Validation of a home-based neurodevelopmental screening tool for under 2-year-old children in Bangladesh

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Abstract

Background Home-based screening to identify young children at risk for neurodevelopmental impairments (NDIs) is needed to guide the targeting of child neurodevelopmental intervention services in Bangladesh. This study aimed to validate such a tool for children under age 2 years. *Methods* A Developmental Screening Questionnaire was administered to mothers of children aged 0–<2 years in an urban community. Inter-rater reliability among the interviewers, who were high school graduates, was determined. All children who were screen positive and a proportion of screen negatives were subsequently assessed for NDIs by professionals. Sensitivity and specificity were calculated by comparing screening with assessment results.

Keywords

disability, frontline worker, impairment, neurodevelopment, screening

Correspondence: Naila Z. Khan, Department of Paediatric Neuroscience, Dhaka Shishu (Children's) Hospital, Sher-e-Bangla Nagar, Dhaka 1207, Bangladesh E-mail: naila.z.khan@gmail.com *Results* Mean kappa coefficient of agreement among interviewers was 0.95. A total of 197 children were screened, of whom 17% screened positive. Fifty-one children, including 24 screen negatives, were assessed for NDIs. Screen-positivity was significantly different between income groups (P = 0.019), and higher in stunted children (odds ratio = 5.76, 95% confidence interval = 1.72–19.28), indicating good discriminant validity Specificity was excellent (84–100%) for all developmental domains. Sensitivity was 100% for vision and hearing; 70% for speech; and 63%, 53%, 48%, and 45% for gross motor, behaviour, fine motor and cognitive impairments, respectively.

Conclusion A tool for screening <2-year-old children at risk for NDIs showed high specificity; and was able to identify all children at risk for vision and hearing impairments, nearly three-fourths with speech impairments, two-thirds with gross motor impairments, and about half with behavioural, cognitive and fine motor impairments. The Developmental Screening Questionnaire tool has potential for use by frontline workers to screen large populations and to link to definitive assessment as well as intervention services.

Introduction

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The estimated proportion of children at risk for disability in Bangladesh has risen more than twofold in the past two decades, from 8% in 1988 (Zaman *et al.* 1990) to 18% in 2005 (UNICEF 2008). Effectiveness of early detection and appropriate intervention is well established in developmental neurobiology (Shonkoff & Phillips 2000), and from our own experiences in working with cohorts of high-risk children (Khan *et al.* 2006). However, in a country where 20% of the estimated 156 million population is <2 years of age, and 85% of deliveries are conducted at home (UNICEF 2007), it is difficult to bring newborns, infants and very young children under an umbrella of developmental surveillance unless innovative and low-cost methods are developed.

The Ten Questions, an internationally developed tool (Durkin *et al.* 1994; Khan & Durkin 1995) most commonly used for home-based screening for childhood disability in low and middle income countries (Maulik & Darmstadt 2007; UNICEF 2008), is applicable only beyond 2 years of age. On the other hand, the International Classification of Function paradigm acknowledges the temporal relationship between early functional limitations, i.e. impairments, and their progression to longer-term limitations or disabilities (World Health Organization 2001); and programmes for early intervention demonstrate best outcomes when focused on impairments (Maulik & Darmstadt 2009). Screening tools for younger children at risk for impairments are, therefore, needed if disability prevention, optimum developmental outcomes and better quality of survival are to be achieved.

The objective of this study was to validate a screening tool for use by frontline workers (FWs) to identify under 2-year-old children at risk for neurodevelopmental impairments (NDIs) in resource-poor home settings.

Methods and materials

The Developmental Screening Questionnaire (DSQ)

The DSQ was developed in Bangladesh, and in keeping with the Ten Questions for older children (Durkin *et al.* 1994; Khan & Durkin 1995), was designed to be administered to mothers of children from birth to <2 years of age to screen their child's neurodevelopment. The questions focus on key skills at each month of age which reflect acknowledged developmental norms (Illingworth 1987; Rogers *et al.* 1997; Sheridan 1997; Bricker & Squires 1999; Oosterling *et al.* 2009; Khan *et al.* 2010). The child's age is determined in months. One question related to each of the following eight functional domains is asked: gross motor, fine motor, vision; hearing, cognition, socialization, behaviour and speech. A 'yes'/'no' format for each item is recorded in the pre-coded form. Table S1 lists the 24 age-sets, with eight questions per set, and 192 items in total, and some repetitions in the questions when deemed appropriate. At the end of the interview, any child positive on one or more functional domain is considered 'screen positive'.

Study design

A two-stage design, i.e. home-based screening by FWs followed by centre-based professional assessment - an internationally recognized approach to epidemiological studies of childhood disability first conducted in Bangladesh (Zaman et al. 1990) and other developing countries (Durkin et al. 1994) - was adapted. In stage one, FWs visited individual households within a given geographical area of Dhaka city where under 2-year-old children had been identified through a demographic survey. They had been issued identity cards with their photographs from the Bangladesh Protibondhi Foundation (BPF), co-signed by the local ward commissioners, and a letter of introduction which they presented (or read) to the mothers and explained that the survey was aimed at determining children's developmental status. After gaining the mother's consent, information on the household and mother's sociodemographic characteristics was collected, followed by interview of the mother (or the primary care providers) on the DSQ.

In stage two, all children for whom parents reported that they could not do one or more age-appropriate items, were considered 'screen positive' on the DSQ, and an equal number of age-matched 'screen negatives', were invited for comprehensive neurodevelopmental assessment by professionals (see below). The outcomes of the assessment were used as the Gold Standard to determine the validity of the DSQ.

Study site

The study was conducted in two urban wards within Dhaka city, the capital of Bangladesh: Mirpur (ward number 7) and Malibag (ward number 54). An initial door-to-door demographic survey was conducted to identify households (n = 197) with children under age 2 years. FWs subsequently visited these households to conduct interviews with the mothers.

Frontline workers

Six female FWs with a high school degree (i.e. Class 12 pass) were recruited for the study. They were provided a 1-week structured training which included tutorials on early child development, concepts incorporated within the International Classification of Function (World Health Organization 2001),

videos of community-based screening for childhood disability, and hands-on interviews with mothers using the screening instrument. Trainers were professionals, i.e. a child health physician and a developmental therapist.

Inter-rater reliability study

Before beginning field testing, 23 mothers of children aged 0–<2 years who were either siblings or neighbours of children attending the Kalyani Inclusive School of the BPF, were explained the purpose of the study (see above) and subsequently interviewed on the DSQ. A main trainee interviewer asked the mother the questions while other trainees simultaneously recorded the mothers' responses on the DSQ, without any consultation among themselves. Each trainee interviewed five mothers.

Gold standard: the Rapid Neurodevelopmental Assessment (RNDA) by professionals

All DSQ screen positive and an equal number of age- and gender-matched screen-negative children were invited to the Child Development Center of the BPF for an assessment. Four developmental therapists, who were blind to the DSQ screening status of the child, assessed the children within 2 weeks of screening by administering the RNDA instrument, i.e. a comprehensive assessment procedure validated against a *Bangla* version (Parveen *et al.* 2011) of the Bayley Scales for Infant Development (BSID II) (Bayley 1993) for children up to age 2 years (Khan *et al.* 2010). They rated the children's functional limitations in the following developmental domains: primitive reflexes (for <1-month-olds only), gross motor, fine motor, vision, hearing, speech, cognition, behaviour and seizures. Assessment results were recorded in a pre-coded Summary Sheet.

Analysis

Kappa coefficients of agreement (Fleiss 1981) were calculated by cross-tabulating the results of the main interviewer with five other trainees for 'screen positivity' on the DSQ. Mean kappa score was determined. Willingness of mothers to answer questions during home visits was considered to be a reflection of the face validity of the DSQ (Bryman & Cramer 1990). Effectiveness of the DSQ in discerning children from vulnerable populations, e.g. low income groups and stunted children (Grantham-McGregor *et al.* 2007), was considered the discriminant validity (Bryman & Cramer 1990).

Screen positivity on the DSQ was cross-tabulated with outcomes in the Summary Sheet of the RNDA to determine the sensitivity and specificity of the DSQ. The analysis considered all screened children (stage one) against those assessed (stage two), weighted for children who were screened but not assessed, as elaborated by Shrout and Newman (1989).

Ethical considerations

A written consent form was signed by literate mothers, while verbal consent was taken from those who were non-literate (12%, Table 1). Logistic support, i.e. travel costs and lunch, was provided to the families for attending the assessment session. All children who screened positive for an NDI were referred to the Child Development Center, Dhaka Shishu Hospital, and offered services appropriate to their condition.

The research protocol was approved by the Affairs of the NGO Bureau, Chief Advisor's Office, Government of Bangladesh (permission letter dated 23 December 2008; reference: ANB/Application-2/BPF/16-3/2008-1869).

Results

Inter-rater reliability of the DSQ

The mean kappa coefficient of agreement for DSQ outcomes on the screening of 23 children was 0.95, and ranged from very good to excellent (range: 0.86–1.00) between all interviewers.

Sociodemographic characteristics of household heads, mothers and children of the screened households

A total of 197 households were visited. One-fourth of the heads of households were unskilled workers and a third were in the lowest income group (Table 1). In total, 41.8% and 53.2% of household heads and mothers, respectively, had had minimal or no schooling. Almost the entire population had availability of electricity, piped water and sanitary toilets. Consanguinity was 4%. Mean numbers of children per family was 1.8. Of the 197 children screened, 17% were positive on the DSQ.

Discriminant validity

The highest proportions of children who screened positive were within the lowest and second-lowest income groups (Table 1), i.e. 4/9 (44%) and 15/50 (30%), respectively (Fig. 1), which was

Table 1.	Household,	maternal	and	child	characteristics	of the	e screened
populati	on $(n = 197)$						

Household characteristics (n = 197)	n (total = 197)	% of tota
Occupation of household head*		20.5
Service Business	56 53	28.5 27.0
Unskilled worker	41	20.9
Skilled worker	14	7.1
Others	32	16.3
Aonthly income of household (in taka)	9	16
<3 000 3 001 to 5 000	50	4.6 25.4
5 001 to 10 000	76	38.6
>10 000	62	31.4
Gender of household head		
Male	186	94.6
Female No information	10 1	5.1 0.5
ducation of household head*	1	0.5
No schooling	22	11.2
Class 1 to 5	60	30.6
Class 6 to 8	32	16.3
Class 9 to 10	28	14.3
Class 11 to 12 Bachelor's degree	15 18	7.7 9.2
Master's degree	21	10.7
ype of house*	21	10.7
Mud wall, thatched roof	1	0.5
Mud wall, tin roof	2	1.0
Bamboo wall, tin roof	1	0.5
Tin wall, tin roof	30	15.3
Brick wall, tin roof Brick wall, brick roof	107 16	54.6 8.2
Apartment	39	19.9
lectricity in house*	57	10.0
Yes	196	100
No	0	0
ource of drinking water*		27.0
Piped into house Piped into common yard	74 116	37.8 59.2
Public tap	5	2.6
Tube well	1	0.5
ype of toilet used*		
Sanitary	186	94.9
Bucket	4	2.0
Pit	6	3.1
Aaternal characteristics (n = 197)		
Aother's education No schooling	24	12.2
Class 1 to 5	72	36.5
Class 6 to 8	30	15.2
Class 9 to 10	30	15.2
Class 11 to 12	23	11.7
Bachelor's degree	8	4.1
Master's degree	10	5.1
an read newspaper	27	13.7
Not at all With difficulty	66	33.5
Easily	104	52.8
onsanguinity		
Present	9	4.6
	Mean	SD
umber of child deaths umber of children alive	1.10 1.87	0.296 1.005
haracteristics of screened children (n =		
ge in years	1.10	0.48
	n	%
ex Male	92	46.7
Female	105	53.3
creening status		
Negative	163	82.7
Positive	34	17.2
and the second have a second second second second	Mean (range)	SD
ge in years by screening status	1.09 (0.085, 1.00)	0.46
Negative Positive	1.08 (0.085–1.99) 1.20 (0.032–1.96)	0.46 0.52
i ositire	Number screened positive	Row %
ender distribution by screening status		
		22.0
Male (total = 92) Female (total = 105)	21 13	22.8 12.4

*Information about one household missing.

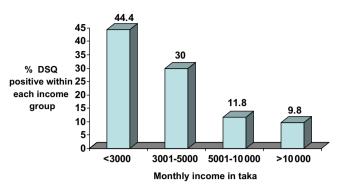


Figure 1. Proportion of Development Screening Questionnaire (DSQ) positive children within each monthly household income group (chi-square = 15.158, *P*-value = 0.019).

statistically significantly different than in the second-highest (11.8%) and highest (9.8%) income groups (chi-square = 15.158, *P*-value = 0.019).

In addition, the odds of having NDIs in the stunted children was high (odds ratio = 5.76, 95% confidence interval = 1.72-19.28).

Study population

In stage two, 51 children were assessed (Fig. 2); 27/34 were screen positive (79.4%) and 24/163 were screen negative (14.7%) in stage one. There was no significant difference in mean age in years [positives: age 1.19 years, standard deviation (SD) 0.53; negatives: age 1.21 years, SD 0.50; P = 0.861] and male : female ratio (positives: ratio 1.7; negatives: ratio 0.71; chi square = 2.313, P = 0.107) between screen positives and screen negatives. Mean age in years of the seven screen-positive children who defaulted in stage two was 1.26 (SD 0.54) years and male : female ratio was 1.3.

Specific NDIs identified in stage two

In all, 24/27 (88.9%) screen positives were found to have ≥ 1 NDI. The most common NDIs identified were for speech (n = 13, 48.1%) and cognition (n = 13, 48.1%), followed by gross motor (n = 9, 33.3%), behaviour (n = 6, 22.2%) and seizures (n = 6, 22.2%). Among the 24 screen-negative children assessed, five (20.8%; four boys, one girl) had ≥ 1 NDI relating to cognition (n = 3), gross motor (n = 1), fine motor (n = 1), speech (n = 1) and behaviour (n = 1) problems.

Sensitivity and specificity of the DSQ

Sensitivity was 100% for vision and hearing impairments and seizures; 71%, 63% and 53% for impairments in speech, gross

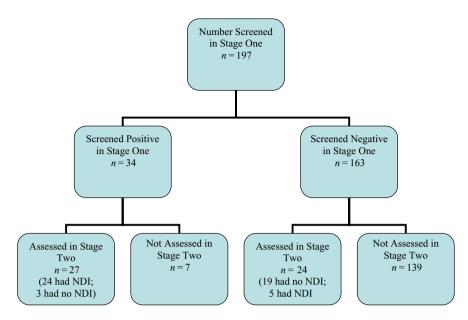


Figure 2. Total numbers of children screened in stage one, and assessed in stage two for any (≥ 1) neurodevelopmental impairment (NDI), by screening status.

 Table 2. Sensitivity and specificity of the Development Screening Questionnaire for screening 'any' and specific neurodevelopmental impairments*

Neurodevelopmental		95% CI			95% Cl	
impairment	Sensitivity (Se)	Lower	Upper	Specificity (Sp)	Lower	Upper
Any impairment	47.1%	36.5	57.6	97.2%	90.9	100.0
Gross motor	62.5%	46.6	78.3	87.3%	84.2	90.4
Fine motor	48.11%	35.0	61.1	84.9%	81.8	88.0
Vision	100.0%	91.0	100.0	83.8%	83.0	83.0
Hearing	100.0%	89.4	100.0	84.4%	84.3	84.3
Speech	70.7%	53.8	87.4	89.9%	86.7	93.0
Cognition	44.6%	27.7	61.3	89.0%	84.0	94.0
Behaviour	52.7%	38.6	66.6	85.5%	82.4	88.5
Seizures	100.0%	86.0	100.0	86.0%	86.0	86.0

*All calculations were weighted for the 139 children screened negative and seven children screened positive, but not assessed (Shrout & Newman 1989). See Fig. 2.

motor and behaviour, respectively; and 48% and 45% for fine motor and cognitive impairments, respectively (Table 2). Specificity ranged from 82% for fine motor impairments to 87% for speech impairments. Sensitivity and specificity was 47% and 97%, respectively, for 'any' (i.e. \geq 1) NDI.

Discussion

A home-based neurodevelopmental screening tool for newborns, infants and young children has been developed for use by lesser trained personnel in countries with limited resources. Several measures of test validity were verified in this study. Agreement between interviewers in the use of the DSQ was high, indicating that it can be administered reliably by FWs after a 1-week training course. This is important for Bangladesh, where primary health care services are provided to households through health assistants and family welfare assistants who have the same level of education as the study FWs (Human Resources for Health 2011). Moreover, other low and middle income countries utilize FWs with similar characteristics to deliver primary health care services (Bhutta *et al.* 2008), and the present tool has potential for use in those countries.

The tool had good face validity since mothers answered questions with active interest and there were no refusals. As 43% of mothers surveyed were first-time parents, it could be a useful approach for disseminating awareness regarding early neurodevelopment and its trajectories, which is often lacking even among parents in resource-rich countries (Bertrand *et al.* 2008; Rikhy *et al.* 2010).

Links between poverty, undernutrition and childhood disability have been well documented, both within Bangladesh (Islam *et al.* 1993; Durkin *et al.* 2000), other regional countries (Durkin & Hasan 1998) and internationally (Grantham-McGregor *et al.* 2007; Walker *et al.* 2007). Therefore, the ability of the DSQ to discriminate between income groups, and those chronically undernourished, adds to its potential for use in resource-poor countries.

Specificity of the DSQ was high overall, and for specific NDIs, so that health and disability services need not be over-burdened with large numbers of inappropriately referred children. This is a strength of the DSQ, if a tiered system of community-based screening by FWs using the DSQ and referral to secondary health care services for definitive professional assessment, e.g. using the RNDA (Khan *et al.* 2010) is to be developed.

Sensitivity of the DSQ was excellent for impairments of vision, hearing and seizures; good for speech and gross motor impairments; and fair (i.e. just about 50%) for behavioural, cognitive and fine motor impairments. Its overall sensitivity was 47% for \geq 1 NDI. In principle this implies a need to subsequently screen older children from high-risk populations in order to identify those with NDIs which early screening missed. The UK system of screening toddlers for NDIs is an example of such a universal practice (Department of Health, UK 2009).

There was a male preponderance for screen-positivity, as almost double the proportion of boys compared with girls screened positive in the surveyed population (i.e. 22.9% males vs. 12.4% females, Table 1). Male preponderance of poor neurodevelopment has been reported by us (Khan *et al.* 2006) and others (Wood *et al.* 2005), and the present finding could be part of that same trend. Paradoxically, four out of five of the false negatives (i.e. screened negative, but found to have NDIs) were boys, suggesting that under-recognition or under-reporting of potential neurodevelopmental problems in male children could be a social construct – including an element of denial related to family preference for male children (Gausia *et al.* 2009).

The study findings have far-reaching implications for the children of Bangladesh. Since 2008, the Ministry of Health and Family Welfare, Government of Bangladesh, has initiated the establishment of multidisciplinary (i.e. child health physician, child psychologist, developmental therapist as the core team) Child Development Centers (http://www.hsmdghs-bd.org/SBK.html) in every government medical college hospital across the country. These services, established with the objective of providing early diagnosis and intervention for children with a range of neurodevelopmental delays and disorders, are expected to link up with a universal system of developmental surveillance within communities, which is now possible with the availability of screening using the DSQ.

Study limitations must be considered. Only a small subpopulation of matched, screen negatives was assessed. The lesser degree of sensitivity of the DSQ to cognitive and fine motor impairments poses a matter of concern for use in its present format. Larger sample sizes, parental reassurance prior to asking the questions, and alternative (or additional) questions for cognition and fine motor skills could improve sensitivity. Formative research to better understand the reasons for, and to reduce, false negatives, may also aid in improving sensitivity of the DSQ. Any potential differences in sensitivity and specificity by age of the child could not be determined because of the small subsample sizes. Predictive validity of the DSQ, and its usefulness when linked with intervention strategies, also needs to be determined through prospective studies.

In conclusion, the DSQ is a new screening tool for identifying children at risk for NDIs early in life, i.e. before 2 years of age, and represents an important advance for large-scale programmes. The tool can be used reliably by FWs. Its high specificity suggests that despite its limitation in sensitivity, it could offer an important advance, particularly in low-resource settings where programmes to identify and intervene for NDIs are nascent. Further research to improve the sensitivity of the tool, particularly for detection of cognitive and behavioural impairments, is recommended.

Key messages

- A validated tool for home-based screening developed in Bangladesh for older children, i.e. the Ten Questions, is the most widely used and documented instrument worldwide; however, such a screening tool does not exist for under 2-year-old children.
- A home-based screening tool, the DSQ, has been fieldtested in a community setting for use in 0- to <2-year-old children and was found to have good reliability and validity.
- The tool was able to discern the large proportion of children who were unimpaired, thus allowing for focusing of time, resources and expertise on the smaller group who needed detailed neurodevelopmental assessment.

Conflict of interests

The authors have no financial disclosures or conflicts of interest to declare.

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Supporting Information

Additional Supporting Information may be found in the online version of this article:

Table S1. Development Screening Questionnaire (DSQ).

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