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A Neuroepidemiological Survey in Rural Bolivia: Background and Methods

Key Words

Epidemiological methods · Developing countries · Neurological diseases

Abstract

A door-to-door survey was carried out in rural areas of the Cordillera province, Santa Cruz Department, Bolivia. A cluster sample of 10,124 inhabitants was selected. The aim was to determine the prevalence of the most common neurological diseases (epilepsy, stroke, parkinsonism and peripheral neuropathy) in this population using a modification of the World Health Organization screening instrument. 1,130 subjects screened positive and were then investigated by neurologists. In this paper we describe the background and methods of the survey and the characteristics of the population.

Introduction

Studies on the magnitude of neurological diseases are limited. Most neuroepidemiological data come from surveys carried out in developed countries. Methodological approaches used in such studies are often unsuitable for developing countries, especially rural populations. Major difficulties are the relative absence of accurate case registers or medical records, the lack of sophisticated

technology, and the relative lack of specialty trained medical staff in urban centers [1]. Large-scale surveys have been carried out to assess the suitability of research protocols for measuring the prevalence of neurological disorders [2, 3].

We describe the background and methods of the first neuroepidemiological study performed in Bolivia, a two-phase survey to ascertain the prevalence of key neurological diseases. The disorders of interest were epilepsy,

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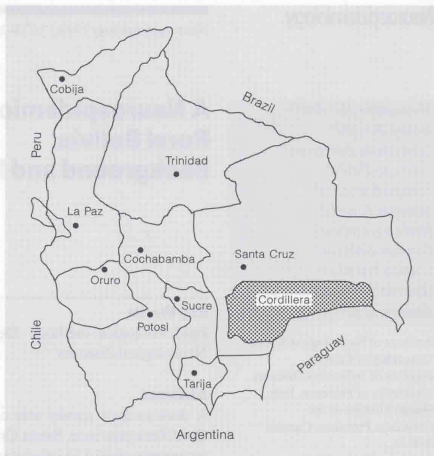


Fig. 1. Bolivia and Cordillera province.

peripheral neuropathy, parkinsonism and stroke. In Bolivia, as well as in other developing countries, epilepsy is a major health problem and the determination of its prevalence was the main goal of this survey. Data from this prevalence study served also as a basis for carrying out a case-control study to identify the relationship between the parasitic infections, cysticercosis and toxocarasis, and epilepsy.

The methodological approach we describe provides a useful strategy to determine disease frequency (prevalence) and to identify risk factors which can assist in developing disease control and prevention strategies. The prevalence and case-control study will be presented in subsequent articles.

Methods

Population

The study was carried out in the Cordillera province, Santa Cruz Department, in the south-eastern part of Bolivia. Bolivia has a total land surface of 1,098,591 km² and a population of 6,400,000, with a density of 5.8 persons per km². According to the Bolivian National Institute of Statistics (Instituto Nacional de Estadística INE) life expectancy at birth is 60 years, while infant mortality rate is 88/1,000 and 145/1,000 for urban and rural areas, respectively. The Santa Cruz Department covers 370,621 km², with a population of 1,359,383 inhabitants. The Cordillera province covers 86,245 km² and borders the Department of Chuquisaca, Tarija, and Paraguay (fig. 1). According to the Bolivian National Census [4], Cordillera province has a population of 88,628 inhabitants, 32,953 urban residents and 55,675 rural residents. The province is administratively divided into ten areas with 3,000–8,000 inhabitants each. Racially the population is a

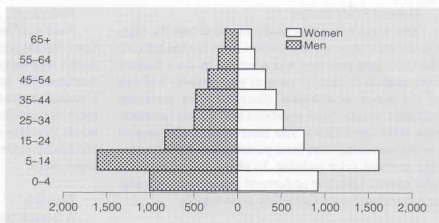


Fig. 2. Age and sex distribution of the study population.

Table 1. Age and sex distribution of the study population

Age years	Men		Women		Both sexes	
	n	%	n	%	n	%
0-4	997	19.9	921	18.4	1,918	19.2
5-14	1,593	31.6	1,605	33.0	3,198	32.3
15-24	827	16.2	749	15.2	1,576	15.7
25-34	485	9.8	521	10.5	1,006	10.2
35-44	471	9.3	445	9.0	916	9.2
45-54	323	6.6	318	6.4	641	6.5
55-64	206	4.0	198	4.1	404	4.0
65+	133	2.5	163	3.3	296	2.9
Total	5,035		4,920		9,955	

mixture of Mestizos, descendants of intermarriage between Spanish colonists and the native tribes (the Guaraní-Chiriguano), and approximately 30% pure Guaraní Indians. The majority of the population speak both Spanish and Guaraní while a minority speak only Guaraní. Agriculture and animal husbandry are the major occupational activities. The health-care infrastructure consists of a district hospital, nine area hospitals, and rural health centers.

The study was conducted with the agreement of the Bolivian National Department of Epidemiology of the Ministry of Social Welfare and Public Health and with the support of the Guaraní political organization (Asamblea del Pueblo Guaraní, APG).

Sample Selection

This prevalence survey served as the framework for a case-control study to estimate the association be-

tween cysticercosis, toxocaríasis and epilepsy. The sample size was calculated to provide sufficient cases for the latter study. A random cluster survey method was used, with each community constituting one cluster. Clusters were randomly selected communities, designed to represent approximately 20% of the population in each area. Urban areas, defined as a community with more than 2,000 inhabitants, were excluded from the sampling frame. In total we selected 10,124 people in 55 communities of whom 9,955 were effectively screened (table 1, fig. 2). Demographic data obtained from the Bolivian National Census combined with records at area hospitals allowed us to estimate that the rural population in 1994 was 54,324 [4].

Inhabitants were only eligible if they had been resident in the communities for the 6 months preceding prevalence day, November 1, 1994. The estimated point prevalence refers to this date.

General Study Design

This was a two-phase study: In phase one, the sample of rural communities selected from the ten areas of the Cordillera province was screened by door-to-door examination to identify persons who possibly had one of the target neurological disorders. The screening included standardized questions and simple examination tasks (see below). The interviewers who carried out the screening were local paramedical workers who had received prior training. In phase two all subjects who screened positive underwent a complete neurological examination performed by neurologists.

Screening Instrument

We adopted the Sicilian Neuroepidemiologic Study (SNES) screening instrument [5], a slightly modified version of the WHO Neuroscience Research Protocol [6]. In the SNES study the sensitivity of the screening instrument was 100% for parkinsonism, 96% for peripheral neuropathies, stroke and epilepsy; and the specificity was 86% [7]. The questionnaire had been translated into Spanish and pretested.

The instrument consisted of four main sections. The first concerned eligibility and demographic information, such as residency (on prevalence day), birth date, sex, relationship to the head of the family, education, occupation, hygienic condition, presence of latrine, household pig ownership, and pork consumption. The second section was a brief symptom questionnaire with precoded answers. Adults (i.e., persons 13 years of age or over) were asked directly about impairment of consciousness, uncontrolled limb movements, speech changes, palsy, mouth drooping, limb weakness or paralysis, limb sensory abnormalities, rigidity and bradykinesia, and tremor. Children (12 years of age or younger) were not asked questions directly. The child care giver, usually the mother or grandmother, served as the surrogate and answered questions on impairment of consciousness, uncontrolled limb movement and absence spells. The third section consisted of a brief physical examination based on simple tasks to assess arm strength, hand sensation, coordination, gait, standing balance with eyes open and eyes closed, and biceps muscle tone. This examination was restricted to persons 13 years of age or older. The fourth section concerned self-reported diagnoses of parkinsonism, epilepsy, stroke, and peripheral neuropathies.

A screening test was defined as being positive if one of the following occurred: (1) a positive response to any item of the symptom questionnaire; (2) an abnormal finding on a simple examination task, or (3) a self-reported diagnosis of one or more of the study diseases [5].

Field Staff

Field staff was composed of 4 neurologists, 3 infectious disease physicians, 2 local general physicians, the health representative of the APG and 26 paramedical workers. The health workers were selected from the Guarani students of the 'Program for rural development of Cordillera' (Programa de Desarrollo Campesino de Cordillera, PDCC). The 2 local physicians and the health representative of the APG worked as field supervisors.

Training

A 2-week training phase preceded the field study. During the 1st week training sessions were held in a classroom setting for 8 h a day. The topics were generally the same as those covered in the interviewer training for the SNES project [5]. The purpose and procedures for the study were presented and discussed. The interviewers were told that: (1) the study was important for the Cordillera province, especially to establish disease prevention programs (epilepsy); (2) their role was very important to warrant success of the study; (3) a successful study might lead to work on additional research projects. The importance of data quality was stressed. The screening questionnaire was reviewed question by question and an interviewer manual was given to each participant. Interviewers administered questionnaires to each other and discussed the results. Transportation and payment procedures were discussed. A meeting with supervisors was held to explain their responsibilities. Field materials were given to each interviewer. These included a water-resistant bag, a ballpoint pen, the interviewer's manual, and the questionnaire.

During the 2nd week the interviewers went in pairs to pretest the questionnaire on hospital patients and subsequently on residents of select rural communities.

Pilot Investigation

A pilot investigation was performed to determine compliance with the screening instrument and to evaluate clarity of items. The pilot investigation was performed in October 1994 in two small communities of 291 inhabitants (Cañon Segura and Guasuigua). The pilot investigation was carried out by all the members of the field staff. The questionnaires were then revised and minor changes were made.

Phase 1: Household Screening

This phase was performed by interviewers during a 5-month period (November 1994–March 1995). During this period the 26 health workers were transported by 4-wheel drive vehicles, trucks or buses to all com-

munities after agreement by the leader of each community. A supervisor who accompanied the interviewers explained and discussed the project to the leader of the community and then to the head of each family.

Initially the health workers drew a map of each community. This was essential to determine the number and geographical position of all houses in each area. The interviewers worked every day (except holidays) during the morning and the afternoon to administer the screening instrument to all eligible subjects. All subjects who were selected but not present initially, were listed and subsequently contacted at least twice. For each household, an adult was selected when the head of the household refused to cooperate or was absent.

The 26 health workers always operated in the communities under a supervisor, and random spot checks were made by a Bolivian physician.

All phase 1 data were double-entered into a computer using the Epi-Info package [8]. Data were checked for consistency and errors were corrected.

Phase 2: Neurological Examination

The staff was composed of 4 Italian neurologists. All the neurological examinations were completed in 4 visits to Bolivia of about 2 months each (total 8 months). During each visit the neurologists worked fulltime, every day, morning and afternoon, except holidays. The Health Representative of the APG assisted with all the neurological fieldwork. All subjects who screened positive were contacted 1 or 2 days before the neurologist's visit. Subjects were gathered at the health post or a community school. The neurologists examined subjects at home if they could not travel to the meeting place. Subjects who were not present at the time of the examination were listed and examined later. At least 2 visits were performed to each community. A typical examination lasted 30-45 min, with about half of that time devoted to history taking and clinical measurements (e.g. pulse rate, blood pressure). For each subject the neurological evaluation included the cranial nerves, motor function, deep tendon reflexes, coordination, gait and posture, and sensory perception. All clinical data were collected on preprinted forms and then added to the computerized data base using the Epi-Info program [8]. During the examination, the study neurologist did not prescribe treatment but referred subjects to the local doctor at the area hospital when indicated. Neurologists met with local physicians to discuss case findings.

Community Relationship

To obtain a successful survey a high level of community cooperation was required. Local radio was used to inform persons about the survey. Meetings with the head and the adults of each community were held before the start of the field work to explain the aim of the survey and to answer questions about the study. Priests and the nuns of the local missions supported the project and the disseminated information about the program in the target communities.

Quality Control

During the household screening the interviewers worked under the auspices of 2 supervisors. For the 1st month all data were checked at the end of each day to detect errors which were discussed with the interviewer the day after. Furthermore, 20% of the subjects screened on that day were randomly interviewed by a physician-supervisor. Once an area survey was completed, all data were checked by a Bolivian physician before entry into the computer.

The neurological examinations were performed in the field by at least 2 neurologists. The neurologists always worked together in the same community to allow for discussion of cases.

Results

The survey was completed in November 1996. Fifty-five communities were selected by cluster sampling from the ten areas of the Cordillera province (table 2). They contained 1,941 households in total. The eligible population consisted of 10,124 subjects. At the end of the household screening 9,955 subjects had been studied. 199 (2%) were not included because 72 were not living in the area and 127 (1.3%) refused to participate. Table 1 and figure 2 show the age and sex distribution of the study population. Table 3 shows the main occupations of persons over 12 years old. The population under 13 years of age was 4,563 while the adult population was 5,392, of which 70% were directly and 30% indirectly interviewed. Of the 9,955 subjects, 1,130 (11.3%) screened positive, of which 1,027 were examined by neurologists in phase two.

Table 2. Population distribution per area

Area	Total population	Selected population	Clusters selected
Boyuiibe	3,701	485	5
Cabezas	8,007	1,264	5
Camiri	5,652	1,417	8
Charagua	8,000	1,313	7
Cuevo	3,329	245	3
Eiti	5,500	848	3
Gutierrez	5,698	1,889	9
Izozo	7,464	1,291	5
Lagunillas	4,581	804	7
San Antonio Parapeti*	2,992	399	3
Total Cordillera province	54,324	9,955	55

Table 3. Occupation

	%	95% CI
Housewife	45.9	44.6-47.2
Farmer	43.3	42.0-44.5
Fishermen	0.2	0.1-0.3
Breeder	1.1	0.4-1.7
Workman	0.8	0.5-1.2
Laborer	0.9	0.6-1.3
Tradesman	0.4	0.2-0.6
Civil servant	0.4	0.1-0.5
Professional	0.5	0.2-0.8
Unemployed	3.9	2.8-5.1
Other	2.5	1.9-3.2

One-hundred and three (9%) were not examined because 7 refused, 86 were lost to follow-up and 10 died during the study (fig. 3). Table 4 shows the proportion giving a positive response to each element of the screening instrument (symptom questionnaire and physical examination).

Discussion

Neuroepidemiological studies can provide important information regarding the frequency, causes, and potential for prevention of neurological diseases in developing countries. The results obtained can be used to assist in planning health services and intervention strategies.

One of the essential requirements for the successful implementation of neuroepidemiological studies in developing countries is community collaboration. In this study the establishment of a working relationship with the APG (the political organization which represents the Guaraní population) was particularly important. We presented and discussed the aim of our project with the representatives of APG and found that they were interested in supporting the study, mainly because it targeted epilepsy, a major health problem in this area.

The involvement of local health workers favored acceptance by this rural population, and thus, the refused rate was low. We did not find language to be a major barrier. In fact, the vast majority of the population understood Spanish. In a few isolated communities, where some women and children spoke only the Guaraní language, health workers translated the study information.

The screening questionnaire, previously used in a prevalence survey of neurological diseases in Italy, was found to be a valid instrument and only minor changes were necessary when it was retested for this field study. Major costs of the study were transportation (i.e. petrol, vehicle maintenance, etc.) and payment of the health workers. The long distances and the poor condition of the roads and the length of time needed to conclude the field work underlied the financial costs. Flooding due to heavy rains sometimes made it impossible to reach communities for weeks.

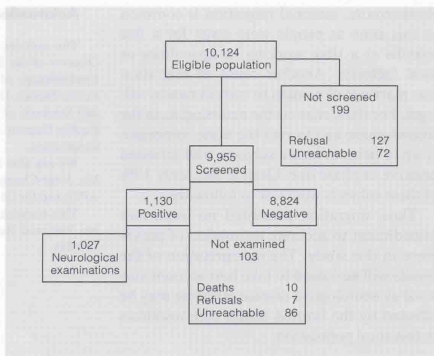


Fig. 3. Flow chart of the screening phase.

Table 4. Evaluation by single items of the screening instrument

Items	Positive		Negative	
	n	%	n	%
<i>Symptom questions</i>				
Impaired consciousness	399	4.0	9,555	96.0
Uncontrolled movements of limbs	247	2.5	9,707	97.5
Absence spell (only for children)	69	1.5	4,506	98.5
Paralysis or weakness of limbs	222	2.2	9,732	97.8
Sensory changes in limbs	331	3.4	9,623	96.6
Changes in speech ¹	105	2.0	5,274	98.0
Paralysis of face ¹	55	1.0	5,324	99.0
Drooping of mouth ¹	80	1.4	5,299	98.6
Rigidity ¹	148	2.8	5,231	97.1
Tremor of head or limbs ¹	135	2.6	5,244	97.4
Diplopia ¹	171	3.1	5,208	97.0
<i>Physical tasks¹</i>				
Holding arms extended	64	1.9	3,587	98.1
Distinguishing textures	16	0.4	3,639	99.6
Touching finger to nose	56	1.5	3,589	98.5
Walking heel-to-toe	53	1.5	3,547	98.5
Standing, eyes open	28	0.8	3,599	99.1
Standing, eyes closed	55	1.5	3,751	98.5

¹ Adults only.

Furthermore, seasonal migration is common in this zone as people were away for a few months at a time working in agriculture or local factories. Another cause of migration was marriage of women to men in nearby villages. For these reasons the neurologists in the second phase had to visit the same community up to twice to locate subjects who screened positive in phase one. Despite this, only 10% of these subjects were lost to follow-up.

Thus, migration presented no important impediment to accurate estimation of prevalence in this study. The interpretation of the results will also need to take into account survival as neurological disease outcome may be affected by the limited health care resources in this rural population.

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