Disability from Japanese encephalitis in Cambodia and Viet Nam

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Summary
A cohort of Japanese encephalitis (JE) survivors in Cambodia and Viet Nam were assessed at least 4 months after hospital discharge in order to understand the extent of disability after JE. We used a simple assessment tool which focuses on the impact on daily life. In total, 64 disability assessments were conducted: 38 in Cambodia and 26 in Viet Nam. In Cambodia, 4 (11%) children had severe sequelae, suggesting the children would likely be dependent, 15 (39%) had moderate sequelae and 17 (45%) had mild sequelae. In Viet Nam, two (8%) persons had severe sequelae, five (19%) had moderate sequelae and eight (31%) had mild sequelae. In many JE-endemic areas there are no multi-disciplinary teams with sophisticated equipment to assess patients after JE disease. This assessment tool can assist with patient management and generate data to support the need for programmes to prevent disease and improve outcomes for survivors.

Key words: Japanese encephalitis, disability evaluation, burden of illness.

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Introduction
Japanese encephalitis virus (JEV) is the most important cause of encephalitis in children in Asia based on frequency and severity [1]. About 10–30% of patients die and there typically is a high rate of sequelae among survivors [2].

Japanese encephalitis (JE) sequelae research usually focuses on identifying specific types of impairment such as motor deficits or psychiatric problems. This information does not always clearly indicate the extent a disability affects daily life. The studies usually require specialized equipment and medical staff, and each assessment may take many hours [3, 4]. Recognizing the need for a simple and meaningful method to measure disability among JE survivors, in 2006 University of Liverpool researchers in the
United Kingdom and colleagues in Asia developed the Liverpool Outcome Score [5]. This assessment tool can be used in a wide range of settings by non-specialist health professionals, and the outcome measure focuses on the likelihood a person will be able to live independently after illness.

The high percentage of JE survivors that experience life-long disabilities means the disease’s impact is substantial, even though absolute numbers of cases and deaths are lower than some other childhood diseases. Data that demonstrate the disease’s impact can be powerful to support advocacy for preventive immunization programmes, and for programmes to improve outcomes for survivors. Two countries in Asia, Cambodia and Viet Nam, used the Liverpool Outcome Score to measure disability in a cohort of JE cases in their respective countries. The objective was to understand the extent of JE-related disability, focusing on the impact on daily life.

Methods

Participants and setting
Participants with laboratory-confirmed JE were identified through routine surveillance system databases in Cambodia and Viet Nam. In Cambodia, there are six JE sentinel surveillance sites: two pediatric hospitals and the pediatric wards of four provincial hospitals. In Viet Nam, two provincial hospital surveillance sites were included, one each in the north and the south of the country.

In both countries, patients who present at a surveillance site with acute encephalitis or meningoencephalitis routinely have diagnostic testing conducted. JE is confirmed by detection of anti-JEV immunoglobulin M (IgM) antibody in cerebrospinal fluid or serum by an IgM antibody capture enzyme-linked immunosorbent assay (ELISA). The Panbio JE–Dengue IgM Combo ELISA was used in both countries [6]. In Viet Nam, the National Institute of Hygiene and Epidemiology’s in-house ELISA was also used.

Conduct of the assessment
In Cambodia, JE cases that occurred from June 2006 through July 2007 were included, and assessments occurred from December 2007 to March 2008. In Viet Nam, cases from October 2006 through July 2008 were included, and patient follow-up was conducted from October to December 2008.

As selection of cases for inclusion occurred retrospectively, the period varied between each patient’s discharge from hospital and assessment. However, all assessments took place at least 4 months after discharge. This interval was considered appropriate because a deterioration or improvement in condition may occur in the weeks following the acute illness but after this is uncommon. A similar study demonstrated that 18 (75%) of 24 patients assessed from 3 to 6 months after hospital discharge had the same score when the assessment was repeated later [7].

Physicians or other health professionals in each province conducted the assessments after being trained on use of the Liverpool Outcome Score. Assessments were usually conducted at the patients’ homes. Activities were conducted as part of routine disease surveillance activities and ethics approval was therefore not required.

Assessment tool
The Liverpool Outcome Score consists of a series of questions posed to a parent or caregiver and observations of the case performing selected actions. The tool aims to be uncomplicated to implement, widely culturally appropriate and easy to use by non-specialist health professionals. Its development has been described elsewhere, and the tool is freely available [5, 8]. In brief, there are 15 assessment items that evaluate basic motor and self-care skills, as well as simple cognitive and behavioural functioning (Table 1). Several items require a comparison of the case’s ability with that of his or her peers in the community. For each item, the person receives a score based on the grading scale for that question. The final outcome score for each individual is the lowest score received for any question, and reflects the likelihood a case will be able to live independently (Table 2).

In both countries, the assessment tool was translated and pretested prior to use. An accompanying user’s manual was available.

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Items evaluated with the Liverpool Outcome Score assessment</th>
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<tbody>
<tr>
<td>A. Questions to the parent or caregiver</td>
<td>1. Speech or communication skills</td>
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<td></td>
<td>2. Feeding ability</td>
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<td></td>
<td>3. Whether the case can be left alone without coming to harm</td>
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<td>4. Behaviour</td>
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<td>5. Recognition of people</td>
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<td>6. Performance at school, work, or in routine activities at home</td>
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<td>7. Occurrence of seizures</td>
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<td>8. Dressing ability</td>
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<td></td>
<td>9. Bladder and bowel control</td>
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<td>10. Hearing</td>
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<td>B. Observations</td>
<td>11. Sitting</td>
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<td></td>
<td>12. Standing up</td>
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<td></td>
<td>13. Walking</td>
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<td></td>
<td>14. Putting hands on head</td>
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<td></td>
<td>15. Picking up an object (pincer grip)</td>
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</tbody>
</table>
Data management
Data were entered and analysis was conducted using Excel (Microsoft Corporation, Seattle, Washington, USA) and Epi Info version 3.3.3 (US CDC, Atlanta, Georgia, USA).

Results
In total, 64 disability assessments were conducted: 38 in Cambodia and 26 in Viet Nam. In Cambodia, 50 JE cases had occurred during the selected time frame. However, seven patients had died during the acute JE illness or following discharge from hospital, and five survivors could not be contacted. Overall in Cambodia, 38 (88%) of the 43 surviving JE patients were assessed for disability. In Viet Nam, 2 of the 29 JE cases had died and one was lost to follow-up. Therefore, 26 (96%) of 27 surviving JE patients were assessed for disability. In both countries, each participant’s assessment took from 15 to 30 min.

Cambodia
The median age of the 38 children assessed was 7 years (range 1–15 years), and 25 (66%) were male. The mean time interval between acute JE illness and assessment was 12.8 months (range 4.4–20.2 months).

Almost all children had sequelae at different levels of severity. Four (11%) had severe sequelae (score = II), suggesting a likelihood the child would be dependent. Fifteen (39%) had moderate sequelae (score = III) and 17 (45%) had mild sequelae (score = IV) (Fig. 1).

The four children with severe sequelae were all younger than 9 years of age. All scored II for one or two items, and three also scored III or IV in one to six other items. The severe sequelae included seizures on most days, incontinence or needing assistance with toileting, severely abnormal behaviour, always needing help with dressing, or inability to be left alone without coming to harm; the latter four items were all inappropriate for the child’s age.

The most common sequela overall was behavioural problems; 31 (82%) of the 38 children had behaviour that was abnormal. For 26 (68%), behaviour was aggressive or the child easily became angry. Education also was commonly affected. Three (8%) of the thirty-eight children had dropped a school grade or could not attend school. Ten (26%) were not doing as well at school or, if they were not school attendees, were not able to perform the same tasks at home as effectively as before illness.

Viet Nam
The median age of the 26 persons assessed was 14 years (range 2–73 years), and 18 (69%) were male. Assessments were conducted a mean of 14.2 months (range 4.0–24.0 months) after acute JE illness.

Overall, two (8%) had severe sequelae (score = II), five (19%) had moderate sequelae (score = III) and eight (31%) had mild sequelae (score = IV) (Fig. 1). The two children with severe sequelae, who were likely to be dependent, were 10 and 15 years of age. One of these children was unable to stand or walk and required assistance with feeding, toileting and dressing; this child scored II or III in 9 (60%) of the 15 assessment items. The other child was having daily seizures and had behavioural problems.

The most frequent sequela among the 26 cases was behavioural problems, identified in 10 (38%) cases. School or work performance had deteriorated in 5 (19%) of the 26 cases, and the child with multiple severe sequelae was no longer attending school.

Discussion
These assessments demonstrate that severe disability is common after JE, with about 10% of JE patients in both countries having sequelae that were serious enough to be incompatible with independent living. Furthermore, the majority of survivors had sequelae; even those with good physical recovery often had...
more subtle sequelae, such as behavioural or cognitive problems. These sequelae are unlikely to make the person dependent, but nonetheless could affect daily life.

The high rate of patient follow-up (88–96%) was the strength in the conduct of these assessments, and provides confidence that the results are broadly representative of patients’ outcomes in these countries. There were some potential limitations. Ideally, all assessments would have been conducted at the same interval after illness. Some assessment items addressed relatively subjective areas, such as school or work performance, with responses possibly reflecting the perceptiveness of the caregiver. The assessments were also conducted at eight different sites. However, with acceptable inter-interviewer variability documented during the tool’s validation phase, this should have had minimal impact [8]. Finally, the two countries’ results are not comparable because the age ranges of the cohorts were different.

The Liverpool Outcome Score was easily implemented. In many JE-endemic countries, there are no multidisciplinary teams with sophisticated equipment to assess patients. This easy-to-use tool can provide information for individual patient management as well as generate population-level data on disability after JE. The information can be used to raise awareness and understanding of JE-related disability and its full economic and social impact.

Awareness of the impact of JE will help to facilitate control. Ministry of Health staff in Cambodia used these data, along with surveillance and cost-effectiveness data, to support the decision to introduce a JE immunization programme, commencing in three provinces in 2009. Increased attention to the extent of JE-related disability also can be an important step in raising awareness of the need for improved long-term management of patients to minimize the handicaps caused by disability. In many places, follow-up of patients after JE disease is limited, and there is no advice on activities, or provision of devices, that may assist with rehabilitation and long-term care, despite the availability of resources appropriate for use in less-developed settings [9].

The Liverpool Outcome Score assessment tool has not yet been validated for use following other neurological illnesses. However, there is potential for use of this tool as part of a wider strategy to assess and follow up children following neurological disease. An integrated approach would result in system efficiencies and the availability of comprehensive information to reinforce the benefit of immunization programmes for the vaccine-preventable neurological diseases.

Disability is an important part of JE disease burden, it contributes to poverty, and its prevention is an important consideration when determining the potential benefits of a JE immunization programme, as the ability to avert long-life disability contributes to the cost-effectiveness of such programmes [10, 11]. To highlight the issue of JE-related disability, a global JE control plan currently under development will include country targets for implementation of policies or plans on assessment and rehabilitation of patients after JE [12].

References