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Setting global research priorities for developmental disabilities, including intellectual disabilities and autism

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Abstract

Objectives—The prevalence of intellectual disabilities (ID) has been estimated at 10.4/1000 worldwide with higher rates among children and adolescents in lower income countries. The objective of this paper is to address research priorities for development disabilities, notably intellectual disabilities and autism, at the global level and to propose the more rational use of scarce funds in addressing this under-investigated area.

Methods—An expert group was identified and invited to systematically list and score research questions. They applied the priority setting methodology of the Child Health and Nutrition Research Initiative (CHNRI) to generate research questions and to evaluate them using a set of five criteria: answerability, feasibility, applicability and impact, support within the context and equity.

Findings—The results of this process clearly indicated that the important priorities for future research related to the need for effective and efficient approaches to early intervention,

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Conflict of Interest
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empowerment of families supporting a person with developmental disability and to address preventable causes of poor health in people with ID and autism.

Conclusions—For the public health and other systems to become more effective in delivering appropriate support to persons with developmental disabilities, greater (and more targeted) investment in research is required to produce evidence of what works consistent with international human rights standards.

Keywords

autism; developmental disabilities; global research priority setting; intellectual disabilities; low and middle income countries

Introduction

In recent years, WHO has expanded working in the area of ID[a] and autism through extending partnerships at the global level with key international stakeholders. Mapping of policy and service provision for people with ID has been completed in 147 countries (WHO, 2007a) and both ID and autism have been included in WHO's flagship mental health Gap Action Programme (mhGAP) (WHO, 2008, WHO, 2010). However, the more WHO has been determined to work in this area, the more the dearth of scientific knowledge that has relevance to the situation of people with ID and autism in low and middle income countries (LMIC) has become obvious (Emerson et al., 2007, Emerson et al., 2012, WHO, 2011). One of WHO's core functions is to shape the research agenda and stimulate the generation, translation and dissemination of knowledge (WHO, 2007b). Developing a global research agenda for ID and autism could help to guide research activities towards priority areas.

Existing epidemiological research suggests that these developmental disabilities contribute to a high amount of disability worldwide. The prevalence of ID has been estimated at 10.4/1000 worldwide with higher rates among children and adolescents and in LMIC's (Maulik et al., 2011). The median prevalence for pervasive developmental disorders (which include autism and Asperger's syndrome) has been estimated at 6.2/1000 (Elsabbagh et al., 2012) with no difference by geographic region. We need to know more about the prevalence, magnitude, impact, causes and correlates of these disabilities.

It is clear that people with disabilities, including people with ID and autism, face extensive and widespread discrimination and violations of their human rights and fundamental freedoms (United Nations, 2003, United Nations, 2006, United Nations, 2011, World Health Organization and the World Bank, 2011). They also, partly as a result of these violations, are vulnerable to experiencing extensive health inequalities (Ouellette-Kuntz, 2005). According to WHO's Atlas of global resources for persons with ID (World Health Organization, 2007); most people with ID do not receive the services and supports they require. The global gap in services is accompanied and, in part, attributable to the absence of relevant scientific knowledge. At present, we simply do not know what the most cost effective services are and which services models are applicable in different contexts.

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[a] ID' is synonymous with the outdated and to many offensive ICD-10 term 'mental retardation'

Research can make a significant contribution to improving the well-being of people with disabilities and to reducing the health inequalities faced by 'vulnerable' groups (World Health Organization, 2008, World Health Organization and the World Bank, 2011, World Health Organization, 2011). However, most existing scientific knowledge is of 'indirect relevance' to people living with developmental disabilities in LMIC and has mostly been conducted in English speaking high income countries (Emerson et al., 2007) a pattern of inequity that is repeated across many areas of health research (Global Forum for Health Research, 2004). Much of the available knowledge is also insensitive to issues of gender, culture and ethnicity (Yasamy et al., 2011). There is a dual need for increased research in LMIC countries, as well as more translational and implementation research to establish the extent to which the existing knowledge base from high income countries has relevance for LMIC. It is safe to say that current research in rich countries is not directly contributing to improving the situation in LMIC. Establishing global research priorities will help identify the appropriate balance between basic science, clinical research and public health research; a dire need in the area of ID and autism.

Methodology

The Child Health and Nutrition Research Initiative (CHNRI) methodology for setting priorities in health research investments was used to identify global research priorities for ID and autism (Rudan et al., 2008). This methodology was adopted because: (a) it is a carefully developed and documented conceptual framework available in the public domain; (b) it has demonstrated usefulness in several previous exercises (including disability and mental health); and (c) it has expanding use by policy makers, large donors and international organizations (Tomlinson et al., 2009b, Tomlinson et al., 2009a, Tomlinson et al., 2007, Rudan et al., 2010). The method also has the advantage of supporting the participation of a range of stakeholders including those concerned with policy, with technical aspects of research, and with social and community issues. The CHNRI process is described below.

1. The first step in the process was to gather a core group who oversaw the process. The authors of this paper formed the core group and provided technical input.
2. The core group defined the context in order to ensure that recruitment of experts to participate in the priority setting exercise was representative (equitable). The following principles guided the process.
    a. Geographical focus; that there was adequate representation from different regions of the world including high income as well as LMIC.
    b. Gender representation: that gender equity was an important consideration
    c. Researcher/NGO interface: that the process was not dominated by academics/researchers and that there was representation from consumer organizations, civil society, NGOs, community based organizations, human rights groups and organizations of persons with disabilities and their families.

   d. Problem focus; the exercise will focus both on research related to mental disorders and psychosocial wellbeing/human rights of people with intellectual and developmental disabilities.

3. There are many possible criteria against which research options can be judged. The CHNRI methodology recommends the use of a limited number in order to ensure feasibility. Five criteria were identified by the core group based on a review of general research domains (see Box 1 for an outline of the specific questions for each criteria). The criteria were: (a) answerability; (b) feasibility; (c) applicability and impact; (d) support within the context; and (e) equity.

4. Library searches and snowball sampling was used to identify experts known for their work on development disabilities and representatives from civil society and service user organizations to independently generate research questions which they believed were priorities. Eligibility to participate to was based on meeting one of three criteria: (a) publication in the scientific literature on ID or autism; (b) being a consumer of ID or autism services; (c) being a member of an organization for persons with ID or autism. All participants fulfilled at least one of these criteria. This group of experts was then asked to generate no more than five research questions that they believed were a priority. This activity generated an initial list of 266 research questions.

5. Questions were collated into a composite set of questions by eliminating overlap. The collation process was undertaken by the core group. This yielded 69 questions. Questions were grouped into a number of sections in order to ease the scoring process. These were Basic Science and Epidemiology (11 questions); Prevention, Promotion, Social Organisation, Public Attitudes & Beliefs (14 questions); Services & Supports – General (18 questions); Services & Supports - Parents and Families (5 questions); Services & Supports - Identification/Screening & Early Intervention (5 questions); Services & Supports – Education (5 questions); Services & Supports - Access to Health (6 questions); Services & Supports – Ageing (2 questions); Services & Supports – Interventions (3 questions). We used the term ‘developmental disabilities’ to cover both ID and autism or other pervasive developmental disabilities. Participants were asked independently to rate each of the 69 questions on each of the five criteria.

6. Scores for competing research options were gathered and computed. In this way, the proposed options received a score on each of the five criteria ranging from 0–100%. They represent a measure of the collective opinion of the experts scoring independently. Surowiecki has shown how once each in individual in a rating process gets a right to express judgement that is treated equally as the judgement of any other individual, then the personal biases that those individuals bring into the process tend to cancel and dilute each other regardless who the participants are. What is left is information based on accumulated knowledge, lifetime experience and common sense of those who took part (Surowiecki, 2004).

Results

The library search and snowballing approach yielded a list of 138 possible members of the expert group, of these 72 (52%) agreed to participate. The expert group was geographically diverse, with 23% from Asia, 17% from Europe, 21% from North America, 12% from the Middle East, 9% from Africa, 9% from Latin America, 6% considering themselves international (WHO or international NGO's), and 4% from Australasia. Sixty three participating members of the expert group contributed potential research questions.

The final list of 69 research questions was sent to the original group of 72 experts. Scoring took place over a three month period and was performed by 49 members; 68% of the original working group. The experts who completed scoring had almost a similar profile to that of the original larger group. They were equally geographically diverse, with 18% from Asia, 16% from Europe, 16% from North America, 8% from the Middle East, 12% from Africa, 10% from Latin America, 12% considering themselves international (WHO or international NGO's), and 6% from Australasia. Experts were also asked to describe their area of expertise – in some cases they listed more than one. Primary areas of expertise were academic researcher (37%); health or allied health care professional (6%); basic scientist (4%); users (2%) (we define a user as a consumer of developmental disability, autism or ID services) non-governmental organization (29%); rehabilitation (4%); public health (14%) and education or special education (8%). In addition, 12% or respondents also stated that they were either the parent or immediate family member of a user.

The scoring of the 69 research questions resulted in the ranking of research topics based on the perceived likelihood that they would be answerable, feasible, have applicability and impact, obtain support or have an impact on equity. An overall research priority score (RPS) was calculated as the mean of each criteria score. Mean scores on the separate domains across the 69 items were all highly inter-correlated (Table 1), while Table 2 shows the mean RPS scores by research domain.

Following Collins et al (Collins et al., 2011) we grouped priority areas into five goals: advance identification, screening and early intervention; improve awareness, prevention, and promotive interventions; identify causes, prevalence, biomarkers, and risk and protective factors; transform health system to improve access and build human resource capacity; and improve support to parents and families. Illustrative research questions for each goal are presented in Table 3 (see Appendix 1 for the full ranking of all 69 research questions).

The goal of advancing early identification, screening and early intervention achieved particularly high scores across the expert group. In this regard, there was a focus on how health systems can improve in early detection of developmental disabilities and how to operationally define and identify people with developmental disabilities. Linked to this was services and support related to identification, screening and early intervention and included questions on the availability, cost and efficacy of early intervention and rehabilitation programs for children with developmental disabilities. In terms of improving awareness and prevention (Goal B) prominent areas of focus were on the rights to health and education of children together with a strong emphasis on stigmatization, and the attitudes of both primary health care workers as well as the broader societal attitudes towards the care of people with developmental disabilities and how these vary across regions and countries. Training emerged as a strong theme – whether of primary health workers to reduce discrimination or establish how to train non-specialist health workers (community health workers for instance) to work with people with developmental disabilities. Providing support to the parents and families of children and people with developmental disabilities emerged as a particularly strong theme with a number of research questions relating to services and support research such as the most efficient ways of supporting and empowering parents/families of people with developmental disabilities and to how they can be most efficiently supported to provide a ‘healthy start’ in life for young children with or at high risk of developmental disabilities. Finally, the priorities recognise that there is much still to be learned in terms of identifying causes, prevalence studies and determining risk and protective factors. Examples of this included questions about the most common preventable causes of poor health among people with developmental disabilities and how these vary across regions and countries (and within countries) as well as questions concerning the most common preventable causes of poor health, social exclusion and nutritional deficiencies amongst children and people with developmental disabilities.

Discussion

There is a dearth of scientific knowledge, other than that derived from basic science that has relevance to the situation of people with developmental disabilities in LMIC’s (Emerson et al., 2007, Emerson et al., 2008, Emerson et al., 2012, World Health Organization and the World Bank, 2011). We applied a proven method (CHNRI methodology) to identify global research priorities in this area (Rudan et al., 2008, Tomlinson et al., 2009b, Tomlinson et al., 2009a, Tomlinson et al., 2007, Rudan et al., 2010). The results of this process clearly indicated that the crucial priorities for future research related to the need for effective and efficient approaches to early intervention and to address preventable causes of poor health among children with ID and/or. Another goal of research in this area was improving support to parents and families, training of health workers and non-specialists, and a strong focus on improving awareness. The main strengths of this priority setting exercise were: (1) the use of a proven methodology (Rudan et al., 2008, Tomlinson et al., 2009b, Tomlinson et al., 2009a, Tomlinson et al., 2007, Rudan et al., 2010); (2) the involvement of a significant number of experts with good gender balance and geographical spread. The main limitations of the approach relate to validity of the CHNRI approach and potential sampling biases. While 63 experts participated generating the ‘research investment options’ that were rated, it is clearly impossible within such an exercise to attain comprehensive coverage of all possible research questions. As such, both the questions generated and the ratings allocated are likely to reflect any biases in the identification, sampling and participation of experts. The nature of the CHNRI methodology as applied in this instance would have limited the participation of experts who were not fluent in English and experts who have difficulty in manipulating complex information. These biases would be expected to have a particular impact in limiting the direct participation of ‘experts by experience’ (e.g., people with ID and autism, many family members). Nevertheless the project was successful in eliciting research questions from 63 experts and ratings from 49 experts from around the globe. The number of participants and the protection against potential bias provided by the CHNRI approach (e.g., by limiting interaction between participants) does reduce the probability that a similar group of experts would produce materially different results. As such, we believe that this research priority setting exercise provides an important contribution to establishing a global research agenda for ID and autism.

Finally, it is important to note that the priority setting exercise did not address the appropriateness of particular research methodologies or paradigms. As has been argued elsewhere, there is clearly a need to seek a balance between investment in basic science, clinical research, strategic research and action research (Yasamy et al., 2011). Agencies such as WHO also support countries in developing relevant policies and plans and providing technical assistance for implementing them. The present exercise has provided grounds for the sound orientation of further research and service development particularly in poor resource settings. Finally, while the focus of this exercise was on LMIC, the aim is to provide guidance in setting research priorities globally. Different countries will certainly have different needs and we are not arguing for the same research questions to be employed within each country. We are also not suggesting that research conducted in rich countries has no relevance (through careful translational work) to LMIC.

In the area of ID and autism, there is also a particular need to increase investment in participatory or emancipatory research (i.e., research that is either co-produced with or controlled by organisations run by and for people with ID and/or autism). The reasons for this are twofold. First, increasing investment in participatory or emancipatory research is consistent with obligations under the UN Convention on the Rights of Persons with Disabilities for States Parties and international organisations to promote the inclusion of people with disabilities in *all* aspects of society. Second, a growing body of participatory or emancipatory research has documented the particular benefits that ‘insider knowledge’ or ‘experts by experience’ may bring to the development of research questions, methods and interpretation and dissemination of results (World Health Organization and the World Bank, 2011, Walmsley, 2010, Ramcharan et al., 2004).

What was perhaps most striking about the results of the priority setting exercise was the emphasis placed on research questions relating to providing more effective and efficient support to children (specifically reflected in 9 of the top 25 priorities) and the importance of understanding/addressing the social exclusion, discrimination and disadvantage faced by people with ID and/or autism (specifically reflected in 11 of the top 25 priorities). In contrast, only three questions in the top 25 related to basic science and epidemiology (one of these being the identification of preventable causes of poor health). The prioritisation of these issues is consistent with two general trends in our understanding of disability and health. First, disability is increasingly being conceptualised as a human rights issue in that people with disabilities are at significantly increased risk of: (1) experiencing inequalities due to discrimination in health care, employment, education, and legal systems; (2) being subject to violations of their dignity; and (3) being denied autonomy (e.g., by being subjected to involuntary sterilization) (World Health Organization and the World Bank, 2011). Second, the growing emphasis within health policy over that last decade on addressing global, regional and national inequalities in health has drawn attention to the general importance of addressing the broader social determinants of health associated with inequalities in access to power, wealth, education and employment (World Health Organization, 2008, World Health Organization, 2011, Marmot and on behalf of the Commission on Social Determinants of Health, 2007). Within this context, a growing body of evidence has highlighted the importance of early child development in setting the scene for the emergence of health inequalities in later life (Irwin et al., 2007, Shonkoff, 2010, Walker et al., 2011). Taken together, these trends stress the importance of taking a broader public health approach to understanding and addressing the health and well-being of people with ID and/or autism; a perspective that is notably lacking in current research investment. Interestingly, health system research was found to be the common priority across all previous global prioritization exercises for mental health research (Yasamy et al., 2011). Interestingly, while many of the research priorities relate to LMIC many of the identified priorities apply equally to higher income countries.

Evidence generated by research is clearly necessary but not sufficient for a real change on the ground. For the public health and other systems to become more effective in delivering appropriate support to persons with developmental disabilities, existing health care and programmes need to be based on evidence of what works consistent with international human rights standards, accompanied by appropriate resource allocation (e.g. financial, training), and based on principles of accountability.

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Web Appendix 1: All Research question ranked by Total RPS

	Research question	**Domain**	**RPS**	**Criteria on Which Item Scored in Top 10%**
1	How can health systems improve in early detection (during infancy and early childhood) of developmental disabilities in low and middle income countries?	SS: EI	74.29	F, A&I, E, S
2	What is the availability, cost and efficacy of early intervention and rehabilitation programs for children with developmental disabilities in low and low-middle income countries?	SS: EI	73.10	Ans, E, S
3	What are the most efficient ways of supporting and empowering parents/ families of people with developmental disabilities (in specific social and cultural contexts)?	SS:PF	72.35	F, A&I
4	What is the effectiveness and efficiency of family-mediated early intervention on the future health and well-being of children with developmental disabilities in low resource settings?	SS:EI	72.11	A&I, E, S
5	What are the most effective ways of promoting the rights to health and education of children with developmental disabilities in low and low-middle income countries?	P	71.67	Ans, A&I, E
6	How can parents/families be most efficiently supported to provide a ‘healthy start’ in life for young children with or at high risk of developmental disabilities in low resource settings?	SS:PF	71.36	A&I, E
7	How can we best operationally define and identify (screen for) people with developmental disabilities?	SS:EI	70.78	F, S
8	What are the most common preventable causes of poor health among people with developmental disabilities? How do these vary across regions and countries (and within countries)?	BSE	69.29	Ans
9	What are the most important priorities for investigation/assessment for children with developmental disabilities in low resource rural settings?	SS:EI	67.59	S
10	What is the impact of global initiatives in disability (e.g. UNCRPD) on policies, laws and services for people with developmental disabilities?	P	67.55	
11	How do public attitudes to developmental disabilities and the care of people with developmental disabilities vary across cultural and language groups and across types of developmental disabilities?	P	67.14	Ans, F
12	What are the most important nutritional needs of children with developmental disabilities in low resource rural settings?	SS:H	67.04	E
13	What are the common experiences and attitudes of primary healthcare workers towards people with developmental disabilities?	P	66.94	Ans, F
14	How best can non-specialists be trained and supervised to work with people with developmental disabilities?	SS:G	65.51	
15	What are the nature and forms of stigmatization in different cultures and contexts (low, middle, high income countries; rural/urban)?	P	65.31	F

	Research question	Domain	RPS	Criteria on Which Item Scored in Top 10%
16	What are the most efficient interventions to support parents/families of children with disabilities to manage their own stress?	SS:PF	65.24	
17	What are the important preventive health measures and barriers to health care for people with ID and how successfully are these implemented?	SS:H	64.66	
18	What are the most efficient ways of training health care workers to reduce the discrimination faced by people with developmental disabilities in health care systems?	SS:H	64.39	
19	What is the prevalence of developmental disabilities? How does this vary with child and environmental characteristics?	BSE	64.18	Ans, S
20	How can employment opportunities for individuals with developmental disabilities and/or autism be increased? What is the impact of employment on the psychological well-being of the adult working (and their families)?	SS:G	63.95	
21	What are the most common preventable causes of developmental disabilities (e.g., neurotoxins, undernutrition, infections, poverty, poor education)? How do these vary across regions and countries (and within countries)?	BSE	63.57	S
22	What would be the most effective way of monitoring across countries the health, well-being and nature/quality of supports used by people with developmental disabilities?	SS:G	63.20	
23	What would be the most effective way of monitoring across countries the inclusion of people with developmental disabilities?	SS:G	62.86	A&I
24	How best can we scale up access to behaviour support practices, including positive behaviour support and minimise the use of restrictive interventions (e.g., chemical, mechanical and physical restraint)?	SS:I	62.86	
25	How can assistive devices (computer software, hand-held devices, touch screen computers) support people with developmental disabilities to learn and communicate?	SS:I	62.79	Ans
26	How can medical training best prepare doctors to work with people with developmental disabilities?	SS:H	62.35	
27	What is the utility and effectiveness of easy read and pictorial information provided to people with moderate and severe ID?	SS:G	62.28	
28	What can be learned from a systematic study of public health systems and health care policy across countries with regard to what has worked and what hasn’t with respect to prevention and intervention for developmental disabilities?	SS:G	62.11	
29	What are the most efficient health promotion models for people with developmental disabilities?	P	61.94	
30	What kind of teacher competencies, teacher education and teacher professional development are required to better support the inclusion and development of learners with developmental disabilities?	SS:E	61.77	
31	What are the most efficient strategies for preventing developmental disabilities (in specific social and cultural contexts)?	P	60.51	
32	What are the most common preventable causes of the social exclusion and reduced quality of life among people with developmental disabilities? How do these vary across regions and countries (and within countries)?	BSE	60.44	
33	How can the situation of people with developmental disabilities be moved up the policy agenda in low and middle income countries?	P	60.31	E
34	How can CBR programmes most efficiently include and intervene with children with or at high risk of developmental disabilities in low resource settings?	SS:G	60.17	
35	What are the most efficient ways of organizing services and supports for people with developmental disabilities?	SS:G	59.76	

	Research question	Domain	RPS	Criteria on Which Item Scored in Top 10%
36	How do public beliefs about the causes of developmental disabilities vary across cultural and language groups and across types of developmental disabilities?	P	59.66	F
37	What are the most efficient ways of delivering reproductive health services for people with developmental disabilities?	SS:H	59.52	
38	What are the most efficient ways of empowering and supporting people with developmental disabilities to control their own lives and supports?	SS:G	59.49	
39	How can we best structure inclusive communities to work and support people with developmental disabilities and their families to be included and overcome marginalization and loneliness?	P	59.18	
40	What is the impact of ‘upstream’ population-based child health and nutrition interventions (including poverty-reduction programmes) on the incidence and prevalence of developmental disabilities in poor resource countries?	SS:G	58.98	
41	How can we most effectively improve research capacity and knowledge transfer and exchange in low resource settings?	P	58.59	
42	How can we reduce the stigma of intellectual and developmental disabilities?	P	58.47	
43	What are the most common genetic and environmental causes of developmental disabilities? How do these vary across regions and countries (and within countries)?	BSE	58.47	
44	How does “family context” interact with the behavioural challenges posed by children with developmental disabilities to produce either positive or negative impact on parents’ mental health and well-being?	SS:PF	57.72	A&I
45	How can empirically-based research findings be disseminated within and among countries?	SS:G	57.59	
46	What are the most efficient ways of enhancing skills that enable individuals with developmental disabilities to participate fully in their communities?	SS:G	57.33	
47	What mix of factors including the level of inclusion are most predictive of children with developmental disabilities making progress in education and better achievements in life.	SS:E	54.93	
48	What are the most effective ways of translating policy into a strategic action for the inclusion of children with developmental disabilities in educational settings?	SS:E	54.69	
49	How does the day to day quality of life and experiences of people with developmental disabilities and their families in all regions compare over time to those of non-disabled people and families in their neighbourhood?	BSE	54.25	
50	What is the impact of aging and related correlates on the ability of people with developmental disabilities to function in society?	SS:A	54.22	
51	How do persons with developmental disabilities participate in their healthcare decisions?	SS:H	53.84	
52	What are the most efficient ways of maintaining intervention effects over long periods of time and across the lifespan?	SS:A	53.40	
53	What are the most efficient ways of supporting people with developmental disabilities to develop and maintain personal relationships, including friendship and intimate partnerships?	SS:G	53.03	
54	How can we best develop self-monitoring tools which can be used by local staff, family members and by disabled people themselves to assess the quality of services and supports to meet their needs and to take action accordingly?	SS:G	52.11	
55	What kind of political, legal and curricular frameworks are playing a positive role in the inclusion of learners with developmental disabilities?	SS:E	51.43	

	Research question	Domain	RPS	Criteria on Which Item Scored in Top 10%
56	What is the prevalence of challenging behaviour among people with developmental disabilities? How does this vary with child and environmental characteristics?	BSE	51.09	
57	How can supported decision making be facilitated in low and middle income countries with different cultures	P	51.05	
58	How can we map the capacity of current services to see whether they reflect current scientific knowledge and whether it is effective enough to support people with developmental disabilities to reach their potential to live an independent life?	SS:G	50.00	
59	What factors are most strongly predictive of people with ID working for pay, irrespective of disability, in a given country?	SS:G	49.25	
60	What are the issues feeding into making informed decisions concerning resource allocation for providing educational opportunities for people with developmental disabilities?	SS:E	44.56	
61	Why do funding organizations and governments not fund research in low and middle income countries on developmental disabilities?	P	43.78	
62	What cultural and communication training is required to ensure that those working in developing countries do so effectively?	SS:G	43.37	
63	How can we most effectively measure the ‘disablement’ of developmental disabilities, so that developmental disabilities can be included in global burden of disease ratings?	BSE	42.52	
64	What are the biomarkers for developmental disabilities like autism?	BSE	42.01	
65	What are the significant markers of social cognition and how can they be assessed effectively and efficiently at key developmental points?	BSE	39.22	
66	What is the impact of wars and war traumas on the incidence of developmental disabilities?	BSE	37.96	
67	How do the dynamic mechanisms in early and ongoing family processes that affect child and parent outcome vary across culture and ethnic identity?	SS:PF	37.93	
68	What are developmentally essential experiences (intervention, support etc.) that promote social cognition and social competence?	SS:G	36.50	
69	How can we identify treatment non-responders early in treatment?	SS:I	35.37	

Table 1

Correlation (Pearson) Between Means Domain Scores and Total Score across Items

	Feasibility	**Applicability & Impact**	**Support**	**Equity**	**Total**
Answerability	.759***	.653***	.807***	.618***	.877***
Feasibility		.742***	.638***	.647***	.873***
Applicability & Impact			.659***	.777***	.874***
Support				.686***	.870***
Equity					.861***

Key:

*** $p<0.001$

Table 2

Mean RPS by Research Domain

Domain	**Mean RPS**
Services and support: Identification, screening and early intervention	71.6
Services and support: Access to health	62.0
Services and support: Parents and families	60.9
Prevention, promotion, social organisation, public attitudes and belief	60.9
Services and support: General	56.5
Services and support: Ageing	53.8
Services and support: Interventions	53.7
Services and support: Education	53.5
Basic science and epidemiology	53.0

Table 3

Priority research areas

	Illustrative research questions
Goal A Advance identification, screening and early intervention	- How can health systems improve in early detection (during infancy and early childhood) of developmental disabilities in low and middle income countries? - What is the availability, cost and efficacy of early intervention and rehabilitation programs for children with developmental disabilities in low and low-middle income countries? - What is the effectiveness and efficiency of family-mediated early intervention on the future health and well-being of children with developmental disabilities in low resource settings? - How can we best operationally define and identify (screen for) people with developmental disabilities? - What are the most important priorities for investigation/assessment for children with developmental disabilities in low resource rural settings?
Goal B Improve awareness, prevention, and promotive interventions	- What are the most effective ways of promoting the rights to health and education of children with developmental disabilities in low and low-middle income countries? - What is the impact of global initiatives in disability (e.g. UNCRPD) on policies, laws and services for people with developmental disabilities? - How do public attitudes to developmental disabilities and the care of people with developmental disabilities vary across cultural and language groups and across types of developmental disabilities? - What are the common experiences and attitudes of primary healthcare workers towards people with developmental disabilities? - What are the nature and forms of stigmatization in different cultures and contexts (low, middle, high income countries; rural/urban)?
Goal C Identify causes, prevalence, biomarkers, and risk and protective factors	- What are the most common preventable causes of poor health among people with developmental disabilities? - What are the most important nutritional needs of children with developmental disabilities in low resource rural settings? - What are the most common preventable causes of developmental disabilities (e.g., neurotoxins, undernutrition, infections, poverty, poor education)? How do these vary across regions and countries (and within countries)? - What are the most common preventable causes of the social exclusion and reduced quality of life among people with developmental disabilities? How do these vary across regions and countries (and within countries)?
Goal D Transform health system to improve access and build human resource capacity	- How best can non-specialists be trained and supervised to work with people with developmental disabilities? - What are the important preventive health measures and barriers to health care for people with ID and how successfully are these implemented? - What are the most efficient ways of training health care workers to reduce the discrimination faced by people with developmental disabilities in health care systems? - How can assistive devices (computer software, hand-held devices, touch screen computers) support people with developmental disabilities to learn and communicate? - What can be learned from a systematic study of public health systems and health care policy across countries with regard to what has worked and what hasn’t with respect to prevention and intervention for developmental disabilities?

	Illustrative research questions
Goal E Improve support to parents and families	• What are the most efficient ways of supporting and empowering parents/families of people with developmental disabilities (in specific social and cultural contexts)? • How can parents/families be most efficiently supported to provide a ‘healthy start’ in life for young children with or at high risk of developmental disabilities in low resource settings? • What are the most efficient interventions to support parents/families of children with disabilities to manage their own stress? • How does “family context” interact with the behavioural challenges posed by children with developmental disabilities to produce either positive or negative impact on parents’ mental health and well-being? • How do the dynamic mechanisms in early and ongoing family processes that affect child and parent outcome vary across culture and ethnic identity?

Box 1

Questions and scoring instructions

Scoring instructions - Firstly, always keep in mind the context. It is not possible to predict all outcomes of health research investments, and therefore the priorities should be set within a specific context, to influence a specific policy. Here, the context will be defined by space, time, population of interest. The space will be global, the time will be the next 5–10 years, and the area of interest on setting priorities for developmental disabilities including ID and developmental disabilities (including autism). Possible answers: Yes=1; No=0; Informed but undecided answer: 0.5; Not sufficiently informed: blank

CRITERION 1: ANSWERABILITY - Likelihood that the research question would be answerable and generate new knowledge

1 Would you say the research question is clear and has well defined endpoints?

2 Would you say that a study can be designed to answer the research question and to reach the proposed endpoints of the research?

3 Would you say that the research question would target a critical gap in knowledge, rather than just advance an already existing level of knowledge?

CRITERION 2: FEASIBILITY - Is the research potentially doable in the majority of countries in the world

1 Taking into account the level of difficulty with undertaking the research (e.g. the complexity of the research, the infrastructure required and human factors involved), would it be possible to undertake this research in most countries?

CRITERION 3: APPLICABILITY AND IMPACT - Likelihood that the knowledge generated through the proposed research would be implemented and have an impact on policy and practice

1 Taking into account the financial resources available to implement the research, would you say that the implementation of the end-points of the research would be affordable within the context of interest?

2 If basic science, is it likely that it will soon lead to significant improvement in practice?

3 Based on the best available evidence and knowledge, do you believe that interventions and programmes developed or improved through the proposed research would be effective?

CRITERION 4: SUPPORT WITHIN THE CONTEXT - Likelihood that the proposed research would, in the context of current policy and funding priorities, be likely to attract the necessary funding support.

1 Will the relevant public sectors such as health, social welfare and education or influential NGOs be supportive of the research in general?

2 Do you think the proposed research is likely to be prioritized for funding by agencies that fund research?

CRITERION 5: EQUITY - Likelihood that the proposed research would address underprivileged people, or provide solutions that would benefit people among all social strata equally.

1 Would you suggest that the proposed research would address primarily people that are among the poorest globally?

2 Would you say that the proposed research would provide solutions that would improve the quality of lives of persons regardless of their social status?