Epidemiological characteristics of hepatic echinococcosis, concurrent cerebral echinococcosis, and pulmonary echinococcosis in Ganzi County, Sichuan Province, China

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Abstract
Human echinococcosis has become a major public health problem in most parts of the world. The objective of this article was to study the demographics of patients with hepatic echinococcosis in Ganzi County to elucidate the main risk factors, as well as to report the concurrent prevalence of cerebral echinococcosis and pulmonary echinococcosis.

We recruited 195 patients with hepatic echinococcosis from the Datongma area of Ganzi County from January 2018 to November 2018. The patients’ demographics, living environments, supported medical resources, knowledge of echinococcosis prevention and control, and hygienic practices were investigated and analyzed. The prevalence of cerebral echinococcosis and pulmonary echinococcosis were also investigated.

The data were analyzed to identify risk factors for human echinococcosis. Our analysis showed that the herding Tibetan population within the 20 to 60 age group, and females, in particular, were at the highest risk of human echinococcosis infection. Having stray dogs around habitations and intimate activities with dogs and livestock were also behavioral risk factors. People with poor health literacy and low educational qualifications had possible risks of infection. In terms of hygiene, not using tap water as the drinking water source and lack of medical staff were significantly correlated with echinococcosis prevalence. Four patients were diagnosed with cerebral echinococcosis. Among them, 1 patient had both cerebral echinococcosis and pulmonary echinococcosis.

Possible high-risk factors for echinococcosis were being female, herding population, in the 20 to 60 age group, having stray dogs around habitations, having activities with dogs and livestock, having poor health literacy, having low educational qualifications, and not using tap water as a drinking water source. The detection rate for brain echinococcosis in patients with hepatic echinococcosis was high (2.05%). Effective preventive strategies should be implemented in epidemic areas. Head CT scans should be applied for early detection of cerebral echinococcosis to carry out the treatment.

Abbreviations: AE = alveolar echinococcosis, CE = cystic echinococcosis.

Keywords: cerebral echinococcosis, hepatic echinococcosis, pulmonary echinococcosis, risk factors

1. Introduction
Echinococcosis (hydatid disease) is a severe zoonotic parasitic disease caused by larval forms of Echinococcus in animals and humans.\textsuperscript{[1]} The prevalent forms of echinococcosis are of 2 types, cystic echinococcosis (CE) and alveolar echinococcosis (AE),\textsuperscript{[2]} caused by infection with the larval stage of E. granulosus and E. multilocularis, respectively. The treatment of echinococcosis requires continuous chemotherapy for many years, or even for the whole life.\textsuperscript{[1]} It can lead to economic losses in the livestock source and lack of medical staff were significant correlated with echinococcosis prevalence. Four patients were diagnosed with cerebral echinococcosis. Among them, 1 patient had both cerebral echinococcosis and pulmonary echinococcosis.

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industry and cause severe harm to health and even death in humans.\textsuperscript{2–8} The conservative estimate for the minimum average global human cost of CE is approximately 285,000 disability-adjusted life years and an annual loss of US $194,000,000.\textsuperscript{9,10} CE is highly endemic in pastoral areas, especially in regions of South America, Eastern Europe, the Middle East, East Africa, China, and Russia. AE is strictly distributed in Europe, the Near East, Russia, and China. CE and AE have aroused wide public health concerns.\textsuperscript{11} In general, CE in the early stage can be asymptomatic. It has been reported that its asymptomatic rate can reach 33.88\%.\textsuperscript{12} The onset symptom of CE is a subtle pain that is not noticeable. The mortality rate of CE is approximately 2\% to 4\%. However, AE is more invasive and progressive compared to CE, and the clinical symptoms are more serious,\textsuperscript{13} including epigastric pain and cholestatic jaundice. If AE remains untreated or undertreated, the mortality rate can exceed 90\% in less than 15 years after diagnosis, which is similar to the death rate of liver cancer.\textsuperscript{9} China is one of the countries with the highest prevalence of human echinococcosis. Echinococcosis is mainly prevalent in rural, semiagricultural, and semipastoral areas in western China, especially in Xinjiang Uygur Autonomous Region, Qinghai Province, Gansu Province, Ningxia Autonomous Region, Tibet Autonomous Region, Inner Mongolia Autonomous Region, and Western Sichuan Province.\textsuperscript{9,11–13} It is one of the main factors reducing the income of herdsmen and farmers.\textsuperscript{14,14,15} Therefore, it is essential to identify the prevalence of echinococcosis and the demographics of these patients to elucidate risk factors to help policymakers develop effective control and prevention strategies.

2. Materials and methods

2.1. Settings and eligibility criteria

This cross-sectional study was undertaken at the People’s Hospital of Ganzi County, which is located in the Ganzi Tibetan Autonomous Prefecture in the western Sichuan Province of southwestern China. The mean elevation of Ganzi County is 3390 m above sea level, and the annual mean temperature is 5.6°C. By the end of 2010, the total population of Ganzi County was 72,000, and the Tibetan population accounted for over 95\%. Their primary economic income is from livestock production and herding. Their lifestyle is nomadic or seminomadic. In addition, this area has many domestic dogs, stray dogs, and wild canids, such as foxes. Ganzi County had already been confirmed to have a focus of human echinococcosis cases, estimating human echinococcosis prevalence of 1.86\%.\textsuperscript{16} The Diagnostic Criteria for Echinococcosis (WS257–2006)\textsuperscript{17} was adopted to screen the eligible participants. The patients who met inclusion and exclusion criteria were selected as participants. Inclusion criteria: participants with hepatic echinococcosis diagnosed by abdominal ultrasound. Exclusion criteria: individuals with poor compliance, the tendency for inadequate follow-up, and severe mental or physical disorders.

2.2. Questionnaire

The questionnaire was developed based on the Program of National Epidemiological Survey on Echinococcosis released by the Chinese Centers for Disease Control and Prevention in 2011. It included information on demographics, animal ownership, sources of drinking water, hygiene-related behaviors, lifestyle practices, and knowledge of echinococcosis prevention and control.

The following demographic data were collected in the questionnaire: age, gender, nationality, religion, occupation, educational qualification, and living styles (nomadic, seminomadic, or settled). The numbers of owned livestock and species (cattle, sheep, camel, and pig) were documented. The surrounding environment evaluated in this study focused on dog care, the presence of stray dogs in the community, sources of drinking water, and accessible medical resources in villages. Additionally, health knowledge about human echinococcosis prevention and control were collected to find the relationship between educational level and health literacy.

2.3. Procedures

From January to November 2018, we screened 13,097 residents from 1 town and 22 villages in Ganzi County through abdominal ultrasound; 509 were diagnosed with hepatic echinococcosis in the People’s Hospital of Ganzi County. Prior to the survey, information about the survey, provision of free treatment to patients diagnosed with hepatic echinococcosis were clearly explained. They could choose to participate by their own will and withdraw their participation without any disadvantage. A total of 195 participants provided oral consent and were finally enrolled. Full-time investigators interviewed all participants. Each participant completed the questionnaire during the interview. All the analyzed data were anonymized. The investigators only explained the questions of the questionnaire and avoided using inductive language to ensure the results were objective and accurate. Furthermore, all patients received a chest CT and a head CT to detect cerebral and pulmonary echinococcosis.

2.4. Statistical analysis

All data were coded and analyzed using SPSS version 18.0 (IBM). The missing data were not included in the analysis. The qualitative data were described by the number of cases and the percentage. The quantitative data were displayed by showing means and standard deviations. A Chi-Squared test ($\chi^2$) was performed to assess the between-group differences. $P$-values ($\alpha$) of less than .05 were considered significant.

3. Results

3.1. Demographics of the participants

During the analysis of questionnaire information and responses, all participants were computed for each question. The demographics of the 195 participants are presented in Table 1. Analysis of demographics for all participants indicated that females were at a higher risk of infection in comparison to males, with 120 females and 75 males included in this study. The age of individuals ranged from 7 to 75 years, with an average age of 39.95 ± 15.68 years. The highest prevalence by age group was in the 41- to 50-year-old group (28.21\%), followed by the 31- to 40- and 51- to 60-year-old groups (18.97\% and 15.38\%, respectively). There was a positive association with involvement in herding and farming compared to other occupations (61.03\% and 26.15\%, respectively). Herdsmen were at the highest risk of echinococcosis infection compared to other occupations (61.03\%). The second high-risk occupational group was farmers, accounting for 26.15\%. All were Tibetans, and their religion was
Buddhism. In terms of educational qualification, most of them (86.67%) were illiterate, and 62.56% lived in settlements.

3.2. Raising livestock

Categories and numbers of livestock raised in each participant’s household were investigated. The results showed that the average numbers of sheep and cattle raised per household were 0.03 and 9.80, respectively. The number of cattle raised per household was significantly higher than the number of sheep raised. The proportion of households raising cattle was 37.10%, and the proportion of households raising sheep was 1.08% (Table 2).

3.3. Living environment and medical resources

Among the 195 patients with hepatic echinococcosis, 41.03% reported stray dogs near their habitations. Sixty percent of the participants said their neighbors raised dogs, and 48.21% of participants chose tap water as their drinking water source. Additionally, 24.61% of them said they did not have a doctor or did not know if there were doctors in their village. Relevant information is shown in Table 3.

3.4. Echinococcosis prevention knowledge and hygienic practices

Among the 195 participants, 8.21% of them knew or partially knew the route of transmission of echinococcosis. There was a significant difference in the percentage knowing the transmission route among the different groups ($\chi^2=12.534$, $P<.05$), and the student group had the highest proportion (44.4%). In terms of hygienic practices, 5.64% of the participants often washed their hands, 56.41% sometimes washed their hands, and 37.95% rarely washed their hands. Patients with different educational qualifications and living styles showed significant differences in handwashing before eating ($P<.05$). Approximately 99.49% of the participants were willing to deworm dogs if it was free of charge (Table 4 and Table 5).

3.5. Detection of echinococcosis outside the abdominal cavity

Among the 195 patients with hepatic echinococcosis, 4 patients were found to have cerebral echinococcosis, and the detection

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**Table 1**

Assessed hepatic echinococcosis patients’ demographic characteristics ($n=195$).

| Category          | Characteristic factor | No. of participants investigated | Composition ratio (%) |
|-------------------|-----------------------|----------------------------------|-----------------------|
| Gender            | Male                  | 75                               | 38.46                 |
|                   | Female                | 120                              | 61.54                 |
| Age               | 1~                    | 3                                | 1.54                  |
|                   | 10~                   | 20                               | 10.26                 |
|                   | 20~                   | 29                               | 14.87                 |
|                   | 30~                   | 37                               | 18.97                 |
|                   | 40~                   | 55                               | 28.21                 |
|                   | 50~                   | 30                               | 15.38                 |
|                   | 60~                   | 14                               | 7.18                  |
|                   | 70~                   | 7                                | 3.59                  |
| Ethnicity         | Tibetan               | 195                              | 100.00                |
| Religion          | Buddhism              | 195                              | 100.00                |
| Occupation        | Part-time herdsmen    | 5                                | 2.56                  |
|                   | Herdsmen              | 119                              | 61.03                 |
|                   | Farmer                | 51                               | 26.15                 |
|                   | Others                | 2                                | 1.03                  |
|                   | Student               | 13                               | 6.67                  |
|                   | Religious people      | 5                                | 2.56                  |
| Educational qualification | Illiteracy     | 169                              | 86.67                 |
|                   | Elementary School and above | 26                          | 13.33                 |
| Living styles     | Settled               | 122                              | 62.56                 |
|                   | semi-nomadic          | 56                               | 28.72                 |
|                   | nomadic               | 17                               | 8.72                  |
| Total             |                       | 195                              | 100.00                |

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**Table 2**

Raising livestock investigation of 186 households.

| Livestock species | Number of households surveyed | Feeding quantity | The average number of livestock | The proportion of households |
|-------------------|-------------------------------|------------------|-------------------------------|-------------------------------|
|                   |                               | No. of households (head/household) | Number of households | Proportion (%) |
|                   |                               | Feeding quantity | The average number of livestock | Number of households | Proportion (%) |
| Cattle            | 186                           | 1823             | 9.80                          | 69                           | 37.10          |
| Sheep             | 6                             | 0.03             |                               | 2                            | 1.08           |
| Camels            | 0                             | 0.00             |                               | 0                            | 0.00           |
| Pigs              | 0                             | 0.00             |                               | 0                            | 0.00           |
| Total             | 186                           | 1829             | 9.83                          | 71                           | 38.17          |

186 households were investigated. 115 households were working for other households instead of owning livestock.

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**Table 3**

The surrounding environment and medical resources.

| Living environment and medical resources | Characteristics | No. of people investigated | Composition ratio (%) |
|----------------------------------------|-----------------|---------------------------|-----------------------|
| Stray dogs around                      | Do not know     | 4                         | 2.05                  |
|                                        | None            | 111                       | 56.92                 |
|                                        | Yes             | 80                        | 41.03                 |
| Neighbors raise dogs                   | Neighbors do    | 78                        | 40.00                 |
|                                        | not raise dogs  |                           |                       |
|                                        | Almost every    | 3                         | 1.54                  |
|                                        | family raises dogs |                   |                       |
|                                        | Some raise dogs | 114                       | 58.46                 |
| Water resources                        | Ditch           | 16                        | 8.21                  |
|                                        | River           | 53                        | 27.18                 |
|                                        | Well            | 28                        | 14.36                 |
|                                        | Spring          | 4                         | 2.05                  |
|                                        | Tap water       | 94                        | 48.21                 |
| Doctors                                | Do not know     | 8                         | 4.10                  |
|                                        | None            | 40                        | 20.51                 |
|                                        | Yes             | 147                       | 75.39                 |
rate was 2.05%. One patient was found to have pulmonary echinococcosis for a detection rate of 0.51% (Table 6).

4. Discussion
Sichuan Province is one of the most widespread epidemic areas for human echinococcosis in China.\(^{18}\) It is a mixed epidemic area of CE and AE\(^{4,18,19}\), which seriously endanger the public health and cause economic losses in the livestock industry in the epidemic areas of this province.\(^{20,21}\) According to the authoritative survey of the epidemiological status of echinococcosis in Sichuan Province in 2012\(^{16}\), the prevalence of echinococcosis in Ganzi Tibetan Autonomous Prefecture was the highest, reaching 1.86%, which was much higher than the average level of echinococcosis in the epidemic area of the province (1.08%). The survey results also showed that the estimated prevalence of echinococcosis in Ganzi County was 2.26%, which ranked fourth in the 18 counties in Ganzi Tibetan Autonomous Prefecture.\(^{22}\) Therefore, it was important to investigate the prevalence of echinococcosis and evaluate the status quo of prevention and control knowledge of echinococcosis for policymakers to develop targeted strategies for echinococcosis prevention and control. Therefore, this study selected the Datongma District, which has the highest prevalence of echinococcosis in Ganzi County, as the investigation area. A total of 195 patients with hepatic echinococcosis were enrolled to investigate the demographic characteristics, the number of livestock raised by each household, living environment, medical resources, knowledge of echinococcosis prevention and control, and hygienic practices.

The results of this survey showed that of 195 patients with evidence of hepatic echinococcosis, 120 were female and 75 were male. Females had a higher risk of getting infected with echinococcosis than males. Similar results were also demonstrated.

**Table 4**

| Knowledge of echinococcosis prevention and control and hygiene practices. |
|---------------------------------------------------------------|
| **Prevention of knowledge and behaviors** | **Characteristic factor** | **No. of people investigated** | **Composition ratio (%)** |
| Mode of transmission | Know or partially know | 16 | 8.21 |
| | Do not know | 179 | 91.79 |
| Hand washing | Frequently | 11 | 5.64 |
| | Sometimes | 110 | 56.41 |
| | Seldom | 74 | 37.95 |
| Free deworming for dogs | Willing | 194 | 99.49 |
| | Unwilling | 1 | 0.51 |

**Table 5**

| Analysis of the factors affecting the knowledge of echinococcosis prevention and control and the status quo of health behaviors. |
|---------------------------------------------------------------|
| **Categories** | **How do people get echinococcosis (n)** | **Wash your hands before eating (n)** |
| | Know or partially know | Not aware | Wash hands frequently | Wash hands sometimes | Wash hands rarely |
| Total | 16 | 179 | 10 | 110 | 75 |
| Gender | | | | | |
| Male | 3 | 72 | 3 | 40 | 32 |
| Female | 13 | 107 | 8 | 70 | 42 |
| \(\chi^2, P\) | \(\chi^2=2.861, P=.091\) | \(\chi^2=1.501, P=.472\) |
| Age | | | | | |
| 1~7 | 7 | 45 | 2 | 33 | 17 |
| 30~ | 6 | 116 | 6 | 67 | 49 |
| 60~ | 3 | 18 | 3 | 10 | 8 |
| \(\chi^2, P\) | \(\chi^2=5.048, P=.059\) | \(\chi^2=4.178, P=.362\) |
| Occupation | | | | | |
| Herders or Part-time herdsmen | 12 | 114 | 7 | 72 | 47 |
| Farmer | 0 | 51 | 1 | 28 | 22 |
| Student | 4 | 9 | 2 | 9 | 2 |
| Religious people | 0 | 5 | 1 | 1 | 3 |
| \(\chi^2, P\) | \(\chi^2=12.543, P=.004\) | \(\chi^2=10.046, P=.004\) |
| Educational level | | | | | |
| Illiteracy | 12 | 154 | 7 | 97 | 62 |
| Preschool | 0 | 3 | 0 | 0 | 3 |
| Elementary school and above | 4 | 22 | 4 | 13 | 9 |
| \(\chi^2, P\) | \(\chi^2=2.904, P=.414\) | \(\chi^2=8.977, P=.040\) |
| Living modes | | | | | |
| Settlement | 9 | 113 | 10 | 57 | 55 |
| Settlement during winter and nomads during summer | 7 | 49 | 0 | 44 | 12 |
| Nomads | 0 | 17 | 1 | 9 | 7 |
| \(\chi^2, P\) | \(\chi^2=2.446, P=.273\) | \(\chi^2=17.866, P=.001\) |

\(\ast\) Fisher exact probability test.
were many stray dogs in Tibetan areas.\[18,24,25\] Another risk factor increasing the likelihood of human infection was feeding domestic dogs and livestock, the chances of becoming sick were substantially increased.\[18\]

The age of the infected participants ranged from 7 to 75 years, with an average age of 39.95 ± 15.68. Cases of echinococcosis were found in different age groups: the 40- to 49-year-old group had the highest prevalence, followed by the 30- to 39-year-old group (18.97%) and the 50- to 59-year-old group (15.38%). The possible reason was that adults, as the primary labor force of the family, had a higher probability of contact with the contaminated environment or infected animals. Thus, they were more easily infected.

Among the different occupational groups, the infection prevalence in herders was significantly higher than that in other occupational groups, accounting for 61.03%. The main activities of herdsman included hazing, which increased the possible risk of exposure to sheep and dogs. Some other behaviors, such as allowing dogs to sleep indoors at night[18] and playing with dogs, were also high-risk behaviors for infection. Therefore, the Tibetan herdsman population was at a significantly higher risk of infection by echinococcosis. This population should be the focus of control strategies.

Approximately 41% of the participants said that there were often wild animals in their residence, and 60.00% of the participants said they had dogs, or their neighbors kept dogs. The presence of stray dogs was another significant risk factor impacting the prevalence of human echinococcosis. Qian et al[23] verified that the *E. multilocularis* prevalence in owned dogs was significantly associated with the number of stray dogs.\[16\] In our study, all enrolled participants were Tibetans, and they all practiced Buddhism, which does not support killing and is associated with close activities with dogs.\[18\] Therefore, there were many stray dogs in Tibetan areas.\[18,24,25\] Another risk factor increasing the likelihood of human infection was feeding dogs viscera.\[16\] Since the Tibetan participants did not like eating livestock viscera, most viscera of the slaughtered livestock were fed to the dogs, and the dogs were infected by the contaminated viscera. The transmission of echinococcosis to humans was caused by accidental ingestion of parasite eggs excreted in the feces of the infected dogs, and a cycle of echinococcosis transmission formed. Consequently, feasible control measures with priority on the plateau should be explored to treat livestock, domestic dogs, and stray dogs, for example, deworming of both owned and stray dogs. The harmless disposal of infected animals and viscera should also be strongly facilitated.

Regarding the surrounding environment and medical resources, only 48.21% of the 195 participants used tap water for drinking. Other water resources were significantly correlated with echinococcosis prevalence, for example, ditch or river water. Approximately 25% of the participants did not know if there were doctors in the village or not. Thus, there is an urgent need to provide tap water resources and increase the number of medical staff in epidemic areas.

According to the survey, only 8.21% of the participants knew or partially knew the route of transmission of echinococcosis. There were significant differences in the awareness rate among different occupational groups, with students having the highest awareness rate (44%). In terms of hygiene practices, 5.64% often washed hands, 36.41% sometimes washed their hands, and 37.95% rarely washed their hands. Patients with higher educational qualifications performed better handwashing self-discipline. Patients with lower education qualifications had fewer opportunities to access knowledge and understanding of echinococcosis prevention and control, which might inhibit the implementation of effective behaviors for echinococcosis prevention and control. In addition, most of the Tibetan population lives in remote areas, where living conditions are poor and health literacy is generally inadequate, which might be the reason for the high frequency of echinococcosis in the Tibetan population. Therefore, a health education program aligned with the population’s educational background and local culture is necessary to effectively disseminate knowledge and improve hygienic practices.

Echinococcosis outside the abdominal cavity (cerebral echinococcosis and pulmonary echinococcosis) was also detected. According to the sampling survey of the epidemiological status of echinococcosis in Sichuan Province in 2012,\[16\] the prevalence of human echinococcosis in the province was 1.08%. In this survey, we enrolled 195 patients with hepatic echinococcosis. Four patients were diagnosed with cerebral echinococcosis. Among them, 1 patient had both cerebral echinococcosis and pulmonary echinococcosis, which has higher mortality than CE.[9,12]

Therefore, an early head CT scan should be applied to detect and implement early treatment measures for cerebral echinococcosis in patients with echinococcosis.

The first limitation of this study is the sampling method, which might introduce potential unknown bias. This limitability might jeopardize the representativeness of the sample and generalizability of the results. Another limitation is the small sample size. Although we diagnosed 509 patients with hepatic echinococcosis, 195 patients agreed to participate in our study and share their information. We could not present all the data of all screened local people. Based on the data of enrolled participants, we still can realize the objectives of this study, which is to identify the possible risk factors for human echinococcosis.

### Table 6

| No. of participants | No. of patients | Detection rate(%) | No. of patients | Detection rate(%) | No. of patients | Detection rate(%) |
|---------------------|----------------|-------------------|----------------|-------------------|----------------|-------------------|
| 195                 | 4              | 2.05              | 1              | 0.51              | 4              | 2.05              |

5. Conclusions

Possible high-risk factors for echinococcosis identified by this study were being female, herding population, in the 20 to 60 age group, having stray dogs around habitations, having activities with dogs and livestock, having poor health literacy, having low educational qualifications, and not using tap water as a drinking water source. Effective preventive strategies should be implemented, such as deworming both owned and stray dogs,

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providing tap water drinking resources, disposing of infected animals and viscera, holding health education activities, and increasing the number of medical staff in epidemic areas. The detection rate for brain echinococcosis in patients with hepatic echinococcosis was high (2.05%). Therefore, head CT scans should be applied for early detection of cerebral echinococcosis to carry out the treatment.

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