International Journal of Epidemiology 1998,27.672–676

Comparison of key informant and survey methods for ascertainment of childhood epilepsy in West Bengal, India

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Background This study aimed to compare efficacy and cost of key informants and survey for ascertainment of childhood epilepsy within a treatment context in rural India.

Methods The study was set in a non-governmental, community programme for the functional and socioeconomic rehabilitation of children with disabilities in rural West Bengal, India. Ascertainment was by two methods: house-to-house survey of 15 000 households and also by 430 key informants including village leaders, health workers and 670 schoolchildren. Methods were compared for positive predictive value, and sensitivity by capture-recapture technique. Ninety four children were enrolled into treatment. Predictors of treatment success were determined by multiple logistic regression analysis, giving adjusted odds ratios for remission. The costs of identifying one case and one treatment success were measured by costing personnel, materials and overheads.

Results The survey was four times as sensitive as key informants although the positive predictive values were similar (36%, 40%). The survey had an absolute sensitivity of only 59%. Identification by key informants strongly predicted successful treatment outcome (odds ratio [OR] = 4.74, 95% confidence interval [CI] : 1.19–18.85). The cost of finding one case was US$11 and US$14, and of finding one successful treatment outcome US$35 and US$67 for informants and survey respectively. Key informants were essential in attaining longer term programme objectives.

Conclusions In the context of a treatment programme, key informants were the more cost-effective method, but community involvement was traded against low sensitivity in the short term. Overall ascertainment costs were significant in the context of primary health care in India.

Keywords Epilepsy, India, ascertainment, costs, key informant, children

Accepted 5 November 1997

Epilepsy is the single largest neurological problem facing developing countries today. Globally, 50 million people have epilepsy, and 33 million of them are children in developing countries, 90% of them untreated.[1] The World Bank ranks epilepsy higher than malnutrition as a health priority for school-age children both because of its high psychosocial morbidity and its potential for control by cost-effective, low technology solutions.[2]

A successful control policy for developing countries has to be affordable, sustainable, acceptable and effective. For 20 years, WHO has recommended a strategy integrating epilepsy services at the primary level and using low-cost drugs.[3] Primary level activities in India include the primary health centre (PHC) now a feature in almost every rural Indian district; the Integrated Child Development Scheme (ICDS),[4] a national programme integrating nutritional, health and educational services for children, mothers and pregnant women; and community based rehabilitation (CBR), a community level strategy building on local resources, which aims at equalization of opportunities and social integration of all people with disability.[5]

Previous studies of epilepsy ascertainment in developing countries have focused on estimating prevalence by 'key informants' (KI), and the house-to-house survey. KI can be defined as long-term residents who occupy positions of trust and respect and have roles that bring them into contact with individuals and groups in the local community.[6] KI have been used extensively in developing countries for qualitative studies

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and rapid community health assessments.[6,7] The major criticism of the KI method has been lack of sensitivity. In a Jamaican study, sensitivity for epilepsy was less than 12% reportedly because of low levels of health care staff and referral facilities.[8] A Kenyan KI prevalence study had sensitivity five times less than a random cluster sample survey.[9,10]

The two-stage survey is often the preferred method of community ascertainment of generalized tonic-clonic seizures and several questionnaires exist for their detection.[11,12] Surveys are expensive in terms of money, time, opportunity, planning and supervision. In Kenya the cost of locating one case was US$37.70 by survey, seven times more than by KI.[10] A commercially funded epilepsy study in Ecuador employed 275 people to screen a population of 75 000 and took over 3 years from planning to data collection,[13] clearly impractical to replicate in other developing countries. Surveys in developing countries have also been criticized as a culturally alien method, treating the community as passive observers, often inaccurate, and sometimes arousing unrealizable material expectations.[14]

We started a community based epilepsy service for children aged 2–18 years integrated into an existing CBR framework in rural west Bengal. Our aim was to offer a low cost, primary level service to reduce children's seizures and their attendant risks, to improve their developmental status, and to reduce social, educational and employment disadvantage. Our longer term aims were to develop families' capacity to analyse and change their socioeconomic situations. The ascertainment exercise was therefore designed to recruit children with active epilepsy into an intervention programme, not as a formal prevalence study. The objective of this paper is to compare the sensitivity, efficacy and costs of KI and survey methods of ascertainment within a treatment context.

Methods

Geography

The study took place in 46 villages of Bishnupur Blocks I and II in district 24 Parganas South, a flat, rural district south of Calcutta of population over 120 000 and area 213 km^2. Income is seasonal, mainly from farming, per capita GDP $380, female literacy 36% and infant mortality ratio 65 per 1000 livebirths. The crude population growth rate is 20 per 1000 per year.[15]

The area has one government hospital, two primary health centres, an ICDS programme, and a considerable choice of private allopathic and traditional practitioners. The epilepsy service was offered jointly by two non-governmental organizations (NGO), one working in children's CBR (SANCHAR-AROD), the other the main facility for maternal and child health (Child-in-Need Institute).

Key informant method

Local figures of authority and special groups known to the NGO in 18 villages were approached as KI by the authors between August and October 1995. We were invited to village development forums (VDF), where villagers discuss ways of improving their environment, and spoke with 51 women and teenage girls at mahila mandals (women's groups) and adolescent centres. We wanted to sound out local views about epilepsy and convince people that we were offering a worthwhile service.

Table 1 Survey questionnaire, West Bengal, 1995–1996

How many children are there in the family between 2 and 18 years of age?
Has any one of them ever had these problems:
1. Sudden jerking or shaking of arms, legs or face which the child could not stop themselves?
2. Suddenly fell over and lost consciousness?
3. Suddenly fell over and bit their tongue?
4. Experienced any of the following: suddenly lost touch, didn't notice what was going around him, at this time experienced a bodily sensation, e.g. smell which no one else did?
5. Had khichuni, khach, mrigi or fit? Did anyone say that's what it was?

All 73 NGO health and disability workers and 20 balwadi (nursery) workers were trained to enquire about epilepsy, counsel and refer appropriately. Routine health records were examined.

We were also interested to find out children's views and whether they knew of affected children in their neighbourhoods. We used a drama sketch with a simulated seizure to generate discussion in 11 primary schools amongst 670 10-year-olds.

House-to-house survey

A five-item questionnaire, adapted from an extensively validated questionnaire used in Ecuador,[12] translated into local Bengali dialect and back translated (Table 1), was used in a two-stage house-to-house survey of 38 villages. Sensitivity and specificity were not validated since its predictive value in a sample with epilepsy would differ from a general population, and no suitable validation sample existed in rural West Bengal; secondly, its pragmatic use was to identify children eligible for treatment and its face validity was judged satisfactory after field testing.

Eight local young adults with secondary education (five men, three women) were given a basic orientation on epilepsy and community survey work. They administered the questionnaire to mothers of children or other adults in the household, and enumerated all children in the field area aged 2–18 years between October 1995 and February 1996. Any positively answered item counted as a positive screening result. Probe questions about associated fever or prior external stimulus were added to improve positive predictive value. Fieldwork was monitored daily.

All children screening positively were interviewed at home by a paediatric neurologist and senior disability worker. Children with epilepsy were invited to attend clinic. Those with other diagnoses were counselled and referred if necessary.

Costs of personnel, training, stationery and travel (but excluding management costs) were calculated, substituting the cost of verification using a local neurologist.

Operational definitions

Operational definitions for seizures, epilepsy and status epilepticus were as used in an international study in Ecuador[13] or, for non-epileptic conditions, based on the work of Stephenson[16] and these definitions were used by the paediatric neurologist in the second stage of screening. Active epilepsy eligible for treatment was defined as two or more unprovoked seizures, or one episode of status epilepticus, in the previous 12 months.

Data handling

Paper records were transferred to a computer database. The odds of treatment success was calculated in a multiple logistic regression model using Stata 5.0 for Macintosh statistical software,[17] adjusted for age, sex, cerebral impairment, prior treatment, lifetime seizures, and dropping out.

Results

Informants varied in knowledge, accuracy, and geographical coverage. Disability workers suggested half of all names, the rest were suggested by villagers at VDF. Local beliefs were that many people with epilepsy died, few got married or led independent lives, and that treatment was expensive and required visits to the city. Epilepsy was sometimes synonymous with madness. Women's groups and adolescents described the stigma, especially for women and girls, and were concerned about febrile convulsions. Teachers and schoolchildren did not suggest many names and very few had ever witnessed a seizure. Health workers and health records provided no useful information.

Reaction to the survey was mainly positive, although there were a few misconceptions, for example that the survey workers represented the government, the bank, or a child sponsorship agency. Some remote neighbourhoods could have been missed since we had no maps or up-to-date population records. As a check, we found our demographic data to be consistent with census statistics of 1991 adjusted for population growth. There was a negligible non-response rate at the first stage of screening (<0.1%). The denominator child population was estimated in those villages where only the KI method was used, using the fraction derived from enumeration.

Table 2 summarizes the results of screening, both methods used in 16 of the 46 villages. A total of 334 children screened positively, 17 by both methods. Seizures and non-epileptic events were diagnosed using the operational definitions referred to above.

Verification

A quarter (16) of 67 children identified by informants were untraceable from details given. Ten had non-epileptic disorders whilst 26 were either normal or had asthma or muscle cramp, the Bengali word for which is the same as for 'tonic'. Twenty seven had epileptic seizures, nine of whom were missed by the survey.

The majority (72%) screened positively by the survey had suffered some disturbance of consciousness and the diagnosis of epileptic seizures was confirmed in 101 of 284 children. The great majority of respondents were mothers, but other family members including older siblings and grandparents sometimes acted as proxy respondents for the index child. The positive predictive value of the questionnaire was 36% and enhanced by probe questions to 39%, similar to that for KI (40%). Question 4 was least predictive, identifying daydreaming children. The probability of being a true case was increased if question 5 (Has your child ever had epilepsy?) or more than one question was answered positively.

Table 3 shows that where both methods were used, the sensitivity of the survey was 3.5 times higher than that of KI (59 versus 17). It is unlikely that prior ascertainment bias (the KI method preceded the survey) accounted for this magnitude of difference. Using the capture-recapture technique to calculate underascertainment by each method,[18] the absolute sensitivity of the survey was estimated as only 59%.

Table 2 Comparison of ascertainment methods, West Bengal, 1995–1996

	House-to-house survey	Key informant method	Methods combined (%)
Villages screened	38	18	46
Total population	108 795[a]	68 468[a]	117 349[a]
Total 2–18 years	37 616	23 673[b]	40 574[b]
Screen positives	284	67	334
Anoxic seizures	18	–	18 (5)
Breathholding attacks	23	5	27 (8)
Febrile convulsions	11	2	13 (4)
Vasovagal syncope	35	–	35 (10)
Unconsciousness undefined	17	3	19 (6)
Normal	45	6	51 (15)
Other	27	8	33 (10)
Untraceable	7	16	22 (7)
All epileptic seizures	101	27	115
Active untreated epilepsy	80	19	90
Inactive epilepsy	10	3	11
Treated epilepsy	10	4	12
Single seizure	1	1	2
Seizures: positive predictive value (95% CI)	36% (32.7–38.4)	40% (34.3–46.3)	34% (31.8–37.0)
Total seizure cases[c]	108	37	132
Prevalence of seizures (95% CI)	–	–	3.3 per 1000 (2.97–3.53)
Prevalence of epilepsy (95% CI)	–	–	3.2 per 1000 (2.87–3.43)
Prevalence active epilepsy[d] (95% CI)	–	–	2.6 per 1000 (2.31–2.81)

[a] 1995 estimates from 1991 census figures using crude population growth rate 2.0%.

[b] Estimated

[c] Includes those traced from clinic.

[d] Active untreated epilepsy. two unprovoked, afebrile seizures in previous 12 months.

Table 3 Identification of children with epilepsy in villages where both methods were used, West Bengal, 1995–1996

	Identified by key informant	Not identified by key informant	Total
Identified by survey	10	7	17
Not identified by survey	49	34	83
Total	59	41	100

Epileptic seizures

One hundred and thirty two children had a history of epileptic seizure: 4 had single seizures, 11 had fewer than two in the previous 12 months (inactive), and 13 were on treatment at ascertainment time. One hundred and four (90%) of those with

active epilepsy were not on treatment. Hence an estimated minimum prevalence of epileptic seizures of 3.3 per 1000 (95% confidence interval [CI] : 2.97–3.53 per 1000) and of active epilepsy of 2.6 per 1000 (95% CI : 2.31–2.81 per 1000). Using the ILAE definition of active epilepsy, at least two seizures with one in the last 5 years regardless of treatment status, the prevalence of active epilepsy would be 3.2 per 1000 (95% CI : 2.87–3.43 per 1000).[19] Adjusting for survey sensitivity over the whole field area raised the estimated prevalence of active epilepsy to 5.52 per 1000 (95% CI : 3.80–7.25 per 1000).

Thirty-six per cent of children had primary generalized tonic-clonic seizures (GTCS), a further 20% GTCS with uncertainty about primary onset or secondary generalization. Nineteen per cent of children had only partial seizures, a further 23% with secondary generalization. One child had absence seizures, one had myoclonic seizures and five had multiple seizure types. One third had associated impairments such as mental retardation or cerebral palsy. Sixty per cent of cases identified by KI had more than one seizure per month or an associated cerebral impairment.

Time and costs

Visits to KI took 48 person-days, using two senior field workers and a doctor. The survey required 555 person-days using eight survey workers. Verification required a doctor and a senior field worker to visit the home. The ascertainment cost per case was Rs393 ($11) and Rs499 ($14) for KI and survey methods respectively.

Predicting treatment success

After one year of treatment, 28 (of 94) children achieved remission in the final 3 months. The adjusted odds ratio (OR) for remission for children identified by KI was 4.74 (95% CI : 1.19–18.85), and by survey, 0.43 (95% CI : 0.12–1.55). Eight children identified by survey dropped out early: six whose parents objected to the diagnosis of epilepsy, and two more who had expected child sponsorship.

Discussion

Our prevalence estimates were consistent with other studies in India,[20,21] and the distribution of seizure types was entirely as expected. The capture-recapture method indicated a significant shortfall in survey sensitivity: some parents had not immediately declared their child to have epilepsy but instead opted to 'wait and see' what the new service was like, before deciding to come for consultation. This introduced the element of perceived service quality as a part of a feedback loop into the ascertainment process.

There were several other possible causes of underascertainment. Probably, some households deliberately concealed epilepsy because of stigma. This was more true for girls whose marriage prospects could be harmed by declaration. This belief was supported by the dropout of six families. Proxy response bias, compounded when the respondent was not the parent or usual guardian, was another possibility. A small number may have been misclassified as unconsciousness of undefined cause when clinical history did not permit a definitive diagnosis. Misclassification was minimized by the use of standardized operational definitions by a single investigator. Recall bias could reduce ascertainment of seizures which occurred many years ago, had a short remission time, or were of subtle types, none of which were important from the treatment perspective. Lastly, the interviewer might have had an indirect effect on response, for example a lone young wife may have replied negatively to questions to expedite the departure of a male survey worker, or the family might have judged the worker too young to receive such sensitive information.

Table 4 Relative costs of ascertainment of childhood epilepsy, West Bengal, 1995–1996

	House-to-house survey	Key informants
Cases identified	108	37
Person-days required	555	48
Total cost of first stage screening	Rs41 941 ($1200)	Rs10 420 ($300)
Cost of identifying one screen positive	Rs388 ($11)	Rs282 ($8)
Cost of screening and verifying one case	Rs499 ($14)	Rs393 ($11)
Cost of identifying one treatment success	Rs2343 ($67)	Rs1212 ($35)

KI tended to detect more visible cases and although four times less sensitive than the survey, the positive predictive value was the same, false positives being mostly due to unlocatable addresses (rural Indian dwellings often have no precise addresses), rather than non-epileptic conditions. KI sensitivity was low, probably because epilepsy is generally less apparent than sensory, locomotor or cognitive impairments. KI were notably more useful in this study than in Jamaica, probably due to a higher number and spread of informants, and awareness of and support for the new service.

Teachers and schoolchildren were not useful sources, probably because affected children were unlikely to attend school, and perhaps also because schoolchildren might not (or indeed might) have been party to concealment. Balwadi teachers mostly identified preschool children with febrile convulsions. Both KI, and the far-reaching publicity produced by the survey, generated continuing referrals.

In a treatment context a method that detects just a few cases is inefficient, and inequitable for children with epilepsy who are not detected. On the other hand, a highly sensitive screen may carry unreasonable temporal and financial costs; detect and label large numbers of children and families with benign conditions, or with infrequent or benign epilepsies who derive borderline medical benefit from treatment, and perhaps for whose families the cost of attendance outweighs the perceived benefit. It is also difficult to measure the indirect costs of screening such as forcing some parents to declare their children's epilepsy, and possibly mislabelling true negatives.

The unit costs, of around Rs400 (US$11) for KI and Rs500 (US$14) for survey detection, were low in comparison to Ecuadorian and Kenyan studies, the latter underestimated by not including the full cost of verification or medical time. Our low costs were partly explained by rapid ascertainment in a densely populated and accessible area. Although cheaper than survey, the opportunity cost of using senior disability workers in the KI method was high and the unit cost was higher than had been

expected; excluding a doctor in the first stage could reduce costs to Rs200 (US$6) and still be significant, but with unknown effects on sensitivity and community acceptance.

The ultimate aim of the ascertainment process was to provide comprehensive management for children with epilepsy in a community development framework. With this in mind, the predictive value of KI assumes considerable importance. Children so identified were over four times more likely to succeed in the treatment programme. The reasons for this effect are unclear. One can speculate that these families were known to KI because they were well motivated and therefore likely to comply with treatment. The KI method costs Rs1212 (US$35) to identify one treatment success, whilst the survey costs Rs2343 (US$67) per treatment success. Therefore, using KI was ultimately twice as cost effective as the survey, although this advantage has to be set against a lower sensitivity.

Our involvement with the community did not go beyond discussion with community leaders, and groups of women and children. We did, however, plan close involvement of families in helping children achieve their educational and social potential. Many of our objectives were focused on them, with wider community involvement a means to achieving these. Although the survey generated publicity it played no part in these forward objectives. The KI method gained the sanction of community leaders, and facilitated subsequent dialogue towards social change, for example in enabling children with epilepsy to enter school. Talking to key informants also helped us to tailor our intervention, for example with regard to counselling parents and training health workers.

Previous studies of epilepsy ascertainment have focused on prevalence estimation as their objective, with sensitivity and specificity the hallmarks of efficacy. This study of ascertainment within a treatment context has shown even a well-performed survey has limited sensitivity, and was not entirely acceptable to the community. KI on the other hand, were invaluable as a means of involving selected community groups in the programme and of giving insight into, and attaining, its longer term purposes. Informants also maintained lines of referral and were able to predict treatment success in a way that is not completely understood. Informants were a fifth less expensive in identifying children with epilepsy, and twice as cheap in identifying children who achieved remission. It seems therefore that within a primary level treatment context, where resources are limited, and sensitivity is of secondary importance in the immediate term, KI offer a cost-efficient method of ascertainment for epilepsy in developing countries.

Acknowledgements

Deb Pal was supported by a Wellcome Trust Research Training Fellowship. The educational work and computer purchase were supported by an International League Against Epilepsy grant. We thank all the staff of SANCHAR-AROD and the Child-in-Need Institute. We also thank Professor Brian Neville for his comments on drafts of this paper.

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