Assessment of a neuro-developmental screening tool in children in Bhutan [version 1; peer review: 1 approved, 2 approved with reservations]

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Abstract

Background: Developmental screening tools are designed to fit the cultural context in which they are utilized, yet often find a wider international audience. This study evaluates the efficacy of one such tool, the Parental Evaluation of Developmental Status: Developmental Milestones (PEDS:DM), developed in the United States and tested in the lower income Asian country of Bhutan. We aimed to test the PEDS:DM instrument to measure neurodevelopmental delay in children in Bhutan.

Methods: In total, 96 community-dwelling Bhutanese children (3-7 years old) without diagnosed neurocognitive conditions were recruited from ambulatory clinics in urban Bhutan in 2016 as part of a larger study on retinal imaging and cognitive and growth parameters. Scoring was based on neurocognitive domains (gross and fine motor, receptive and expressive speech, self-help, social-emotional). Rates of failure (meant to indicate delay) within domains were calculated.

Results: Modifications of some standard questions was deemed necessary by the study staff to suit the cultural context, such as replacing kickball with football in a question regarding games played with rules to maintain local relevance. In a modified PEDS:DM test with these improvised modifications, the mean percentage of age-appropriate domains failed was 58.8% and the mean percent delay was 12.3% (range 0-41.4%, available in n=83). The highest prevalence of failures was 59.4% for receptive language and 76.3% for expressive language, much higher than the lowest rate of failure seen in self-help (5.4%).

Conclusions: The PEDS:DM requires further modifications and validation studies before it can be reliably implemented to assess developmental delay in children in Bhutan. In this pilot study, the rate of delay as reported by the PEDS:DM would be scored as markedly elevated, especially when compared to available epidemiologic studies in the region.
**Keywords**
children, cognition, developmental delay, neurodevelopment, epidemiology

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Introduction
Neurodevelopmental delay is a common finding in United States pediatric practices, with the prevalence measured at 12–16% of all children. For the purpose of referring children to appropriate early intervention services, various screening tools have been designed to help clinicians identify developmental delay along different cognitive domains. While in Western countries there exist robust protocols for screening children to identify developmental delay, there are few available data regarding the utility of such protocols in lower income countries. Nonetheless, it is estimated that the majority of children with developmental disabilities (up to 80%) reside in low- or middle-income countries.

A commonly utilized developmental screening tool in the U.S. is the Parents’ Evaluation of Developmental Status: Developmental Milestones (PEDS:DM). The survey is designed to assess developmental delay in children from birth to age 7–11 years across several cognitive domains: fine motor, gross motor, receptive language, expressive language, self-help, and social-emotional. While the PEDS:DM has been well validated in the United States where the instrument was constructed, there are few studies confirming its validity across cultural contexts.

We aimed to assess the validity of the PEDS:DM survey in the lower-income country of Bhutan, a Himalayan country with no pediatric neurologists. Bhutan is typical of several lower-income countries where English is a language of school instruction and a pediatric neurodevelopmental screen could be valuable, particularly if used by non-physician health care workers in the field.

Methods
Ethics approvals
The study was approved by the Research Ethics Board of Health, convened by the Bhutanese Ministry of Health (PO-2015-011) and the Institutional Review Boards at Massachusetts General Hospital (2015P000159) and Children’s Hospital of Los Angeles (CHLA-15-00539). Neurocognitive screening was implemented as part of a larger study to correlate cognitive functioning and retinal and other growth parameters. Informed consent (written, or via thumb print if parent had low literacy) was obtained from each subject’s parent or next of kin over the age of the majority.

Location
The Kingdom of Bhutan is bordered by China and India. In 2014, the total expenditure on healthcare per capita was 89 USD, compared to 9402 USD in the USA. Subjects were recruited from the Jigme Dorji Wangchuck National Referral Hospital (JDWRNH) located in the capital city, Thimphu (population 138,736 in 2017). Specialist care is available, but there are no pediatric neurologists or subspecialized developmental pediatricians. Patients presenting to the JDWRNH Department of Ophthalmology outpatient clinic for routine vision screening were recruited as subjects so as to recruit community-dwelling children who were not seeking care for neurodevelopmental or neurological issues directly. Dzongkha is the primary spoken language in Bhutan. English is taught in schools and is the primary written language.

Enrollment
Recruitment took place in 2016. Inclusion criteria included all children aged three to seven years old. Children who did not have a parent present and who were unable to complete a retinal scan as part of a concurrent study were excluded from the cognitive testing. Subjects were provided with reimbursement of travel expenses amounting to 500 Bhutanese Ngultrums (equivalent to approximately 12 USD).

Anthropometric characteristics
At the time of enrollment, each child’s height, weight, and head circumference were measured and recorded according to the methods of the World Health Organization. Z scores were calculated for the height, weight, and head circumference of each child by an independent scorer using normative data from the World Health Organization.

Neurocognitive testing
The PEDS:DM survey was conducted on all eligible children by a U.S.-based pediatrician (BW) who was a fellow of pediatric neurology and native Bhutanese research staff (LT, KT). The survey assesses for cognitive development up to a maximum age limit, varying by task category. The domains and their corresponding upper age limits are: fine motor (6 years, 11 months), receptive language (7 years, 11 months), expressive language (6 years, 11 months), gross motor (4 years, 5 months), self-help (6 years, 11 months), and social-emotional (5 years, 5 months). Each domain includes questions of increasing difficulty correlated with specific ages. For each domain, each child was asked the question for their age, and then if incorrect, was asked the question for progressively younger age groups until he or she provided two consecutive answers correctly. Several questions were modified to provide greater cultural relevancy (see Extended data).

The reading and math sections of the screen were not used because those questions were deemed irrelevant to the population. Examples include a math question which requires recognition of a penny when Bhutanese currency does not include coins and a reading question that involved recognition of American “Exit” and “Caution” signs. To control for potential differences in parental literacy levels, surveys were conducted via interview by the pediatrician rather than having parents fill out a written survey. Subjects were only assessed in domains for which they were not over the maximum age limit for that domain.

Although Dzongkha is the primary spoken language in Bhutan, the evaluations were conducted in English due to high levels of English proficiency in the majority of participants and parents. Forward and backward translation to Dzongkha was provided by a Bhutanese research assistant, who was available at every participant encounter, when requested.
With the assistance of the Bhutanese research assistant, some questions were mildly modified to be culturally relevant. For example, the social-emotional domain question, “can your child play games with rules, like board or card games, kickball, or hopscotch,” was altered to the example of “football” which is a more common childhood game in Bhutan.

Scoring
Responses to questions were scored either based on parental reports of the child’s experience, or direct subject demonstration of the required task, as appropriate. For example, on a question in the fine motor domain, “can your child write any of the letters of the alphabet?” parents were asked to answer this question from their past experience of their child, and the subject was asked to perform the task. If there was a discrepancy between parental report and subject performance, then the score was determined by subject performance. No other questions were excluded from the testing.

The PEDS:DM responses were scored as both an age-dependent screen (pass/fail for the age appropriate task) and assessment (percentage delay). For example, if the child was 6 years 5 months old and the highest correctly answered question was for 6 years 0 months, then she was scored as having failed the age-dependent screen, and would be considered 7.7% delayed ($(1-(72 months/78 months))*100$) in the assessment. (Descriptions of each scoring method available in Table 1)

In addition to determining the failure rate (screen) and percentage delay (assessment) within individual domains, a screen composite score (the number of domains failed divided by the number of age appropriate domains assessed) and assessment composite score (mean percent delay) were calculated across all domains per subject. This provided an overall assessment of delay for each individual subject combining each domain assessed. PEDS:DM evaluations were scored by an independent scorer who was not present at the study site during the evaluations. To assess patterns of failure across the study population, the rates of failure for each domain were calculated for all children as well as for each age group individually (Table 2).

Statistical analysis
Basic data analysis was descriptive and included means, proportions, and percentages for the variables of interest. Where sample sizes were reduced due to incomplete data, the analyzable subcohort sample size is provided. Scoring of the PEDS:DM test was performed using the provided testing manual by a study staff member who did not perform the cognitive or anthropometric testing of the subjects directly (SG). All calculations were performed using Stata 12 (StataCorp. 2011. Stata Statistical Software: Release 12. College Station, TX: StataCorp LP).

Results
Subject demographics and anthropometric characteristics
In total, 96 children (37 male) aged 3 years, 1 month to 7 years, 11 months old were enrolled. There were no refusals from parents to have their child participate. The mean age was 6 years 0 months old. 26 subjects were not yet enrolled in school (Table 3 and Underlying data). Head circumference and height measurements followed a normal distribution. Weight and body mass index measurements skewed towards underweight (65.2% of subjects scored were under average weight) (Figure 1).

PEDS:DM Scores
PEDS:DM screen composite scores were generated for 92 of 96 subjects, with possible scores ranging from 0 to 100. For the screen composite score, only an answer to the age appropriate question was required. Four children were missing data on the age appropriate question for their age. Of the remaining 92 children who could be scored, the average score was 41.2 points (Table 2).

The overall assessment score determines the percentage of delay for a child across all domains. It required both the age-appropriate question and younger age groups’ questions to be answered correctly. Two questions in a row answered correctly were required to give a score. Of the 96 subjects, 13 were missing data that made the percent delay impossible to score. Of the remaining 83 children, the average score was 12.3 (range 0–41.4) (Table 2).

Domain screen failure rates were calculated for individual neurocognitive domains (Table 2). The language domains contained the largest rates of failure on the PEDS:DM: 59.4% for receptive language, 76.3% for expressive language. The lowest rate of failure was seen in self-help (5.4%).

Table 1. Definitions of scoring tools.

<table>
<thead>
<tr>
<th>Screening Tool</th>
<th>Subjects Assessed</th>
<th>Domains Assessed</th>
<th>Calculation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Domain specific screen</td>
<td>Individual child</td>
<td>Single</td>
<td>Pass/fail on age appropriate question for the domain</td>
</tr>
<tr>
<td>Screen composite score</td>
<td>Individual child</td>
<td>Multiple</td>
<td>Number domains failed/Number domains assessed</td>
</tr>
<tr>
<td>Assessment specific assessment</td>
<td>Individual child</td>
<td>Single</td>
<td>(1-[(Associated age level of highest correct answered question)/(child's age)]*100)</td>
</tr>
<tr>
<td>Assessment composite score</td>
<td>Individual child</td>
<td>Multiple</td>
<td>Mean percent delay across all domains assessed</td>
</tr>
<tr>
<td>Domain screen failure rate</td>
<td>All children</td>
<td>Single</td>
<td>Number of children who failed the domain/Number of children assessed for the domain</td>
</tr>
<tr>
<td>Mean assessment percent delay</td>
<td>All children</td>
<td>Single</td>
<td>Mean percent delay for all children across a single domain</td>
</tr>
</tbody>
</table>
### Table 2. Parental Evaluation of Developmental Status: Developmental Milestones (PEDS:DM) results.

<table>
<thead>
<tr>
<th>Age [years]</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>PEDS Total</th>
<th>Two-Stage Study [% with disability]</th>
</tr>
</thead>
<tbody>
<tr>
<td>Screen (failure rates)</td>
<td>(n=6)</td>
<td>(n=18)</td>
<td>(n=17)</td>
<td>(n=35)</td>
<td>(n=20)</td>
<td>(n=96)</td>
<td>(n=3491)</td>
</tr>
<tr>
<td>Fine Motor (&lt;7y-11mo)</td>
<td>2 (33.3%)</td>
<td>8 (44.4%)</td>
<td>7 (41.2%)</td>
<td>0 (0.0%)</td>
<td>8 (40.0%)</td>
<td>25 (26.0%)</td>
<td>199 (5.5%)</td>
</tr>
<tr>
<td>Receptive Language (&lt;7y-11mo)</td>
<td>1 (16.7%)</td>
<td>10 (55.6%)</td>
<td>16 (94.1%)</td>
<td>20 (57.1%)</td>
<td>10 (50.0%)</td>
<td>57 (59.4%)</td>
<td>-</td>
</tr>
<tr>
<td>Expressive Language (&lt;6y-11mo)</td>
<td>4 (66.7%)</td>
<td>15 (83.3%)</td>
<td>11 (64.7%)</td>
<td>28 (80.0%)</td>
<td>-</td>
<td>78 (76.3%)</td>
<td>102 (2.3%)</td>
</tr>
<tr>
<td>Self-help (&lt;6y-11mo)</td>
<td>0 (0.0%)*</td>
<td>1 (5.6%)</td>
<td>1 (6.2%)*</td>
<td>2 (5.7%)</td>
<td>-</td>
<td>26 (5.4%)</td>
<td>-</td>
</tr>
<tr>
<td>Social-emotional (&lt;5y-5mo)</td>
<td>0 (0.0%)</td>
<td>6 (35.3%)*</td>
<td>6 (66.7%)**</td>
<td>-</td>
<td>-</td>
<td>76 (37.5%)</td>
<td>-</td>
</tr>
<tr>
<td>Gross Motor (&lt;4y-5mo)</td>
<td>1 (16.7%)</td>
<td>1 (16.7%)**</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>86 (16.7%)</td>
<td>86 (2.0%)</td>
</tr>
<tr>
<td>Average screen composite score by age group</td>
<td>26.6 ± 8.8*</td>
<td>41.8 ± 21.8*</td>
<td>51.6 ± 21.2*</td>
<td>36.0 ± 17.6*</td>
<td>45.0 ± 39.4</td>
<td>41.2 ± 25.3*</td>
<td>-</td>
</tr>
<tr>
<td>Assessment (mean % delay ±SD)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fine Motor (&lt;7y-11mo)</td>
<td>4.5 ± 7.1</td>
<td>13.0 ± 13.0</td>
<td>8.6 ± 14.0</td>
<td>0 ± 0</td>
<td>2.9 ± 4.4</td>
<td>4.8 ± 9.7</td>
<td>-</td>
</tr>
<tr>
<td>Receptive Language (&lt;7y-11mo)</td>
<td>1.9 ± 4.7</td>
<td>18.8 ± 18.9</td>
<td>24.1 ± 12.0*</td>
<td>29.3 ± 12.7*</td>
<td>21.7 ± 22.1*</td>
<td>23.0 ± 17.1</td>
<td>-</td>
</tr>
<tr>
<td>Expressive Language (&lt;6y-11mo)</td>
<td>4.7 ± 4.2</td>
<td>19.6 ± 15.5</td>
<td>22.2 ± 9.1**</td>
<td>28.4 ± 12.0**</td>
<td>-</td>
<td>22.8 ± 13.5</td>
<td>-</td>
</tr>
<tr>
<td>Self-help (&lt;6y-11mo)</td>
<td>0 ± 0*</td>
<td>0.3 ± 1.3</td>
<td>0.3 ± 1.2**</td>
<td>1.3 ± 5.7</td>
<td>-</td>
<td>0.7 ± 4.0</td>
<td>-</td>
</tr>
<tr>
<td>Social-emotional (&lt;5y-5mo)</td>
<td>0 ± 0</td>
<td>5.7 ± 11.0*</td>
<td>6.1 ± 5.5***</td>
<td>-</td>
<td>-</td>
<td>4.7 ± 8.5</td>
<td>-</td>
</tr>
<tr>
<td>Gross Motor (&lt;4y-5mo)</td>
<td>2.9 ± 7.1</td>
<td>10.1 ± 16.0**</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>6.5 ± 12.4</td>
<td>-</td>
</tr>
<tr>
<td>Average assessment composite score by age group</td>
<td>2.3 ± 1.6</td>
<td>11.1 ± 10.0*</td>
<td>12.9 ± 6.3*</td>
<td>14.7 ± 5.8*</td>
<td>12.1 ± 11.5*</td>
<td>12.3 ± 8.6</td>
<td>-</td>
</tr>
</tbody>
</table>

Table 3. Participant background information: age, education, medical comorbidities.

<table>
<thead>
<tr>
<th></th>
<th>Male</th>
<th>Female</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total Participants</td>
<td>37</td>
<td>54</td>
<td>96</td>
</tr>
<tr>
<td>Age, years (n=96)</td>
<td>(n=37)</td>
<td>(n=54)</td>
<td></td>
</tr>
<tr>
<td>Mean ± SD</td>
<td>6.2 ± 1.2</td>
<td>5.9 ± 1.2</td>
<td>6.0 ± 1.2</td>
</tr>
<tr>
<td>Range</td>
<td>3y, 1m - 7y, 11m,</td>
<td>3y, 1m - 7y, 10m</td>
<td>3y, 1m - 7y, 11m</td>
</tr>
<tr>
<td>Years in School (n=93)</td>
<td>(n=37)</td>
<td>(n=54)</td>
<td></td>
</tr>
<tr>
<td>No school</td>
<td>9 (24%)</td>
<td>15 (28%)</td>
<td>26 (28%)</td>
</tr>
<tr>
<td>&gt;0 and &lt;6 months</td>
<td>3 (8%)</td>
<td>1 (2%)</td>
<td>4 (4%)</td>
</tr>
<tr>
<td>&gt;6 and &lt;12 months</td>
<td>4 (11%)</td>
<td>7 (13%)</td>
<td>12 (13%)</td>
</tr>
<tr>
<td>&gt;12 and &lt;24 months</td>
<td>8 (22%)</td>
<td>15 (28%)</td>
<td>24 (26%)</td>
</tr>
<tr>
<td>&gt;24 months</td>
<td>13 (35%)</td>
<td>14 (26%)</td>
<td>27 (29%)</td>
</tr>
<tr>
<td>Medical Complications (n=92)</td>
<td>(n=36)</td>
<td>(n=52)</td>
<td></td>
</tr>
<tr>
<td>Pregnancy complications (mother) (n=93)</td>
<td>1 (3%)</td>
<td>5 (10%)</td>
<td>6 (6%)</td>
</tr>
<tr>
<td>Pregnancy hospitalizations (mother)</td>
<td>4 (12%)*</td>
<td>8 (15%)*</td>
<td>12 (13%)</td>
</tr>
<tr>
<td>Child medical problems</td>
<td>5 (14%)</td>
<td>7 (14%)</td>
<td>12 (13%)</td>
</tr>
</tbody>
</table>

*Sex not recorded for 5 participants.
The domain screen failure rates produced non-uniform results across childhood age groups. Percentage failure of the fine motor domain by age group ranged from 33.3–44.4%, with the striking exception of 0% for six-year-olds. Five-year-old children had the highest failure rate in receptive language (94.1%) compared to other ages, yet had the lowest failure rate in expressive language (64.7%). For the social-emotional domain, failure rate appeared to increase with age.

The assessment composite score (percent delay) measured the degree of delay within particular domains. The score across all domains was 12.3% (range 0–41.4%, n=83). Similar to the screen composite scores, the language domains had the highest assessment composite scores. Within the language domains, the greatest delays were in the five- and six-year-old age groups (receptive language 24.1–29.3%, expressive language 22.2–28.4%). Relatively smaller total percent delays were observed in the motor (both fine and gross), social-emotional, and self-help domains, in a manner that roughly aligns with the failure rates for these domains in the domain specific screen.

Discussion

The validation of culturally relevant screening tools provides a means for more accurate assessment of developmental delay in children in lower income countries as well as prevalence data on the extent of developmental delay in these locations. Here we evaluated the applicability of the PEDS:DM in Bhutan to determine its utility in this non-Western setting.

Cultural differences leading to suboptimal performance

The exceedingly high rates of calculated delay in expressive and receptive language limit the ability to accurately interpret the other domains of the PEDS:DM test. A similar conclusion was reached in a study conducted using the PEDS:DM in a population of Thai children. That study reported a greater proportion of the subjects being classified with medium risk for developmental delay when compared to prior standardization studies in the United States. The authors postulated that local culture influenced certain responses of parental concern about their child’s development (such as expressing worry about a child being left-handed), resulting in an overestimation in the identification of actual delay.

The PEDS:DM was originally designed and intended for implementation in a Western patient population. Bhutanese cultural differences appeared to consistently invalidate certain questions of the test. For instance, for the self-help question, “Can your child get dressed by himself or herself?” almost all of the parents answered “no.” Bhutanese children are dressed in traditional outfits that are more complicated to put on compared to Western-style clothing, and thus children are usually unable to independently dress until an age much later than what was tested on the PEDS:DM. Children were often unable to correctly answer a question regarding irregular plural nouns (i.e. “tooth” and “teeth”), suggesting such irregular English grammar structures are not introduced in the early school years, when most Bhutanese children are taught English. This may also account for the peak for delays in the assessment for expressive and receptive language occurring in five- and six-year old children, ages when subjects are first enrolled in formal schooling.

In school-age children, there was striking consistency in the manner that incorrect answers were provided. This may suggest a
difference in academic curricula between the United States and Bhutan that led to suboptimal performance of Bhutanese children on an American-designed test. One such example may explain the high failure rate in receptive language among five-year-old children. In order to pass this domain, subjects had to identify at least three of the following body parts: shoulders, elbows, heels, ankles. Nearly all five-year-old children could correctly identify shoulders and elbows and very few could identify ankles and heels.

For six-year-old children to earn a “pass” on the fine motor domain, they had to correctly write three or more letters of the alphabet and draw a triangle with all lines connecting correctly. The abnormally high pass rate (100%) for this category amongst six-year-old children may be due to such skills being highly emphasized in school curriculum or at home by this age.

Our study gave credit to subjects who were able to demonstrate tasks, rather than accepting parents’ report on subjects’ ability. For example, subjects on one question were asked to recite the alphabet and would only be scored as “correct” if subjects could do so during the study visit. Subjects for the most part provided attempts at most questions. Valuing parental reporting over subject performance may have been a more representative assessment of the child’s abilities, as parents would more likely know the child’s skill performance in a natural environment.

Comparison to the Two-Stage Child Disability Study

Findings

The Two-Stage Child Disability Study was conducted by UNICEF and the National Statistics Bureau of Bhutan and was the largest recent country-wide screen of disability in Bhutan, including 3491 subjects. The study utilized a first-stage screen with the Ten Questions questionnaire (TQ). Those who were positive were then screened with the second stage: the Rapid Neuro-developmental Assessment (RNDA) for two to five year old children and the Rapid Functional Assessment (RFA) for five to nine year old children. The TQ, RNDA, and RFA were designed for use in low-income settings and focus on general function abilities, avoiding culture-specific skills (i.e. tying shoelaces). These tests were initially developed in large epidemiologic studies in Bangladesh, conducted by the Bangladesh Protibondhi Foundation, who provided these tests to the Two-Stage Child Disability Study. The tools have since been validated in other lower income settings on a country-wide scale in Jamaica and Pakistan.

The Two Stage Child Disability Study provides a worthwhile comparison for our results. It reached a wider group of children geographically and with a much larger and population-based sample. While the authors of that study noted lower sensitivity and specificity of the first-stage TQ methodology when compared to similar studies conducted in other countries, they believed the final determination of disability from the second-stage screen to be accurate. Similar to our study, the Two-Stage Child Disability Study uses assessment tools that were created and validated elsewhere (Bangladesh), however the authors were careful to choose these assessments for their lack of testing of culture-specific skills.

As a rough point of comparison, when our data are compared to that of the Two-Stage Child Disability Study, we see that in all domains for which there was a direct comparison available, the percentage of disability measured by PEDS:DM exceeded that seen in the Two-Stage Study. However, with only one available comparable study, normative data in this population remain unclear.

Limitations and strengths

One limitation of our study is sampling from a more developed region of Western Bhutan. Our study does not generalize to the entire Bhutanese population which is more rural and remote, such as in the Eastern gewogs (Dzongkha term for a group of villages) where we did enroll subjects. We suspect children suffering from significant disability in a lower income country could have accessibility issues, going to a hospital less often to accompany their parents for routine eye care or other appointments, and thus these children would not be well represented here. There is also the potential for selection bias, as subjects who could not complete the retinal scan as part of the larger study were not selected to complete the PEDS:DM. The retinal scan required children to sit still for several seconds; children with severe developmental delay would have had a more difficult time participating and would not be reflected in the PEDS:DM screening here.

While Dzongkha instructions were provided verbally, having written materials may also improve response accuracy during testing. Additionally, the PEDS:DM evaluation is designed as a screening test and therefore is intentionally sensitive in comparison to a confirmatory test such as was used in the second-stage of the Two Stage Child Disability study. This could be one of the reasons for the inflated rates of developmental delay measured in this study. It is also possible that subject performance anxiety increased the number of responses categorized as “incorrect.” If children are not accustomed to directly interacting with a healthcare provider during outpatient clinic visits, then requesting cognitive tasks may be overwhelming, leading to worse test outcomes. Finally, our small sample size overall and within each age group may contribute to the wide variations displayed between groups.

Our study strengths include assessment of a community-dwelling pediatric population that is not well represented in the neurodevelopmental research literature. We are unaware of other studies of the PEDS:DM in Bhutan or neighboring regions. Children were evaluated by a senior fellow of pediatric neurology where none are otherwise available. We also provide a detailed assessment of how and likely why children may be erroneously labeled as delayed using a well-established screening tool in the U.S.A.
We demonstrate that the PEDS:DM may not be an appropriate developmental screening tool for use in children in Bhutan due to cultural differences in questions and required developmental testing tasks. When compared to the Bhutan Two-Stage Child Disability Study which recruited 3,491 Bhutanese children, our study of just less than 100 community-dwelling Bhutanese children using PEDS:DM categorized significantly more subjects with disability in one or more neurocognitive domains, at times more than half of the study population.

Diagnostic assessments that are sensitive to small differences in developmental delay are an important tool for early discovery and allow for early interventions in these cases. Low- and middle-income countries have the highest burden of developmental delay yet lack these important tools to recognize changes early when the largest differences on children’s outcomes can be made through early life intervention. The presumed lack of cross-cultural applicability of the PEDS:DM to a population in Bhutan shows the importance of developing pediatric neuro-developmental tools to better screen children and determine accurate prevalence data in non-Western, non-English speaking countries. Locally-constructed developmental screening instruments such as the Angkor Hospital for Children Milestone Assessment Tool utilized in Cambodia, and the South Africa Road to Health Booklet utilized in South Africa, represent the promise of more valid, culture-specific tools on the horizon in other locations. While we do not suggest that tools such as PEDS:DM have no utility in this setting, we would advise culture-specific revisions of existing assessments before widespread implementation studies.

Data availability

Underlying data


This project contains the following underlying data:

Bhutan_fullTable_4June2019.xlsx (This is raw field data from 96 Bhutanese children undergoing the PEDS:DM evaluation. Cells where “N/A” or “not applicable” is entered reflect that the screening instrument is staged by the age of subject. Several subjects either were too young or failed to progress through the assessment, making more difficult questions no longer applicable to their testing.)

Extended data

Figshare: Extended Data: Field Notes and Modifications to the PEDS:DM Test for Bhutanese Children. https://doi.org/10.6084/m9.figshare.8246282

This project contains the following extended data:

• Modifications to PEDS:DM Testing in Bhutan.

Grant information

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The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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Open Peer Review

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Naila Z. Khan
Clinical Neurosciences Center, Bangladesh Protibondhi Foundation, Dhaka, Bangladesh

The paper reads well. Reading the results, without seeing the tables, was easier. The tables are confusing in the discrepancies between the language of the column headings and the data provided. I have listed the confusions below.

Table 2:
1. The language of the first column headings and row numbers, as presented, don’t match. For example, in the second row it says ‘Screen (failure rates)’, while all the numbers entered within brackets (why brackets?) in that row, by age group, probably indicate number of children who failed. The first column should have read ‘number of screen positives’ and not ‘rates’. If rates are to be calculated then for each of the age groups (3,4,5,6,7) a ‘total number screened’ should be mentioned, and a percentage calculated. The next 8 rows calculate percentages of the 2nd row ‘number of children’. Therefore, second row first column heading needs to be changed.

2. Last column: Two Stage Survey being used for comparison: Conversely, the second row number is the ‘Total number of children screened’. If we go by the 2nd row heading of ‘Screen (failure rates)’, it does not make sense.

3. First column row 10, under Average Score Composite Score by Age Group, is confusing, as this row does not have a texted heading. How does this row differ from row 19, i.e. last row of the table, which also has number of children in brackets with asterisks? Please clarify, or redesign this table. I understand that each item in the first column has a different set of children seen. Still, too many asterisks above (row 10) and below (row 19) to follow.

Table 3:

Rows and column heading are confusing. Repeats the same pattern as table 2.

1. Column one heading Age, years (96) simply repeats the Total Participants seen listed in row 2. Why not just say Mean Age in Years +/-SD (total=96) which is listed in row 4, and delete this row?
Years in School: This would mean to the reader ‘years of schooling’. However only total male/female numbers are given in the row. The next 5 rows under Years in School are even more confusing, as the age range goes from 0-24 months, i.e., below the pre-school age group. Otherwise, the reasons for their inclusion has to be made clear.

Sex not recorded asterisk below the table for 5 children: The asterisk should be in the 2nd row after total number of participants, i.e., 96, as the numbers of male/female in that row don’t match the total.

My conclusions: The authors should re-design their tables, bearing in mind that each table should be self-explanatory. That is, they should not depend upon the readers reading the text to be able to understand. Headings, sub-headings, and sub sub-headings might be helpful. Just a suggestion.

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

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

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

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

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

Are the conclusions drawn adequately supported by the results?
Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Child Neurology, Neurodevelopment, Neuroepidemiology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.
Charles R. Newton
Kenya Medical Research Programme-Wellcome Trust Collaborative Programme, Kilifi, Kenya

This well-written paper describes the adaption and testing of the Parental Evaluation of the Developmental Status: Developmental Milestones (PEDS: DM) in 96 children (aged 3-8 years) who attended ambulatory clinics in urban Bhutan. The investigators found a high prevalence of developmental delay (59%), particularly in receptive language (59%) and expressive language (76%). They conclude (correctly in my view) that the PEDS: DM may not be culturally applicable to assess children in Bhutan.

It is not clear why the investigators choose to examine this tool in this setting when there are other screening tools e.g. Rapid Neurodevelopmental Assessment, Malawi Developmental Assessment Tool. A justification needs to be made more explicit.

There is little information provided on the adaption of the tool to the circumstances in Bhutan. There is a well-established process of adapting tools to different cultures and circumstances (van de Vijver and Tanzer, 2004).

The children were recruited from a clinic in a national referral hospital, and thus it is unclear how representative they are of the population. Most evaluations were conducted in English, suggesting that the population sampled were the more educated population. No data is provided on the socio-economic status of the parents or the maternal education.

The influence of sex does not appear to have been taken into consideration during the analysis. There was no information on the psychometric properties of the adapted PEDS: DM in Bhutan.

References

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

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

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

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

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

Are the conclusions drawn adequately supported by the results?
Partly

Competing Interests: No competing interests were disclosed.
Reviewer Expertise: Neurodevelopmental disorders; epidemiology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response 15 Aug 2019

Farrah Mateen, Harvard Medical School, Boston, USA

“This well-written paper describes the adaption and testing of the Parental Evaluation of the Developmental Status: Developmental Milestones (PEDS: DM) in 96 children (aged 3-8 years) who attended ambulatory clinics in urban Bhutan. The investigators found a high prevalence of developmental delay (59%), particularly in receptive language (59%) and expressive language (76%). They conclude (correctly in my view) that the PEDS: DM may not be culturally applicable to assess children in Bhutan.

It is not clear why the investigators choose to examine this tool in this setting when there are other screening tools e.g. Rapid Neurodevelopmental Assessment, Malawi Developmental Assessment Tool. A justification needs to be made more explicit.”

Thank you for making us aware of the other options available. We were not aware of such developmental screening tools at the time of this study. We now provide some examples of other relevant tools at the end of discussion.

“There is little information provided on the adaption of the tool to the circumstances in Bhutan. There is a well-established process of adapting tools to different cultures and circumstances (van de Vijver and Tanzer, 2004).

The children were recruited from an outpatient clinic in the national referral hospital. They represent a convenience sample only. It is unclear how representative they are of the full pediatric population. Most evaluations were conducted in English, suggesting that the population sampled were a more educated population. No data were queried on the socio-economic status of the parents or on the level of maternal education.”

We plan to include this citation in the revised version.

We now discuss the potential for selection bias in our sample. You are correct that the sample is limited to a population living within travelling distance of the major urban center of the country. Since there is only one national hospital, it is possible that many children traveled long distances but more likely, these children have a marginally higher socio-economic status when compared to the rest of the country, which is primarily rural with less access to healthcare. Children were specifically selected as receiving routine vision screening, and maternal education likely play a part in which parents decide to seek such care. However, the entire health system is public, so all children would be seen free of direct costs.

“The influence of sex does not appear to have been taken into consideration during the analysis. There was no information on the psychometric properties of the adapted PEDS: DM in Bhutan.”
The following text will be included in results regarding gender distribution:

The female preponderance in our sample is thought to be a spurious finding. The percentage of population of males to females ages 5-9 years at time of study was 50.9% male to 49.1% female. To our knowledge there are no gender disadvantages at the pre-school or early school years. Nor are there known cultural considerations that may favor school age females for receiving routine medical care over males.

**Competing Interests:** None

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**Reviewer Report 02 August 2019**

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Philip Wilson
Centre for Rural Health, University of Aberdeen, Inverness, UK

Thank you for asking for an opinion on this interesting paper. It is a useful contribution to the world literature on developmental assessments and raises some important issues about the transfer of tools across cultures. The key finding is that, when using the PEDS: DM, a far higher proportion of children in Bhutan aged 3-7 years fail to meet expected milestones compared to children raised in the USA. The reasons for many such ‘failures’ lie in societal child-rearing practices described by the authors and are relatively easy to understand. Most would be relatively easy to remedy and the authors give valuable insights into how this might be achieved.

I have two significant comments to make about the paper, and a few minor points which are largely stylistic.

1. The first major point is that it is not clear how representative of the general population of Bhutan (or indeed of the locality from which the sample was drawn) the 96 children are. Is the female preponderance a chance finding? Is the ophthalmological screening programme from which the children were drawn free of charge? Is it well attended, apart from by children with severe disabilities? Are parents with worries about visual abilities of their children more likely to attend? These sorts of concerns are known to be more common among parents of children with autism, for example. There is some discussion of these matters on page 7 in the Limitations and Strengths section, but a much more critical analysis of the potential for selection bias and its possible contribution to the findings is required. It is well understood that a completely representative sample is extremely difficult to obtain in lower income countries but readers will want to have some sort of idea about the proportion and nature of the potentially eligible population who would not have attended the clinic.

2. My second comment is about the use of the word screening. It is commonplace to refer to developmental screening, but no neurodevelopmental assessments have to date met standard WHO screening criteria - largely because there is little evidence that early intervention is any better
than late intervention for most problems in most of the tested domains. Screening is most appropriately used for one-off assessments conducted in the context of an organised programme: ‘surveillance’ more closely reflects what is being done here. The authors might be interested in the discussion of this matter in a recent paper comparing child developmental assessments in five countries (Wilson et al., 2018).

More minor points:

- I found Table 2 a little difficult to follow. The use of parentheses is confusing and the final column on the two-stage study does not make sense until the reader reaches the Discussion section.

- Ages would be best expressed in Months in both Table 2 and Table 3, and probably elsewhere in the text.

- The legend to Figure 1 requires more information including total n.

- The discussion about language assessment on page 6 should perhaps reference the literature on bilingualism (I assume that at least some of the children fall into this category). Young children reared in bilingual households commonly appear to have delayed language acquisition in each language but eventually achieve fluency in both.

- There are some minor typographical issues, such as in the first sentence of the Results section of the Abstract and the second sentence of the Strengths and Limitations section.

References


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

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

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

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

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

Are the conclusions drawn adequately supported by the results?  
Partly

Competing Interests: No competing interests were disclosed.
Reviewer Expertise: Developmental assessment of children, primary care, epidemiology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.