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Adaptation of the "ten questions" to screen for autism and other neurodevelopmental disorders in Uganda

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Abstract

Neurodevelopmental disorders are recognized to be relatively common in developing countries but little data exist for planning effective prevention and intervention strategies. In particular, data on autism spectrum disorders are lacking. For application in Uganda, we developed a 23-question screener (23Q) that includes the Ten Questions screener and additional questions on autism spectrum disorder behaviors. We then conducted household screening of 1169 children, 2–9 years of age, followed by clinical assessment of children who screened positive and a sample of those who screened negative to evaluate the validity of the screener. We found that 320 children (27% of the total) screened positive and 68 children received a clinical diagnosis of one or more moderate to severe neurodevelopmental disorders (autism spectrum disorder; cerebral palsy; epilepsy; cognitive, speech and language, hearing, or vision impairment), including 8 children with autism spectrum disorders. Prevalence and validity of the screener were evaluated under different statistical assumptions. Sensitivity of the 23Q ranged from 0.55 to 0.80 and prevalence for ≥1 neurodevelopmental disorders from 7.7/100 children to 12.8/100 children depending on which assumptions were used. The combination of screening positive on both autism spectrum disorders and Ten Questions items was modestly successful in identifying a subgroup of children at especially high risk of autism spectrum disorders. We recommend that autism spectrum disorders and related behavioral disorders be included in studies of neurodevelopmental disorders in low-resource settings to obtain essential data for planning local and global public health responses.

Keywords

autism spectrum disorder screening and assessment, developing countries, low- and middle-income countries, neurodevelopmental disorder screening and assessment, Uganda

Introduction

The World Health Organization (WHO) (2006) has identified neurological disorders, including neurodevelopmental disorders (NDDs), as one of the greatest challenges to improving global public health. NDDs are a diverse group of chronic conditions, often severe, that originate during neurologic development and typically persist throughout the lifetime. Etiology can be congenital (with genetic and nongenetic contributors) or acquired through trauma, infections commencing early in life, toxic exposures, nutritional deficiencies, and combinations of these factors (Durkin, 2002; Uganda Bureau of Statistics, 2007). Estimates of prevalence based on household screening range from 1.6/100 in Bangladesh (Zaman et al., 1990) to 6/100 in South Africa (Couper, 2002). With reductions in mortality in many low- and middle-income countries, a new population of children with developmental disorders may be emerging (Scherzer et al., 2012). Among the more common NDDs that often lead to severe impairment and result in considerable personal and public costs are autism spectrum disorders (ASDs), cerebral palsy (CP), epilepsy, cognitive impairment (CI),

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speech and language disorders, hearing impairment, and visual impairment.

Accurate data on the burden of NDDs are necessary for planning effective prevention and intervention strategies. However, in most low-resource settings, administrative databases and medical records are neither widely available nor complete, necessitating different case identification methods than the relatively efficient and inexpensive record-based approaches frequently used in the developed world. An intensive effort was launched in the 1980s to develop and evaluate a method of case identification for children with disabilities as an indicator of health status in countries with scarce resources (Durkin et al., 1991, 1994, 1995a, 1995b; Shrout and Newman, 1989; Thorburn, 1993; Thorburn and Desai, 1991; Thorburn et al., 1992, 1993; Zaman et al., 1990). The result was the formulation of a two-stage approach using door-to-door screening with a simple caregiver questionnaire, followed by clinical assessment of children who screen positive.

The screening instrument (Ten Questions (TQ)) was designed for applicability across cultures by using a simple yes–no response format, focusing on universal abilities that are typically acquired by children in all cultures, and asking the parent to compare their child to others of the same age and from their cultural setting. The age range of 2–9 years was selected because it is very difficult to distinguish typical from atypical neurodevelopment in children younger than 2 years, and children older than 9 years would, in general, require different screening questions (Durkin et al., 1991). The validity of the TQ for screening for serious disability has been shown to be high in Bangladesh, Pakistan, and Jamaica (Durkin et al., 1994) and among 6- to 9-year-old children in Kenya (Mung'ala-Odera et al., 2004). Subsequent to these pioneering efforts, the TQ has been applied in various low-resource settings including Kenya (Muga, 2003) and South Africa (Christianson et al., 2000; Couper, 2002). The TQ was constructed to identify children with cognitive and motor impairment; seizure disorders; and serious speech, vision, and hearing impairments. ASDs and other behaviorally defined disorders were not specifically included. Since the development of the TQ, widespread public concern has developed regarding a possible true increase in the occurrence of ASDs and related disorders (reviewed in Elsabbagh et al., 2012). Despite a growing body of population-based descriptive and prevalence studies of ASDs from around the globe, epidemiologic data from low-resource settings remain lacking.

We conducted a planning and feasibility study (Tumaini Child Health (TUCH) Project) in the sub-Saharan country of Uganda using an expanded version of the TQ that includes additional questions intended to screen for ASDs. The overall aim of this study was to develop, pilot, and evaluate screening and assessment tools and procedures to identify children with moderate to severe NDDs, including ASDs, for use in Uganda, a low-resource setting that shares many cultural features with other countries in sub-Saharan Africa.

Methods

Setting and study population

This study was conducted from August 2010 to March 2011 and included children 2–9 years of age in randomly selected parishes from one urban and one rural district. Kampala District, the only urban district in the country, is the nation's capital and includes the Makerere University—Mulago National Referral Hospital. Wakiso District is a primarily rural district adjacent to Kampala with communities similar to those in more remote areas of the country but with reasonable accessibility to Makerere University—Mulago Hospital.

A Technical Advisory Group (TAG) that included Ugandan and American clinicians and epidemiologists was established to provide guidance to project activities. Prior to data collection, the local project team made several visits to the study areas to discuss the project with district and community leaders and seek their active cooperation in publicizing the project and its purpose, assuring confidentiality, and reassuring parents that participating children would be provided with health services as needed. With this information coming from recognized community leaders in personal conversations, public announcements at markets, churches, and local council meetings (which all citizens in the village are supposed to attend), we sought to maximize community engagement and contribute to a reduction in stigma associated with developmental disabilities in the communities.

Screening

Screening was conducted with a 23-item screener (23Q) that includes all questions in the original TQ, 10 additional ASD questions (5 for all children 2–9 years of age and 5 for children <5 years of age only), and three other questions intended to increase screening capability for visual, hearing, and seizure impairments (see Appendix 1 for copy of 23Q). Screening questions for ASDs were developed by the TAG in consultation with additional American clinicians.

The 23Q was translated into the local language, *Luganda*, and back translated by a team of certified professional translators. Pilot testing was conducted in Mulago National Referral Hospital, and further revisions made as necessary to assure that the questions were querying the intended behaviors. Using a broad or global interpretation of the screener (and following the procedures established in the TQ validation studies), a result was considered positive if a child failed one or more items on the 23Q.

A cluster sampling strategy (Bennett et al., 1991) with probability proportionate to the most recent population

census (Uganda Bureau of Statistics, 2002) was used to select 10 rural parishes (ranging in total population size from 1800 to 6600), 5 parishes in the urban slum area (total population ranging from 15,000 to 57,000), and 4 parishes in the urban upscale area (total population ranging from 11,000 to 23,400). In each selected parish, the project interviewer, with the assistance of a parish mobilizer known in the community, determined the midpoint of the parish and then randomly selected a direction in which all eligible households were screened until the desired sample size was achieved.

At each household, the parish mobilizer introduced the research assistant to a primary caregiver (defined as a person who had direct responsibility for the child and played a parental role), solicited written informed consent for participation, and then departed for the 23Q to be administered by the project interviewer in private. Following the screening interview, all children who screened positive and every third child who screened negative were invited to participate in a clinical assessment, and an appointment was made, usually for the following day. The Global Positioning System (GPS) coordinates and mobile phone contact numbers for each household were recorded for follow-up contact as needed. All field data collection staff were Ugandans fluent in English and Luganda. No incentives for participation were offered other than the provision of a free medical examination with referrals, as appropriate and available, for any health problems that were identified.

Clinical assessments

Based on TAG discussions about local feasibility, a two-step assessment process was devised for establishing NDD diagnoses. In Step 1, all children who screened positive and the sample of every third child who screened negative were invited to participate in a comprehensive Medical Officer Examination (MOE) held in their local community and conducted by a project Medical Officer without prior knowledge of the child's screening results. The MOE followed a protocol derived from the Medical Assessment Form used in the original TQ validation studies but with some modifications to shift specialized components to Step 2 examinations. Upon completion of the MOE, referrals for specialist examinations were made without regard to initial screening status for all children with suspected disability, using protocols designed for the project (copies of the MOE and referral protocol are available from corresponding author upon request).

In Step 2, experienced specialists at Mulago Hospital conducted systematic examinations for referred children based on examination protocols for each of the NDDs. The protocols were developed by the TAG and incorporated the specialized testing not included in the MOE and additional elements as deemed necessary by the specialists for diagnosis and management. ASD was diagnosed by an experienced child psychiatrist using *Diagnostic and Statistical Manual of Mental Disorders* (*DSM-IV-TR*) criteria (American Psychiatric Association, 2000), guided by the use of a standard data collection form. CI was determined by a neuropsychologist using an adapted *Kaufman Assessment Battery for Children* (*KABC-II*) for children above 5 years (Kaufman and Kaufman, 2004) and the *Mullen Scales of Early Learning* for those below the age of 5 years (Mullen, 1995). Results were compared with a normative sample of Ugandan children of similar age (Bangirana et al., 2009); children scoring less than −2 SDs on any one of four areas assessed were considered to have moderate to severe CI, and those scoring less than −3 SDs were rated severe. A diagnosis of epilepsy was made based upon standard clinical criteria for the diagnosis of each major seizure type and epilepsy syndrome, including history and neurology examination findings (Meinardi et al., 2001; Thurman et al., 2011). Using standard clinical criteria, diagnoses of CP (Rosenbaum et al., 2007), speech and language impairment (De Lamo White and Jin, 2011, hearing impairment (Kieran and Fenton, 2007), and vision impairment (Adoh and Woodhouse, 1994; Hyvarinen, 2000) were recorded on project data collection forms. Specialist assessments were conducted at Mulago National Referral Hospital, using a triage protocol for children with multiple referrals. The presence of a hearing impairment referral in a child was always given priority because it was pivotal in determining whether the child could qualify to be assessed for other co-existing impairments such as speech and language, ASDs, or CI. Due to scheduling constraints, multiple referrals sometimes required multiple trips to Mulago Hospital occurring on different days (the triage protocol and specialist assessment forms are available from corresponding author upon request). For the clinical assessments (both Step 1 and Step 2), transportation and light snacks were provided; children received medical examinations free of charge and were offered appointments for follow-up care and/or were referred to specialized schools or other services as needed. No financial incentives were provided.

Statistical analysis

Primary analyses focused on a global interpretation of the 23Q for which a child was considered to have screened positive if they failed one or more items. To estimate population prevalence and validity of the screener, we used the statistical techniques recommended by Shrout and Newman (1989) and Morvan et al. (2008) to adjust for a two-stage design with unequal sampling fractions between the screen positive and negative. We treated screening as the first stage and the MOE followed by specialty evaluations as a combined second stage, using diagnoses established in the specialty examinations as the final determination of the presence/absence of one or more of

Table 1. Characteristics of children in Tumaini Child Health (TUCH) Project, Uganda 100812.

	Total (N = 1169)
Gender	
Boys	536 (45.8%)
Girls	633 (54.2%)
Residence	
Urban	580 (49.6%)
Rural	589 (50.4%)
Age at screening	
24–60 months	653 (55.9%)
61–108 months	515 (44.1%)

the seven NDDs. The adjusted estimates assume the children who screened either positive or negative but were lost to follow-up between screening and the clinical assessments had the same rate of disability as children who participated in the clinical assessments (here called as "Estimate B"). We also computed alternate estimates that assume the children who screened negative but were lost to follow-up after screening had no disabilities ("Estimate A"). Because of the uncertain assumptions around attrition and the lack of statistical independence due to the cluster sampling of households and children, we do not present formal confidence intervals around the estimates. Rather, we recommend that the two estimates be interpreted as providing boundaries to parameters that cannot be more precisely known.

Ethical approval

Ethical clearance was granted by the School of Medicine Research and Ethics Committee, Makerere University College of Health Sciences and Uganda National Council of Sciences and Technology, and by the Committee for the Protection of Human Subjects, California Health and Human Services Agency.

Results

The study sample of screened children included 1169 children, 2–9 years of age, from the sampled parishes (Table 1), 50% urban and 50% rural. Fifty-four percent of the screened children were girls (46% boys) and 56% were in the younger age range of 24–60 months (44% in the older group). Among all screened children, 320 (27%) failed one or more items on the 23Q and met the definition of screening positive (Table 2). Boys, urban children, and children in the older age group were somewhat more likely to screen positive (Table 2). Considering only the original TQ items on the screener, 25% of all children screened positive, and based only on the ASD items on the 23Q, 8.9% of all children screened positive (Table 2).

Table 2. Developmental disability screening results from 23Q screener, Tumaini Child Health (TUCH) Project, Uganda.

Positive screen criteria	Total (N = 1169)	
	n	Percentage of screened
≥1 Positive items 23Q	320	27
Boys	160	30
Girls	160	25
Urban	166	29
Rural	154	26
Age 24–60 months	166	25
Age 61–108 months	154	30
≥1 Positive items TQ	289	25
≥1 Positive items ASD	104	8.9

TQ: Ten Questions; ASD: autism spectrum disorder.

All 320 children who screened positive on the 23Q were invited to participate in the MOE, as were 283 children who screened negative. Of the 603 invited children, 379 participated (68% of children who screened positive and 58% of children who screened negative; Table 3). The remaining children invited to participate in MOE were considered lost to follow-up (104 who screened positive and 120 who screened negative). Half of all children who participated in the MOE received a referral for a specialty evaluation (n = 189), and 141 of these children participated in one or more specialty evaluations, representing 75% of referred children who screened positive and 75% of referred children who screened negative (Table 3).

Based on the specialist evaluations, 68 children (55 who screened positive on the 23Q and 13 who screened negative) received one or more diagnoses of an NDD (Tables 3 and 4). The most common disability was moderate to severe CI, followed by epilepsy, speech and language impairment, hearing impairment, and ASD (Table 4). Inclusion of those with moderate CI resulted in an estimated population prevalence of 10/100 children (Table 5, Estimate A) to 12.8/100 children (Table 5, Estimate B). Excluding those with moderate CI reduced the estimate of prevalence to 7.7/100–8.9/100. The estimated prevalence of the six NDDs included in the TQ ranged from 10.4/100 to 12.7/100 with moderate CI included and 7.8/100–9.1/100 with moderate CI excluded (Table 5). Eight children with ASDs were identified, yielding estimated prevalence of 1.2/100–1.3/100 children in this population (Table 5).

Validity of screener

Using the global interpretation of disability for the 23Q and with moderate CI included, sensitivity is 0.55 under Estimate B and 0.68 under Estimate A. With moderate CI excluded, sensitivity increases to 0.70–0.80 (Table 5). For the six NDDs included in the TQ and evaluating only TQ

Table 3. Participation and follow-up for children in MOEs and specialist exams, TUCH Project, Uganda.

	Screened positive (n = 320)		Screened negative (n = 849)		Total children (N = 1169)	
Invited to participate in MOE	320	100%	283	33%	603	52%
Clinically evaluated by MOE	216	68%[a]	163	58%[a]	379	63%[a]
Received referral for specialist evaluation	122	56%[b]	67	41%[b]	189	50%[b]
Clinically evaluated by specialist	91	75%[c]	50	75%[c]	141	75%[c]
Received specialist diagnosis ≥1 NDDs	55	60%[d]	13	26%[d]	68	48%[d]

MOE: Medical Officer Examination; NDD: neurodevelopmental disorder; TUCH: Tumaini Child Health.
[a]Percentage of invited.
[b]Percentage of evaluated (MOE).
[c]Percentage of referred.
[d]Percentage of evaluated (specialist).

Table 4. NDDs specialist diagnoses, TUCH Project, Uganda.

Received ≥1 NDD specialist evaluations	N = 141	
	n	%
Received ≥1 NDD diagnoses from specialist	68	48.2
Cerebral palsy	5	3.5
Epilepsy	15	10.6
Speech and language	12	8.5
Hearing	11	7.8
Vision	1	0.7
Cognitive (including moderate)	37	26.2
Cognitive (excluding moderate)	21	14.9
Autism spectrum disorders	8	5.7

NDD: neurodevelopmental disorder;
TUCH: Tumaini Child Health Project.

items on the screener, the sensitivity is 0.49–0.60 with moderate CI included and 0.60–0.70 with moderate CI excluded (Table 5). These results indicate that, depending on which assumptions are used, up to 80% of the children with a specialist-diagnosed disability screened positive on the 23Q and up to 70% screened positive on the TQ.

For receiving a specialty diagnosis of ASDs, the sensitivity of the ASD items is 0.52–0.57. Of the eight children who received a diagnosis of ASDs, one child screened negative on all items on the 23Q, one child screened negative on the ASD items but positive on TQ items, and six children screened positive on both ASD and TQ items. Sensitivity for receiving a diagnosis of ASDs increases to 0.69 (Estimate B) for the combination of screening positive on both ASD *and* TQ items and decreases to 0.49 when only the original TQ items are considered.

Specificity of the 23Q was 0.77 under both Estimates A and B (with moderate CI included or excluded); for the TQ items only, specificity was 0.79 (Table 5). These results indicate that among the children without NDD, over 75% screened negative. For the ASD questions only, specificity is above 0.90. Positive predictive value (PPV) for the 23Q and the TQ alone are both 0.22–0.26, indicating that only about one-fourth of the children who screened positive received a specialty diagnosis of one or more NDDs; PPV was substantially lower for ASD items alone (0.08) indicating that less than 10% of children who screened positive on these items received a diagnosis of ASD. The negative predictive value (NPV) was above 0.90 for all comparisons, indicating that, in this population, a very high proportion of those who screened negative on the 23Q, the TQ items only, or the ASD items only did not meet the criteria for a diagnosis of a moderate to severe NDD (Table 5).

Discussion

The TUCH Project in Uganda offers a population-based evaluation in a low-resource setting of a simple household screener for initial identification of children with moderate to severe developmental disabilities, including ASDs. To our knowledge, this represents the first-reported validation in a developing country of a relatively low-cost, community-based screener for NDDs that includes questions specifically intended to screen for ASDs. To construct the 23Q, we modified the TQ, previously validated for screening other moderate to severe developmental disabilities by adding age-appropriate items for ASDs that are similar to those used in ASD screeners in pediatric clinical settings in developed countries. Household screening was followed by clinical assessments of children who screened positive and a sample of those who screened negative to obtain diagnoses (and treatment) for the children. For the clinical assessments, we used a two-step approach with a general pediatric examination conducted by a medical officer who then referred children for specialty evaluations as indicated by study protocol. This approach was requested by the local hospital-based specialists to accommodate their schedules and patient waiting lists. While the two-step assessment provided both comprehensive and highly specialized services to a large number of children, including many who would not otherwise have received care, compliance was challenging for some families due to the time commitment involved.

Table 5. Estimated prevalence and validation of screener, TUCH Project, Uganda.

	Estimate A					Estimate B				
	Prevalence/100	Sensitivity	Specificity	PPV	NPV	Prevalence/100	Sensitivity	Specificity	PPV	NPV
≥1 Positive items 23Q, with moderate CI included	10.3	0.68	0.77	0.26	0.95	12.8	0.55	0.77	0.26	0.92
≥1 Positive items 23Q, with moderate CI excluded	7.7	0.80	0.77	0.23	0.98	8.9	0.70	0.77	0.23	0.96
≥1 Positive items TQ, with moderate CI included	10.4	0.60	0.79	0.25	0.95	12.7	0.49	0.79	0.25	0.91
≥1 Positive items TQ, with moderate CI excluded	7.8	0.70	0.79	0.22	0.97	9.1	0.60	0.79	0.22	0.95
≥1 Positive ASD items, received ASD diagnoses	1.2	0.57	0.92	0.08	0.99	1.3	0.52	0.92	0.08	0.99

TUCH: Tumaini Child Health; TQ: Ten Questions; ASD: autism spectrum disorder; CI: cognitive impairment; PPV: positive predictive value; NPV: negative predictive value.
Estimate A assumes children who screened negative but were lost to follow-up after screening had no disabilities.
Estimate B assumes all children lost to follow-up after screening had the same rate of disability as children who participated in the clinical assessments.

Using the 23Q, we found that 27% of children screened positive on one or more items. Using the original TQ items only, 25% of children screened positive. A substantially higher proportion of children in our study failed the TQ than has been reported elsewhere—8.2% of children in Bangladesh; 14.7% in Pakistan; 15.6% in Jamaica (Durkin et al., 1994); 9.3% in rural Kenya (Mung'ala-Odera et al., 2004); 16% in Bondo, Kenya (Muga, 2003); and 8.2% in KwaZulu-Natal, South Africa (Couper, 2002).

Based on the specialty diagnoses, we obtained an estimated population prevalence of 10–13/100 children for the seven NDDs combined (prevalence of 8–9/100 if moderate CI is excluded), with similar results for the six NDDs included in the original TQ. Our estimated prevalence is higher than has been reported in Bangladesh (1.6/100; Zaman et al., 1990), Pakistan (4.4/100; Durkin et al., 1995a), or South Africa (6/100; Couper, 2002) and indicates a very substantial burden of disability among children in the sampled communities.

The estimated prevalence of 1.2–1.3/100 for ASDs in this population is within the range reported recently in the United States (Autism and Developmental Disabilities Monitoring Network, 2012) and above the median reported from a worldwide review (Elsabbagh et al., 2012). If we assume that the eight children with ASDs diagnosed in the specialty exams represent all ASD-affected children in this population of screened children (i.e. no loss of cases due to attrition or sampling of children who screened negative), the unadjusted prevalence is 0.68/100. These estimates suggest a rate of moderate to severe ASD occurrence in this developing country that is similar to that reported from more highly developed settings. However, as this initial planning study was not designed to investigate complete population-based prevalence and focuses only on moderate to severe impairment, it may be premature to conclude that these ASD estimates accurately reflect the true prevalence in this population. It is important to note that with our very modest sample size, the estimates would change noticeably with the addition or subtraction of just one child.

For the 23Q, the sensitivity ranged from 0.55 to 0.80 and for the TQ alone, from 0.49 to 0.70, depending on which set of assumptions we used and whether moderate CI was included as a diagnosis or not. As would be expected, sensitivity is improved with moderate CI excluded and under Estimate A in which we assume that the children who screened negative but were lost to follow-up after screening had no disabilities. The TQ screening items did not perform as well in this Ugandan population as previously reported from Bangladesh, Pakistan, and Jamaica where sensitivity of the TQ was around 0.80 for detecting serious cognitive, motor, and seizure disabilities (Durkin et al., 1994). Our specificity results are also somewhat lower than reported by Durkin et al. (1994) but indicate that among children without a NDD, a very high proportion screened negative (>77%). We also found that approximately one-quarter of

all children who screened positive on the 23Q or TQ items alone obtained one or more specialty diagnoses (PPV) and that among all children who screened negative, 95% or more did not have an NDD (NPV). These results are consistent with those obtained elsewhere that support the usefulness of the household screener as a practical tool for identifying a pool of high-risk children who are likely to benefit from follow-up clinical assessments.

Although modeled on screening and assessment projects conducted elsewhere, some design features of our feasibility study likely contributed to the differences between our results and those reported from other TQ validation studies. Our sample size was smaller, and in contrast to the other studies that used one comprehensive clinical assessment to assign NDD case status, we implemented a two-step assessment protocol. The cognitive component of the MOE was derived from an instrument designed for application in more developed settings (*Parents' Evaluation of Developmental Status–Developmental Milestones* (*PEDS-DM*; Glascoe and Robertshaw, 2007)) and led to an unrealistically high number of referrals, particularly among the rural children, even when we used modified criteria that excluded school curriculum–based items. Our cognitive specialty evaluations, which used different tools than those in the prior validation studies, also relied on instruments originally designed for application in more industrialized settings, although they were scored for our study using norms derived for a Ugandan population of similar age. Measures of adaptive behaviors may have been more appropriate for our population. It should also be noted that we did not conduct formal inter-rater reliability for either screening or clinical assessments, as doing so was beyond our capability in this feasibility study. We also were not able to conduct formal evaluation of the success of our community sensitization efforts, but our household interviewers reported that caregivers typically already knew about the project and were very welcoming, especially in the rural areas.

A further consideration is our attrition rate for the clinical assessments, which is noticeably higher than in the other validation studies in which participation in the clinical assessments was reported to be between 81.8% and 86.4% (Durkin et al., 1994). Although MOEs were held in a location close to the residential community and transportation was provided, participation was time-consuming for families because of transportation logistics and waiting time. Similarly, the specialized evaluations, particularly for rural residents, required considerable travel at prescheduled appointment times, putting demands on the family that likely contributed to nonparticipation. Despite these challenges, rural children participated to a greater extent in the follow-up assessments than the urban children. (In fact, some rural families brought nonstudy children to the MOEs, requesting that they be examined, as health-care services were otherwise inaccessible for the children.) Children with multiple referrals were scheduled to be seen by multiple specialists, and some children did not receive all relevant examinations and diagnoses, leading to an undercount for some specific diagnoses. For this and other reasons as discussed above, we advise against disorder-specific interpretations of our data.

Cultural factors likely contributed to our findings and may be relevant to NDD screening in other low-resource communities. In many poor Ugandan communities with extremely limited access to outside influences through media and personal contact, there is low awareness or concern about developmental disabilities and differences. If the child can perform the basic skills of daily living through household accommodation, CI may not be identified until the child is in school. In the rural areas, many children were not yet in school, even among the older children, and had not yet been exposed to numbers and letters or environments outside their very local neighborhood. Homes in poor communities were typically lacking commercially made toys or other objects, including books and magazines. In the upscale urban community, many children with disabilities had previously been diagnosed and were receiving care, so parents saw little advantage to participation. For the MOE, we found higher rates of lost to follow-up among those who screened negative, especially in the urban setting in which health care for children was relatively accessible. Both among upscale and poor families, participating in the two-step assessment was difficult for parents, as many needed to spend that time doing their work to support their family. Lost to follow-up, in rural areas in particular, may also be related to caregivers believing that their child's disability was due to some evil spirit and that it would be a waste of time to come for further medical evaluations and treatment.

For identifying children with ASDs, we found that screening positive on a combination of ASD *and* TQ items had a sensitivity of 0.69, higher than when only the ASD items were considered. Cultural factors may, at least in part, explain these results. In traditional Ugandan society, children with the behavioral characteristics of ASDs are likely to be viewed with far more negativity than a child with physical disabilities because of cultural perceptions that these behaviors result from possession by evil spirits or neglect of traditional taboos or myths. Children with only behavioral features of ASDs but without other impairments may have screened negative on the 23Q due to caregiver reluctance to acknowledge behaviors or misinterpretation of the questions. Another possibility is that some children with behavioral features of ASDs may have screened positive but failed to participate in the clinical assessments due to concerns related to stigma. Further study may be helpful in sorting out these and other cultural factors.

A unique aspect of the project is the expansion of the TQ to include questions on ASDs for application in a largely poor setting with little cultural awareness of this complex

behavioral condition. While the addition of ASD items to the screener failed to uniquely identify children with ASDs who would not otherwise have been identified by the TQ alone, failure of one or more ASD items in combination with failure of one or more TQ items was more sensitive than either set of items alone in identifying a subgroup of children at especially high risk of ASDs. Improvement of the sensitivity of the screener to identify children with ASDs may benefit from focus groups and other qualitative approaches for development of more culturally specific questions. We recommend further studies in low-resource settings to evaluate the addition of ASDs and other behavioral screening questions so that ASDs and related disorders can be incorporated into culturally appropriate and comprehensive screening, assessment, and intervention programs.

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Conflict of interest

The content is solely the responsibility of the authors and does not necessarily represent the official views of the Fogarty International Center or the National Institutes of Health.

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Appendix 1

Tumaini Child Health (TUCH) Project

23 question screener (23Q)

(Shaded questions were added to the original Ten Questions Screener (TQ))
Please circle the respondent's answer following each question.

Compared with other children, did the child have any serious delay in sitting, standing, or walking?	YES*	No	DK	N/A
Does the child have difficulty in walking or moving his/her arms or does he/she have weakness and/or stiffness in the arms or legs?	YES*	No	DK	N/A
Compared with other children, does the child have difficulty seeing either in the day time or at night?	YES*	No	DK	N/A
In school going children ask: Does the child read well what is written on the blackboard when sitting at the back of the class?	YES	No*	DK	N/A
Does the child appear to have difficulty hearing?	YES*	No	DK	N/A
Does the child often ask you to repeat what you have said?	YES*	No	DK	N/A
When you tell the child to do something, does he/she seem to understand what you are saying?	YES	No*	DK	N/A
Does the child learn to do things like other children his/her age?	YES	No*	DK	N/A
Compared with other children of his or her age, does the child appear in any way mentally backward, dull, or slow?	YES*	No	DK	N/A
Does the child sometimes have fits, become rigid, or lose consciousness?	YES*	No	DK	N/A
Does the child have episodes of staring when you cannot get their attention by talking to them or touching them lightly?	YES*	No	DK	N/A
Does the child speak at all (can he or she make himself or herself understood in words; can he or she say any recognizable words?	YES	No*	DK	N/A
a. For 3- to 9-year-olds ask: Is the child's speech in any way different from normal (not clear enough to be understood by people other than his/her immediate family?	YES*	No	DK	N/A
b. For 2-year-olds ask: Can he or she name at least one object (for example: an animal, a toy, a cup, a spoon)?	YES	No*	DK	N/A
Does the child have difficulty making and maintaining eye contact?	YES*	No	DK	N/A

Does the child cry or get upset if you do not do particular routines the same way every day like using the same plate/ cup to serve his/her food/drink, letting him/her sit on a particular stool/chair in the house?	YES*	No	DK	N/A
Does the child take an interest in playing with other children?	YES	No*	DK	N/A
Does the child usually turn to look at you when you call his/her name?	YES	No*	DK	N/A
Does the child repeat phrases over and over exactly as they were said or heard from someone (or on the radio)?	YES*	No	DK	N/A
Additional questions for children <5 years of age				
Does the child engage in pretend play like "Mama ne Tata," driving, cooking?	YES	No*	DK	N/A
Does the child usually use his/her index finger to point, to indicate interest in something?	YES	No*	DK	N/A
Does the child often bring objects over to you (parent) to show you something?	YES	No*	DK	N/A
Does the child imitate you? (e.g. if you make a face, will the child imitate it?)	YES	No*	DK	N/A
If you point at a toy or a person across the room, does the child look at it?	YES	No*	DK	N/A

Screening result is positive if any one or more of the responses with an asterisk (*) is circled.