EPILEPSY IN RURAL SOUTH AFRICAN CHILDREN — PREVALENCE, ASSOCIATED DISABILITY AND MANAGEMENT

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Objective. To determine the prevalence of epilepsy and its associated disabilities in rural South African children aged 2 - 9 years.

Setting. Eight villages in the district of Bushbuckridge, Northern Province, South Africa.

Design. A two-phase design was used. The first phase involved screening children on a house-to-house basis by interviewing mothers or caregivers using an internationally validated questionnaire for detecting childhood disability in developing countries. The second phase consisted of a paediatric/neurodevelopmental assessment of the children who screened positive.

Results. A total of 6 692 children were screened; 722 (10.8%) had a paediatric evaluation and 49 (0.73%) had epilepsy. The lifetime and active prevalences of epilepsy in these children were 7.3/1 000 and 6.7/1 000 respectively. Associated developmental disability was recorded in 35 affected children (71.4%), including 8 (16.3%) in whom this was moderate to severe. More than a half of the children with epilepsy (57.1%) did not receive anticonvulsant medication.

Conclusion. The prevalence of epilepsy in the rural childhood population investigated is higher than that recorded in most similar studies from sub-Saharan Africa, and the poor utilisation of appropriate anticonvulsant treatment is cause for concern. This study highlights the paucity of relevant information on the epidemiology of epilepsy in South Africa and that the system available for its management, especially

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in rural areas, appears to have functional deficiencies. Appropriate research is needed to identify the problems in service delivery and to enable the planning and implementation of an appropriate primary health care-based system for the diagnosis and management of epilepsy in children.

Epilepsy affects individuals throughout the world irrespective of age, ethnicity, socio-economic class or geographical location. Because of its high prevalence in developing countries and the potentially severe consequences of the disorder, a World Health Organisation (WHO) Study Group recommended in 1978 that the control of epilepsy should receive top priority.[1,2]

Before 1978 most of the information available on the epidemiology of epilepsy in developing countries was derived from hospital-based studies.[3] Subsequently some data have been reported from community-based research undertaken mainly in rural populations, using a two-phase study design in which fieldworkers screen the population using an appropriate interview schedule, and then those screened positive are evaluated by clinicians. Although such studies have methodological limitations, they currently represent the optimal method for conducting such research.[3,4]

The prevalence of epilepsy increases with age. It is notably high between birth and puberty, during which period the morbidity of the condition is potentially at it highest level, especially in terms of the neurological and psychosocial development of the individuals concerned.[1,3-7] It is to be expected that morbidity will be exacerbated in circumstances in which appropriate management with anti-epileptic drugs (AEDs) is either unavailable or underutilised. Prevalence rates of epilepsy in childhood and adolescence vary greatly worldwide from 3.4/1 000 in Japan, to 11.2/1 000 in Mexico.[8] As far as the authors are aware, no information is currently available on the prevalence of epilepsy in rural South African children, the disabilities associated with this condition, or how it is managed.

Prevalence studies done on epilepsy provide information that is useful to health care planners for the development of control and prevention strategies.[1,7-9] The present study documents the prevalence of epilepsy, its associated disabilities and its management in rural South African children aged 2 - 9 years in the Bushbuckridge district, Northern Province. This research was undertaken as part of a broader study into the prevalence of five childhood disabilities (intellectual, motor, visual and auditory handicap and epilepsy). Preliminary results on the part of the study pertaining to intellectual disability have been published previously.[10]

STUDY POPULATION AND METHODS

The Bushbuckridge area is situated in the north-east of South Africa, and abuts on the western border of the Kruger National Park. This study was based in the Mhlala district, which is populated by approximately 152 000, mostly Tsonga-speaking people who live in 48 villages, including the major centre, Thulamahashe. Eight of these villages, spread geographically throughout the district and each served by a community-based rehabilitation worker, were chosen for this study. A female resident of the village was selected and trained to administer the internationally validated Ten Questions (TQ) questionnaire on a house-to-house basis in the village. The TQ questionnaire was developed specifically for use in developing countries and was designed to screen out children between the ages of 2 and 9 years with intellectual, motor, visual and auditory disabilities and epilepsy.[11,12] Children who screened positive on the TQ questionnaire were offered a paediatric examination, with a neurodevelopmental assessment if necessary, at subsequent clinic visits to the villages by the investigating team. This team included two neurodevelopmental paediatricians and a clinical geneticist with a neurodevelopmental background.

Those children considered on screening to have had seizures were assessed by the clinicians with specific regard to confirming the diagnosis of epilepsy, determining the type and frequency of the seizures experienced, and ascertaining the management methods used, including treatment by traditional healers. All this was done by obtaining a good history, as no seizures were witnessed. The children were then examined for associated disability and particularly developmental delay. Children who were not on treatment or appropriate medication were referred for management to Tintswalo Hospital (in Acornhoek), the nearest major medical facility.

Ethical approval for the study was obtained from the Committee for Research on Human Subjects of the University of the Witwatersrand.

DEFINITIONS

For the purposes of this study, epilepsy was defined as a history of recurrent, unprovoked seizures, occurring in the absence of an identified acute brain or systemic insult. However, they could occur subsequent to, or as a direct consequence of, such a cerebral insult. Single unprovoked seizures, febrile seizures and neonatal seizures were not included in this definition.[3,4,6,13]

The term 'active prevalence' was defined as the proportion of the population with epilepsy that had experienced a seizure within the preceding 2 years, or had recently been or were currently on AEDs. Lifetime prevalence was defined as that portion of the study population with a history of epilepsy.[1,13]

Diagnosis of the seizure type was made entirely on clinical

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history as no investigative facilities, including EEGs, were available. Given the limitations of the study, it was not possible to gather enough information for confirmation of the seizure types according to the International League Against Epilepsy classification.[14] Working from this classification, however, the seizure patterns were broadly divided into the two groups, namely partial or generalised. The classification of developmental disability was made according to the physician's guidelines of Thorburn et al.[15]

RESULTS

A total of 6 692 children were screened for childhood disability using the TQ questionnaire. Included were 3 575 children (53.4%) aged between 2 and 5 years, and 3 117 (46.6%) who were 6 - 9 years old. Subsequently 722 of these children (10.8%) underwent paediatric evaluation. Altogether 49 children (0.73%) were diagnosed as having had epilepsy, giving a lifetime prevalence in this sample of 7.3 per 1 000 children. The active prevalence of epilepsy, i.e. those children who had had a seizure within the preceding 2 years or who were on AEDs (45, 0.67%), was 6.7 per 1 000 children. The lifetime and active prevalence rates of epilepsy in 2 - 5-year-old children were 5.6 and 5.3/1 000, respectively, and for the age group 6 - 9 years, 9.3 and 8.3/1 000, respectively. Of the 49 children with epilepsy, 47 (95.9%) had generalised seizures and 2 (4.1%) simple partial seizures. The causation of the epilepsy was undetermined in 35 patients (71.4%); it was considered to have a congenital origin in 9 patients (18.4%), while 5 children (10.2%) had an acquired cause, including 4 (8.2%) in whom this was considered to have originated in the perinatal period.

The age and gender of the affected children are detailed in Table I. The male/female ratio of the affected children was 3:2. There were more affected children in the older (6 - 9-year) group than in the 2 - 5-year-old group, with a ratio of approximately 3:2.

Table I. Age and sex of children with epilepsy

Age (yrs)	Male	Female	Total (%)	Prevalence/ 1 000 children Lifetime	Active
2 - 5	13	7	20 (40.8)	5.6	5.3
6 - 9	17	12	29 (59.2)	9.3	8.3
Total (%)	30 (61.2)	19 (38.82)	49 (100)	7.3	6.7

Management sought for the treatment of the epilepsy is documented in Table II. No treatment at all was sought for 17 children (34.6%). When combined with those children who only consulted a traditional healer (11, 22.5%), this gave a combined total of 28 children (57.1%) who did not receive any AEDs. Of those 21 affected patients (42.9%) who received

Table II. Management sought for children with epilepsy

Type of management	N (%)
Western medical management only	11 (22.5)
Traditional medical management only	11 (22.5)
Traditional and Western	10 (20.4)
Nil	17 (34.6)
Total	49 (100)

AEDs, 8 (38.1%) used phenobarbitone and 1 (4.8%) used phenytoin, while the mothers of 12 patients were not sure what medication was prescribed.

Associated developmental disability was recorded in 35 children with epilepsy (71.4%). This included 8 children (16.3%) with moderate to severe developmental disability, of whom 3 also had microcephaly, 3 cerebral palsy and 1 dysmorphic features. Of these 8 children with severe disability, 6 (75%) were receiving AEDs. Mild developmental disability was documented in 27 epileptic children (55.1%), including 1 child with associated spina bifida and another with tuberous sclerosis. Of these 27 children with mild developmental disability, the majority (20, 74.1%) had received no medication for their epilepsy.

DISCUSSION AND CONCLUSION

Many of the prevalence studies done on epilepsy in rural communities in developing countries have employed a two-phase study design.[3,4] In the first phase of these studies the community is screened by means of interviews conducted by fieldworkers, while in the second phase a medical evaluation is done of those individuals who screen positive. Despite the basic commonality in the methodology employed, comparison of the data from these studies is fraught with problems. Differences that exist between studies include those associated with the definition and classification of epilepsy, the age range of the populations screened, and the particular screening tools utilised. Further problems encountered have included underreporting of epilepsy because it is stigmatised, and the exclusion of minor seizures. Notwithstanding these and other methodological problems, these surveys are useful in terms of obtaining information on the epidemiology of epilepsy.[3,4]

The present study was undertaken using the TQ questionnaire, which is designed and validated to screen for five serious disabilities (intellectual, motor, visual, auditory and seizures) in developing countries. Of the 10 questions, only one relates to seizures: 'Does the child sometimes have fits, become rigid or lose consciousness?'.[11,12] It has been documented that this tool has relatively high rates of false-negative results for epilepsy. In order to overcome this, previous studies using the TQ questionnaire evaluated samples of those screening negative, and the effect of the false-negatives was 'calculated'

into the final estimated prevalence rate using validated statistical methods.[11,12,16] It was not possible to undertake this last step in the present study and consequently our results should be considered as 'minimum' observed prevalence estimates.

The lifetime and active prevalence rates for epilepsy in this study sample, namely 7.3 and 6.7/1 000, respectively, for children between 2 and 9 years of age, are slightly higher than some of the rates reported in other sub-Saharan countries, although as noted above these studies are not absolutely comparable. In Nigeria, Osuntokun et al.[17] documented an active prevalence of epilepsy of 5.8/1 000 in infants and children between birth and 9 years old. The community surveyed was urban, from a small town of 20 000 people. In a pilot study carried out 5 years previously in a rural village 20 km from this town, Osuntokun et al.[18] had recorded an active prevalence of epilepsy in infants and children of 26/1 000. The authors ascribed this marked difference in prevalence in these two neighbouring populations to the presence of an effective primary health care system that emphasised the prevention of childhood infectious diseases, antenatal care and health education in the town. However, there were differences in methodology as well as in the definitions used.[17,18] Other data from sub-Saharan Africa include a low active prevalence of epilepsy of 3.5/1 000 in birth to 9-year-olds in rural Tanzania, and 2.9/1 000 in Kenyan children aged 6 - 9 years.[19,20] The latter figure is considerably lower than the active prevalence rate of epilepsy (8.3/1 000 children) among similarly aged children in the present study.

Three previous studies have assessed the prevalence of childhood epilepsy in developing countries using the TQ questionnaire and statistical methodology to compensate for false-negative responses obtained during the screening procedure.[12,16] In two of these studies, from Bangladesh and Jamaica,[12,16] the lifetime and active prevalence rates were 5.8 and 6.5/1 000 children and 5.2 and 5.8/1 000 children, respectively. These results are slightly lower than those documented in the present study, especially when one considers that the results obtained in this study were not corrected for false-negatives and should be considered as minimal observed prevalence rates. The third study, from Pakistan, had lifetime and active prevalence rates of 12.4 and 15.5/1 000 children, which is relatively high when compared with our findings.[12]

The sex-specific prevalence of epilepsy in this study documented the male/female ratio as being 3:2 for both the younger and older age groups, as well as overall. This is a reversal of the tendency seen worldwide and in sub-Saharan Africa where the prevalence of epilepsy in female children tends to be slightly higher than in males.[8,7,19] No apparent reasons for this sex-specific reversal of prevalence were immediately obvious.

The present study was limited by the lack of diagnostic tools available to the investigators and their reliance on information obtained from mothers and their children during one screening procedure and one clinical interview, together with the examination of the patient. The epilepsy could therefore only be classified into generalised (95.9%) and partial (4.1%). It is, however, probable that partial seizures, which may rapidly become secondarily generalised, may have been erroneously classified.[3,4] Similarly, partly because of the limitations in the methodology used in the study, a definitive cause for the epilepsy could be designated in only 14 patients (28.6%). Nevertheless, this figure is comparable to figures obtained in other studies from developing countries, in which a cause for epilepsy could be established in less than 40% of cases.[3,4] An acquired cause was recorded in 5 children with epilepsy (10.2%) and in 4 of these cases (8.1%) it was considered to be perinatal in origin. The extent to which perinatal problems result in epilepsy is uncertain, but it is notable that Osuntokun et al.[17] from Nigeria reported an abnormal perinatal history in 2 out of 101 individuals (2%) with epilepsy.

Conversely, figures as high as 11% and 14% have been reported from Brazil. These wide-ranging data, together with the figure of 8% from the small sample (too small to draw conclusions, but useful to indicate a possible trend) obtained in the present study, are indicative of the need for further research to clarify the situation.[4,7,17]

Developmental disability was present in a significant proportion of the epileptic children (35, 71.4%), but only in 8 (16.3%) was this moderate or severe. Of these 8 children, 7 had other clinical health problems in addition to the epilepsy. Six of them were receiving AEDs, suggesting that it was the multiplicity and severity of their problems that resulted in their receiving medication. In contrast, of the 27 children with epilepsy and mild developmental disability, the majority of whom had no other problem, 20 (74.1%) had received no AEDs. It remains a matter for conjecture as to what role the lack of medical management and the continuation of seizures played in the genesis of developmental disability in these children.

More than one-third (17, 34.6%) of the epileptic children in this study had not been taken to any type of health care service (Western medical or traditional) for management. Twenty-one children (42.9%) had been treated by a traditional healer, either alone or together with Western medical management. Over half of the epileptics (57.1%) had never received AEDs (Table II). It is not unusual for epileptics in developing countries not to receive effective orthodox Western anti-epileptic management.[1,9] In their treatise on epilepsy in developing countries, Shorvon and Farmer[4] detailed the many possible reasons for this situation. They included the limited availability of medical manpower and deficiencies in the training of doctors with regard to the management of epilepsy. They noted that the Western model of epilepsy management may also be

inappropriate, especially in a rural setting. This model requires that patients with seizures present to the health care system for diagnosis and long-term treatment with AEDs. Possible barriers to the utilisation of this system include community attitudes towards seizures. The disease may not be considered a problem that should present for management, either because its significance is not understood or because of stigmatisation. Even if management is desired, distance from facilities and cost of obtaining regular treatment and follow-up may be excessive. In addition, language and cultural differences may prevent some medical attendants from making a correct diagnosis. When a diagnosis is made and appropriate treatment instituted, barriers to its effectiveness still exist. AEDs need to be taken on a long-term basis and do not effect a cure immediately, as might be expected by the patient. Consequently treatment may be intermittent and only in response to crises. Finally, and possibly the greatest impediment to effective anti-convulsant treatment in the developing world, is the lack of a constant supply of AEDs at facilities easily accessible to the patients.[4]

While undertaking this study the authors observed that many of the barriers to effective anti-epileptic management, noted above, were potentially present in the villages of Bushbuckridge, thereby explaining how 57.1% of epileptic children had not received AEDs. It was noted that one specific local factor may be operational in this regard. Historically the management of epilepsy in the rural clinics of Bushbuckridge has been the function of the visiting psychiatric clinic. Nursing staff in the rural clinics suggested to the authors that this practice stigmatised epilepsy; they said that the parents of children with epilepsy did not like their children being categorised with psychiatric patients and were therefore less likely to keep follow-up appointments.

In conclusion, this study has documented the first data on the prevalence of epilepsy in rural South African children, the disabilities associated with this chronic debilitating condition, and the fact that in the rural setting of the Bushbuckridge district the orthodox medical management system available for its treatment is far from ideal. These data preface the need for further research in South Africa to delineate those factors, especially in rural areas, that mitigate against effective control and prevention of epilepsy. Thereafter an appropriate, simple, effective, primary health care-based management system that is acceptable and accessible to the communities it serves can be planned and implemented.

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