Human and environmental risk factors of leptospirosis in Gunungkidul, Indonesia: a case-control study

By SULISTYAWATI
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Human and environmental risk factors of leptospirosis in Gunungkidul, Indonesia: a case-control study

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ABSTRACT

Background: Leptospirosis is a bacterial, zoonotic disease associated with environmental factors and human behavior related to occupation. This research aimed to investigate the association between human behavior and leptospirosis. Case mapping and topographical mapping were done for a comprehensive visualization.

Methods: A matched case-control study design was conducted in Gunungkidul, Indonesia, from December 2017 to January 2018. Cases were selected from those reported as suspicious of leptospirosis by the Gunungkidul District Health Office during 2017 and controls were matched according to sex and age. Chi-squared, Fisher exact test, and Odds ratios were employed to find out the association between exposure and outcome for a significance level of 0.05. Quantum GIS-Web Map-Stamen terrain was used to overlay case and landscape.

Results: Bivariate analysis showed that four exposure variables that enhanced the risk for leptospirosis though not significantly associated were history of injuries, habit of taking a bath or wash the clothes in the river, not using personal protection during work and presence of an animal fence surrounding the house. Most of the leptospirosis cases (>70%) resided in a hilly area. This finding assists in developing prevention strategies concerning leptospirosis infection.

Conclusions: Human behaviour is vital in leptospirosis prevention. Accordingly, this study can broaden the understanding horizon, particularly for a decision-maker where and how to implement the Leptospirosis prevention program. Prevention should address the current situation in the field and based on population and local wisdom to result in the successful implementation.

Keywords: Environmental, GIS, Human, Indonesia, Leptospirosis

INTRODUCTION

Leptospirosis is a severe problem in tropical countries with global, annual mortality close to 60,000 given according to a recent review.1 *Procrustes* from the bacterial genus *Leptospira* is responsible for this infection among humans.2,3 Transmission occurs when humans with skin cuts and/or abrasions come in direct contact with soils or water contaminated by urine containing the spirochetes coming from infected animals.2 Several studies have discussed the potential risk factors. In a study from southern India, outdoor occupational activities can produce cuts or wounds, often superficial but still a risk when there is either frequent direct contact with rodents or contact with soil or water that may contain urine of infected animals.4 A recent review mentions that leptospirosis is also a risk in high-income countries when people's occupation puts them in contact with outdoor freshwater and animals.5 Along with the globalization era, the transmission of this infection can
also be attributed to people from non-endemic countries who visit areas where transmission of leptospirosis exists, for example, for tourist purposes. Previous research holds that farmers in the agriculture sector are vulnerable to leptospirosis as they usually have low levels of knowledge, particularly about this disease and its transmission patterns, and therefore are not inclined to protect themselves. At present, a large number of leptospirosis studies in Indonesia have been conducted. However, none of them combine the human risk factor and environmental coordinates such as topography and soil type. That type of research is essential to comprehensively see the pattern for a better understanding of the situation and build effective evidence-based prevention programs. The current study was aimed to investigate the potential effect of geographical, environmental parameters such as elevation over the mean sea level and human behavior as possible risk factors for leptospirosis.

METHODS

Study area

This study was conducted in the Gunungkidul District (110°21' - 110°50' East longitude and 7°36' - 8°09' South latitude), situated on the coast in south-eastern of Yogyakarta Province, Indonesia (Figure 1). The district has a population of 698,825, has been given by a census in 2014, and covers 1,483.36 km² divided into 18 sub-districts—Gunungkidul District experiences a tropical climate, rain, and drain. The topography varies between 0 and 700 m above the mean sea level, with the North being hilly, the Centre plain, and the South mountains. The ground consists mostly of limestone structures that make the soil infertile. Some areas have caves with an underground river, while only a few are suitable for agriculture as there is a canal that can be used for irrigation.

![Figure 1: Gunungkidul district position towards Yogyakarta province.](image)

Study design

This matched case-control study design was carried out to explore associated risk factors for leptospirosis occurrence in the Gunungkidul District. Cases and controls were matched with respect to sex and age, while a +5 years range of age of the controls was accepted considering the possibility of not getting enough controls. We performed a 1:1 ratio for cases and controls.

The case defined as persons residing in Gunungkidul District according to his/her identity card and confirmed by a serology test as leptospirosis-positive during the period January to December 2017. This person should also have been registered in Gunungkidul Health Office's monthly report and visited for a minimum of three times. Cases who were not in place during the three visits or had migrated to/from other areas were excluded from this research. The control was people residing in Gunungkidul District refer to his/her identity card. They were confirmed by a serology test to be leptospirosis negative during January to December 2017. So, they were not being listed in the Gunungkidul Health Office monthly report.

Data collection and study tools

The research was done based on data covering the period from December 2017 to January 2018. In total, 63 cases and suspected cases were listed in the district health office of Gunungkidul; among them, 24 were selected based on the inclusion criteria. Matching controls were sought nearby the cases ensuring that they met with the inclusion criteria. In total, 48 respondents were included in this study, consisting of 24 cases and 24 controls. All of them worked in the agriculture sector. They were between 31 and 75 years old. The majority of the participants were males (83.7%). More than 60% of the respondents had primary school education. We used a semi-structured questionnaire adapted from previous research. It was offered to both cases and controls and divided into five sections: household head information, respondent information, behavior, history of injury, and environmental conditions. Data were collected through a door-to-door visit in the residential area. At the end of the interview, the research team observed whether there was a fence around the house. We examined for potential exposure factors, such as occupation; history of skin injuries; habit of bathing or washing clothes in the river; lack of personal protection during work; “Kerja Bakat,” i.e., cleaning sewers or gutters; and presence or lack of a fence surrounding the house.

Statistical analysis and mapping

Data were input in the SPSS version 24.0 (IBM Corp., Armonk, NY, USA). Univariate analysis was used to tabulate and calculate the percentage of each category. We employed the chi-square and the Fisher exact test to assess the association between exposure and the outcome, considering a significance level of p<0.05. Odds ratios with 95% confidence interval was calculated. To get geographical information on the study area's environment, we used a digital elevation model (DEM) overlay...
produced by Quantum GIS through Web Map-Stamen terrain background.

**RESULTS**

**Bivariate analysis of potential risk factors for leptospirosis**

Among the six potential exposure factors, we found four to carry some risk for leptospirosis, but none was significantly associated. The occupation was not countable due to the uniformity between cases and controls groups, and having a history of outdoor social activities potentially vulnerable to leptospirosis infection was not found to be a real risk for infection (Table 1).

**Leptospirosis mapping**

In our study, more than 70% of the leptospirosis cases were found to live in the hilly areas in the North, while only a few of them live in the plains in the Centre (Figure 2).

**Table 1: Bivariate analysis of risk factors.**

<table>
<thead>
<tr>
<th>Variable</th>
<th>Number of cases (%)</th>
<th>Number of controls (%)</th>
<th>Crude OR (95% CI)</th>
<th>P-valueb</th>
</tr>
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<tbody>
<tr>
<td>Occupation</td>
<td>24 (100)</td>
<td>24 (100)</td>
<td>-a</td>
<td>-a</td>
</tr>
<tr>
<td>At risk</td>
<td>24 (100)</td>
<td>24 (100)</td>
<td>-a</td>
<td>-a</td>
</tr>
<tr>
<td>History of skin injuries</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>6 (25)</td>
<td>2 (8.3)</td>
<td>3.667 (0.658 - 20.421)</td>
<td>0.245</td>
</tr>
<tr>
<td>No</td>
<td>18 (75)</td>
<td>22 (91.7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bathing or washing clothes in the river</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>3 (12.5)</td>
<td>1 (4.2)</td>
<td>3.286 (0.317 - 34.083)</td>
<td>0.609</td>
</tr>
<tr>
<td>No</td>
<td>21 (87.5)</td>
<td>23 (93.8)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Use of personal protection during work</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>1 (4.2)</td>
<td>2 (8.3)</td>
<td>2.091 (0.177 - 24.734)</td>
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</tr>
<tr>
<td>No</td>
<td>23 (95.8)</td>
<td>22 (91.7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&quot;Kerjabaki&quot;, i.e. cleaning sewers and gutters</td>
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<td></td>
<td></td>
<td></td>
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<tr>
<td>Yes</td>
<td>10 (41.7)</td>
<td>10 (41.7)</td>
<td>1.000 (0.317 - 3.151)</td>
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</tr>
<tr>
<td>No</td>
<td>14 (58.3)</td>
<td>14 (58.3)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Presence of cattle fence surrounding the house</td>
<td></td>
<td></td>
<td>0.217 (0.022 - 2.108)</td>
<td>0.348</td>
</tr>
</tbody>
</table>

'a' could not be calculated; 'b' according to Fisher exact test.

**Figure 2: Overlay leptospirosis cases and terrain.**

**DISCUSSION**

All respondents included in the study, both cases and controls, turned out to work in the agriculture sector, so they were unable to calculate an occupational risk. Among the suggested risky habits of leptospirosis, we found four to be potential risks, even though they were not significantly associated (Table 1).

According to the USA Centers for Disease Prevention (CDC), the risk of leptospirosis can be reduced by avoiding contact with possibly contaminated water and infected animals. If such contact cannot be avoided, personal protective equipment such as footwear and protective clothing are mandatory. In our study, most of the participants reported that they did not habitually take bath or wash clothes in the river. The association of that variable could be significant even if we could not show it at the set level of statistical significance because cases and controls had almost the same behavior in this respect. However, a study conducted near the Indian ocean demonstrated that this variable is a potential risk factor for leptospirosis. Iranian research shows that swimming in a river was a risk factor for leptospirosis but not cloth washing, the better quality of life among our respondents, allowing them to bath and wash in their private facility, made it difficult to investigate this variable properly.

Regarding personal protection, the two groups of the respondent had a similar habit of not using personal protection that again made it difficult to investigate this variable. So, in this study, lack of personal protection was
not a leptospirosis risk factor. The low awareness of the respondents regarding the individual protection was similar to a study done in Brazil.25 However, our result was different from the research done in Thailand. More than 80% of participants used personal protection during their job,27 which implies that health promotion regarding personal protection is necessary to elevate the farmers' knowledge regarding prevention.

Cleaning sewers or gutters ("Kerjabakti") offers a chance for bacteria to infiltrate the human body. But in our research, both cases and controls had a similar level of this activity before infection. Accordingly, this variable was not as a potential risk factor for leptospirosis. Our finding was in contrast with recent research in Indonesia, which stated that the type of activity was a significant risk factor for leptospirosis.28

In Gunungkidul, most of the households integrate their farming activities with livestock. It is known that leptospirosis can be transmitted from these animals as well as cats, rodents, and dogs.29 Accordingly, our research approached the risk through the presence of the animal fences within 10 meters from the respondent's house. The result showed that both cases and controls had fences; however, the number was higher in the control group.

Leptospirosis is closely related to human activity and environmental aspects, one of which is the topography. In our study, we found the cases were mostly located at high altitudes in Gunungkidul District. Even though assessed the different subjects, our finding was consistent with the research in Switzerland that stated that more than 90% positive dogs lived in the altitude up to 800 m above the mean sea level.30 This study result is also strengthened with research in American Samoa that found leptospirosis cases were significantly associated with altitude and soil type.23 A previous study reported that most cases were found in the Northern (hilly) of Gunungkidul, where the soil type is mostly Lytosol and Latosol.12 This soil is low in absorbing and saving water on their structure; however, it is still possible for Leptospira to survive for a more extended live period, potentially increasing people living there to be exposed more with the exposure risk.

This study has two limitations. First, data is generated retrospectively, making it possible to have a biased memory from the respondents for a particular variable. Second, we did not include the rodent aspect as one risk factor even though it is a primary agent for leptospirosis. For future study, it is suggested to assess Leptospirosis risk factors by including two aspects: environmental factors and people's behavior comprehensively followed by spatial analysis and mapping to see the disease pattern both spatially and temporally.

CONCLUSION

This research provides evidence on a variable that could be associated with Leptospirosis cases. Human behavior is vital in Leptospirosis prevention; accordingly, this study can broaden the understanding horizon, particularly for a decision-maker where and how to implement the Leptospirosis prevention program. Prevention should address the current situation in the field and based on population and local wisdom to result in the successful implementation of the program.

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