obtained from the scorers, independent scoring that limits influence

of strong-minded individuals on the rest of the scorers, and ability to expose points of greatest agreement and controversy.

Although the advantages mentioned above represent attempts to deal with many issues inherent to research priority setting, there are still some potential biases. One of them is related to the fact many possible good ideas ("research investment options") may not have been included in the initial list of research options that was scored by the experts, and to the potential bias towards items that get the greatest press coverage. The spectrum of research investment options listed initially in this exercise was derived through a systematic process, but it is not endless and it cannot ever cover every single research idea. Therefore, the CHNRI process aims to achieve reasonable coverage of the spectrum of possible ideas. After the completion of the exercise, approximate scores and ranks for some specific research questions that are missing in the initial systematic list could still be estimated – either by relating them to the most similar questions on the list or by having those missed questions scored by a single expert (or by a group), using the CHNRI framework and then comparing the computed score to all other scores received for the originally listed research options.
Another concern over the CHNRI process is that its end product represents a possibly biased opinion of the involved group. We tried to balance the group with experts tasked with both research and implementation responsibilities.

Validation of CHNRI methodology

CHNRI methodology combines two ideas:

(i)"Principal component analysis" – a statistical technique which reduces a very complex system of large number of variables to a small number of relatively independent "principal components" which still capture a sizeable proportion of variation in the system; by defining a set of 5 "criteria", CHNRI process effectively reduces a notoriously complex and multi-dimensional task of priority setting, which could be approached through an almost infinite number of "lenses", into an exercise where the 5 most important (and reasonably independent) criteria for priority setting are clearly defined. They can even be weighted afterwards, in order of their importance to the users.

(ii) "Wisdom of the crowds" – this refers to the process of taking into account the collective opinion of a group of individuals rather than a single expert (or small number of experts) to answer a question, because it has been shown that the average of collective guesses are nearly always closer to the truth than any expert judgement. The pre-requisites for this process to work are: (i) Diversity of opinion (each person should have private information); (ii) Independence (people's opinions aren't determined by the opinions of those around them); (iii) Decentralization (people are able to specialize and draw on local knowledge); and (iv) Aggregation (some mechanism exists for turning private judgments into a collective decision – in this case, the CHNRI method).

The validation of CHNRI method based on the exercises conducted to date showed: (i) stability (correlation coefficients of over 90%) of scores given to same questions by the same experts in different points in time; (ii) almost identical scores of the same question scored by a larger group multiple times (score always falls within +1.7 points on a scale 0-100); and (iii) Monte Carlo simulations in random sub-samples of the larger group of scorers showed that the probability that the outcomes of the exercise could be substantially different if another group of experts conducted the scoring becomes incredibly small as soon as each criterion is scored by more than 17-23 rational persons with some knowledge of the problem; (iv) change of the context of the exercise leads the same group of experts to assign significantly different scores to the same research questions.

In comparison to other methods for setting priorities, in "expert panel"-type processes one very loud vote has a potential to heavily bias the process. During the GAPPS conference nine working groups defined priorities using Delphi-type processes, while three working groups used the CHNRI method. At the end of the conference, the rapporteurs from Delphi groups realised that it is not possible to have a discussion on all possible research options and keep in mind all their pros and cons all the time.

Eventually, the group leaders ended up forwarding the ideas which they originally brought to the table and gained support for them from the rest of the group. This did not happen in the CHNRI group.

Supplementary Table S2: Profile of respondents and non-respondents

		Gender		Region					Organisational Base		Total
		Women	Men	Sub-Saharan Africa	Asia	Latin America	Europe	USA	Research (University, WHO)	Implementing NGO, UNICEF, USAID	
Respondents	Numbers	12	19	8	5	4	3	11	15	16	31
Non-respondents	Numbers	3	8	4	2	0	1	4	2	9	11
Total		15	27	12	7	4	4	15	17	25	42

Countries where respondents were based:

Bangladesh, India, Pakistan, Thailand

Burundi, Ethiopia, Niger, Malawi, Senegal, Zambia

Brazil, Argentina, Columbia

Sweden, Switzerland, UK, USA

Countries were non-respondents were based:

India, Nepal

Burkina Faso, Senegal, Kenya, Uganda

Denmark, USA

Supplementary Table 3: All 55 Implementation Research Questions Scored and Ranked

RANK	RESEARCH OPTION	Answerable?	Burden reduct?	Scale-up?	Nat'l policy?	Ownership?	RPS	Difference implementers vs. researchers
1	Evaluate ways to reduce the financial barriers to facility births at the community level (user fee exemptions, emergency loans, conditional cash transfers, transportation vouchers, etc)	0.930	0.663	0.845	0.877	0.895	0.858	0.135
2	Develop and validate strategies to identify preterm babies at community level by CHWs and family members	0.942	0.640	0.750	0.795	0.821	0.832	-0.040
3	Evaluate different methods of behaviour change that overcome harmful practices and promote positive cultural and social norms	0.904	0.696	0.909	0.886	0.772	0.829	-0.065
4	Evaluate effective community-based strategies to reach the poor and marginalized	0.895	0.670	0.843	0.911	0.868	0.825	0.097
5	Evaluate ways to measure and maintain quality of care provided by CHWs	0.967	0.698	0.851	0.737	0.776	0.825	-0.050
6	Evaluate ways to provide thermal care and feeding for the preterm baby	0.958	0.686	0.802	0.737	0.798	0.822	0.041
7	Evaluate financing measures at the community level that improve referral	0.915	0.500	0.848	0.729	0.877	0.817	0.064
8	Evaluate ways to motivate and compensate CHWs and their supervisors	0.983	0.596	0.929	0.700	0.817	0.814	-0.062
9	Evaluate how to maximize referral compliance especially for the poor and marginalized	0.959	0.587	0.796	0.772	0.833	0.813	0.093
10	Evaluate ways to engage communities in birth planning for normal and at risk pregnancies	0.908	0.630	0.740	0.741	0.888	0.812	0.069
11	Evaluate demand-side financing mechanisms (e.g. insurance, demand side subsidies, vouchers)	0.895	0.512	0.840	0.886	0.851	0.805	0.073

12	Evaluate how community audits could improve access and quality of services	0.936	0.534	0.821	0.731	0.768	0.804	0.025
13	Assess the impact of initiation and continuation of Kangaroo Mother Care at home on survival of preterm/LBW babies in setting with high home births	0.907	0.660	0.683	0.806	0.694	0.801	0.025
14	Evaluate how community engagement improves referral and counter-referral	0.925	0.510	0.891	0.678	0.746	0.797	-0.019
15	Evaluate ways to ensure the sustained use of ITNs by pregnant women and newborns	0.974	0.533	0.765	0.809	0.786	0.796	0.002
16	Evaluate how CHWs can improve referral and counter-referral	0.958	0.531	0.782	0.681	0.750	0.795	0.031
17	Evaluate ways to assure continuous supply of essential medicines and inputs for CHWs	0.975	0.558	0.718	0.612	0.847	0.791	0.111
18	Evaluate the barriers at the community and provider level that cause poor referral	0.975	0.521	0.741	0.741	0.793	0.789	0.042
19	Evaluate ways to garner community support to ensure early and sustained breastfeeding	0.921	0.696	0.800	0.686	0.781	0.775	0.111
20	Evaluate ways to improve retention of CHWs	0.967	0.529	0.786	0.703	0.892	0.768	0.135
21	Evaluate how to measure good supervision for CHWs and different ways of providing it	0.950	0.547	0.821	0.667	0.742	0.761	0.068
22	Assess methods to ensure rational drug use among CHWs	0.930	0.520	0.722	0.736	0.728	0.756	0.107
23	Evaluate ways to maintain CHW neonatal resuscitation skills	0.908	0.635	0.740	0.777	0.705	0.752	0.118
24	Assess the optimal number of activities and population coverage required to maintain case load and skills of CHWs	0.917	0.592	0.845	0.603	0.741	0.752	0.099
25	Evaluate the equity impacts and effectiveness of CHW services when delivered with user fees or drug cost-recovery fees	0.825	0.349	0.704	0.754	0.860	0.740	0.026
26	Determine how CHWs can use injectable antibiotics for newborn sepsis safely and effectively	0.800	0.824	0.764	0.789	0.708	0.739	-0.111

27	Determine how to adapt and improve existing diets for malnourished pregnant women based on home available foods	0.925	0.500	0.588	0.691	0.704	0.737	-0.057
28	Evaluate different management structures for CHWs (community based, government based, private sector franchising, etc)	0.956	0.402	0.798	0.661	0.769	0.732	0.080
29	Determine culturally appropriate means to deliver skin to skin care (formative research of the cultural barriers, design of local solutions)	0.900	0.570	0.745	0.821	0.830	0.724	0.060
30	Assess the costs of individual interventions and combined packages of interventions	0.930	0.363	0.685	0.673	0.714	0.721	0.149
31	Determine how to overcome the cultural barriers for the adequate food intake of women during pregnancy in specific contexts like South Asia	0.917	0.431	0.594	0.683	0.676	0.717	0.091
32	Evaluate different ways of supporting facilitators of community engagement (training, supervision, skill maintenance, etc)	0.921	0.451	0.836	0.527	0.684	0.706	0.091
33	Evaluate different training approaches (including refresher training) for CHWs and their supervisors	0.942	0.470	0.781	0.647	0.629	0.705	0.063
34	Evaluate methods to prevent misuse of oxytocics	0.875	0.444	0.660	0.717	0.708	0.705	-0.073
35	Evaluate methods to overcome health professional resistance to CHWs prescribing and administering drugs (e.g. oral or injectable antibiotics for newborn sepsis, injectable vitamin K, etc)?	0.807	0.600	0.647	0.640	0.632	0.684	0.088
36	Evaluate current career pathways for CHWs and methods for improving their prospects	0.917	0.311	0.688	0.658	0.763	0.682	0.057
37	Assess what communities consider as maternal-newborn health priorities and how communities compare maternal-newborn health with other development priorities	0.877	0.290	0.627	0.447	0.772	0.672	0.100
38	Evaluate various forms of community engagement (village health committees, mothers groups, working with religious leaders, community theatre and songs, etc)	0.868	0.302	0.736	0.536	0.741	0.662	0.000
39	Evaluate the different methods of selecting CHWs (community vs. competency based, etc)	0.883	0.406	0.625	0.575	0.758	0.659	-0.044
40	Assess methods to ensure community awareness and practice of rational drug use	0.868	0.388	0.673	0.585	0.759	0.659	-0.033

41	Assess the gender and other equity dimensions of community engagement	0.813	0.330	0.690	0.670	0.589	0.656	-0.058
42	Measure the extent of household expenditures and their equity impacts	0.891	0.226	0.433	0.613	0.704	0.656	-0.018
43	Evaluate different CHW labour arrangements (unionisation, levels of formalisation -volunteers vs. paid, etc)	0.888	0.363	0.675	0.583	0.708	0.653	0.018
44	Evaluate the impact on preventive and promotive aspects of community level interventions, when curative interventions are introduced into the package of services	0.818	0.489	0.604	0.667	0.660	0.651	0.067
45	Evaluate ways in which communities are involved in monitoring and evaluation	0.895	0.367	0.704	0.509	0.763	0.647	0.099
46	Assess the gender distribution of CHWs and its implications in terms of their acceptability and effectiveness	0.925	0.343	0.602	0.619	0.633	0.639	0.114
47	Assess how CHWs and other kinds of frontline health workers are represented in human resource policies, strategies and legislation	0.925	0.271	0.556	0.583	0.692	0.638	0.092
48	Evaluate methods of integrating community-based data collection into district HMIS	0.930	0.298	0.636	0.526	0.579	0.628	0.131
49	Evaluate methods and levels of accountability that can be ensured	0.650	0.345	0.565	0.510	0.608	0.618	0.082
50	Assess the methods of tracking budget allocations and flow	0.889	0.256	0.482	0.636	0.651	0.611	0.062
51	Determine the minimum set of indicators required and the most effective monitoring system	0.825	0.298	0.609	0.535	0.544	0.608	0.082
52	Evaluate the sequencing and linking of different community level interventions	0.696	0.385	0.610	0.479	0.590	0.591	0.055
53	Evaluate different stages of community engagement (consultation, cooperation, co-learning, collective action), including their phasing, cost and effectiveness	0.816	0.267	0.663	0.453	0.548	0.587	0.145
54	Evaluate ways to ensure delayed cord clamping in deliveries assisted by private providers	0.933	0.278	0.478	0.343	0.616	0.573	0.147
55	Assess the optimal number of community groups that a community engagement facilitor can support	0.923	0.208	0.471	0.365	0.611	0.562	0.013