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JOURNAL TITLE: Preventive Veterinary Medicine

VOLUME/EDITION: 121/3-4

PUBLICATION DATE: October 2015

DOI: 10.1016/j.prevetmed.2015.06.016

Reconciling surveillance systems with limited resources: an evaluation of passive surveillance for rabies in an endemic setting

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Abstract

Surveillance systems for rabies in endemic regions are often subject to severe constraints in terms of resources. The World Organisation for Animal Health (OIE) and the World Health Organisation (WHO) propose the use of an active surveillance system to substantiate claims of disease freedom, including rabies. However, many countries do not have the resources to establish active surveillance systems for rabies and the testing of dead dogs poses logistical challenges. This paper explores the potential of using a scenario tree model parameterised with data collected via questionnaires and interviews to estimate the sensitivity of passive surveillance, assessing its potential as a viable low-cost alternative to active surveillance systems. The results of this explorative study illustrated that given a large enough sample size, in this case the entire population of Colombo City, the sensitivity of passive surveillance can be 100% even at low disease prevalence (0.1%), despite the low sensitivity of individual surveillance components (mean values in the range 4.077×10^{-5} – 1.834×10^{-3} at 1% prevalence). In addition, logistic regression was used to identify factors associated with increased recognition of rabies in dogs and reporting of rabies suspect dogs. Increased recognition was observed amongst dog owners (OR 3.8 *(1.3 -10.8)*), people previously bitten by dogs(OR 5.9 *(2.2 -15.9)*) and people who believed they had seen suspect dogs in the past (OR 4.7 *(1.8 – 12.9)*). Increased likelihood of reporting suspect dogs was observed amongst dog owners (OR 5.3 *(1.1 -25*)). Further work is required to validate the data collection tool and the assumptions made in the model with respect to sample size in order to develop a robust methodology for evaluating passive rabies surveillance.

1. Introduction

Rabies is a viral zoonosis that, despite being amenable to control, continues to plague most developing countries across the world. It is estimated that there are 61,000 (95% CI 37,000–86,000) deaths caused by rabies annually across the globe (World Health Organization, 2013). In addition, rabies accounts for 1.9 million (95% CI, 1.3–2.6 million) disability-adjusted life years lost (DALY), and financial costs of US$ 6 billion (95% CI, 4.6–7.3 billion) annually (World Health Organization, 2013). In southeast Asia where 45% of all human rabies deaths occur, rabies is therefore a disease of public health and economic importance (Gongal and Wright, 2011).

In more than 99% of all cases of human rabies, the virus is transmitted directly by dogs (Knobel et al., 2005). Canine rabies can be eliminated, as demonstrated in North America, Western Europe, Japan, areas of South America and parts of Asia (Hampson et al., 2009). Advancements in post exposure prophylaxis (PEP) mean that if a person can access the appropriate post exposure vaccination and immunoglobulin therapy in a timely manner they are likely to survive (Hampson, Cleaveland, & Briggs, 2011). However, this treatment is costly, requires expertise for administration and is often not available in remote or resource poor settings where it is most needed. Without such medical intervention following infection the case fatality rate is close to 100%. Controlling and eventually eliminating the disease in dogs therefore has major benefits for public health and healthcare costs (World Health Organization, 2013).

In Colombo City, Sri Lanka, in the period 2008-2012 a combined approach to rabies management was applied, targeting improved PEP and rabies diagnostics provided by the Ministry of Health and a complementary canine control programme involving vaccination and sterilisation funded by World Animal Protection (at the time the study was conducted called World Society for the Protection of Animals- WSPA) and implemented by The Blue Paw Trust. In 2011 the human death rate due to rabies was 0.08 per 100,000 people (Häsler et al., 2014); declining from the rate of 1.7 per 100,000 in 1990 (Ministry of Health, 2007). During the combined control programme, a steady decrease in the number of confirmed canine rabies cases was observed, from 20 in 2009 to only 3 in 2012 (Häsler et al., 2014).

Surveillance is defined as any number of component activities which generate information on the health, disease or zoonosis status of animal populations to inform intervention (Corner et al., 1990). Adequate surveillance is therefore of paramount importance before, during and after any intervention to monitor technical and economic efficiency, and to inform responses to changes in prevalence (Häsler et al., 2011, Howe et al., 2013). In addition, surveillance is required to substantiate claims of disease freedom in a geographical area or region. In the case of rabies, WHO currently recommend sampling of 0.01-0.02% of the domestic animal population to substantiate freedom from rabies (WHO, 2004) this would require either a random sample of all dogs to be euthanized and tested or would rely upon a small subset of the population who are euthanized for health reasons to be tested. The latter would certainly introduce bias into the sample and the former would likely prove unfeasible and be opposed by animal rights activists. Serological testing is rarely useful for *antemortem* diagnosis because of late seroconversion and the high mortality rate of host species (Mani & Madhusudana, 2013). Differentiation between vaccinated and exposed cases is currently not feasible using serological methods and would not be appropriate in a setting employing vaccination as the control method.

The World Organisation for Animal Health (OIE) specifies that an adequate surveillance system must be in place, following the generic guidelines for surveillance systems described in the Terrestrial Animal Health Code, with no confirmed rabies cases in two years (OIE, 2011). However, while substantial progress has been made with regards intervention measures for rabies, surveillance systems are often 'poorly resourced, particularly in developing countries and especially for zoonosis which require combined veterinary and medical capacity and collaboration' (Townsend et al., 2013). This conflict, between a requirement for surveillance based on active sampling to substantiate freedom, and the availability of resources in affected countries, creates a need to evaluate alternative rabies surveillance systems with lower resource input requirements.

The evaluation of surveillance systems is an area that has grown rapidly over the past 10 years. Many techniques have been developed to assess the effectiveness of disease surveillance, of note here is the scenario tree method first described by Martin et al. (2007). In the scenario tree approach, each event from the occurrence of an infection to the detection of the case is represented with specified probabilities. The overall probability or sensitivity to detect at least one positive unit given that the population is truly infected can be calculated for each individual surveillance system component (Hadorn and Stark, 2008). Mainly used in evaluating production animal surveillance systems, it has also been employed to estimate the sensitivity of systems in place for surveillance of zoonoses, such as avian influenza (Knight-Jones et al., 2010).

Currently the surveillance system in place for rabies in Sri Lanka is a passive one. 'The approach is perceived as passive since the decision on inclusion, or exclusion, of individuals is done by the animal owners or practitioners, and not by the investigators or veterinary authorities that require the information' (Doherr and Audige, 2001). The sensitivity of passive surveillance depends on many factors including the probability of infected animals showing detectable clinical signs, the disease awareness of the public, veterinarians and health authorities and their motivation to report, as well as the sensitivity of the confirmatory test (Hadorn and Stark, 2008, Gilbert et al., 2014).

This research aimed to estimate the sensitivity of the current passive rabies surveillance in Colombo City, Sri Lanka through a scenario tree model, as well as to identify factors associated with differing levels of public rabies awareness. Evidence on the effectiveness and representativeness of passive surveillance provides important information for decision-makers in charge of disease control and allows them to identify areas where further research is required.

2. Materials and methods

2.1 Overview

To construct the scenario tree, all components of canine rabies surveillance in Colombo City were identified and detailed, considering every step needed to generate a positive laboratory diagnosis. This was based on data from scientific and grey literature and expert input from four veterinarians from The Blue Paw Trust, a local animal welfare organisation managed by veterinarians. Semi-structured interviews were undertaken with staff at the Medical Research Institute (MRI), Colombo City, and the municipality dog shelter offices to describe the surveillance processes and protocols in place. The information collected was used to derive the scenario tree (Figure 1).

From the scenario tree, the data requirements needed to parameterise the model were identified and primary and secondary data were collected. Primary data collection focused on parameterising the recognition and reporting probabilities in the scenario tree. Questionnaires were composed for members of the community and for private veterinarians (questionnaires available as supplementary materials). The administration of questionnaires is described below, and was facilitated by the community liaison officer of the Blue Paw Trust. Ethical approval for the data collection process was gained from the Royal Veterinary College ethics and welfare committee (*URN 2013 0085H)*.

2.2 Primary data collection

2.2.1 Community questionnaire

A questionnaire was designed to evaluate the ability of people in the community to recognise rabies as well as their reporting behaviour. It was also used to collect data on possible factors that were hypothesised to affect people's ability to recognise and report rabies. Questionnaires were administered in English or Sinhalese by face-to-face interviews. The enumerators gained consent from participants before commencing the interview. To avoid response bias, participants were told that the survey's aim was to gain information on people's attitudes to dogs and the general diseases they can have, rather than being specifically introduced as a rabies study.

The participant was shown five photographs of dogs: one each of dogs with distemper, mange, transmissible venereal tumours and two pictures of rabid dogs and were asked to suggest what diseases they thought the dogs had. They were also given a list of 15 symptoms and asked to identify those that would be seen in a dog with rabies. The list contained eight symptoms that are likely to occur in rabies cases and seven that are not, namely agitation, diarrhoea, excess salivation, vomiting, lethargy, hair loss, loss of appetite, eye discharge, aggression, sneezing, convulsions, nose discharge, fear of water, coughing, collapse (WHO, 2013). Ten Likert style questions with a four scale answering option were used to evaluate general rabies knowledge of the participant ("A technique for the measurement of attitudes (Book, 1932) ," n.d.). Future reporting behaviour was also evaluated using Likert style questions. A copy of the questionnaire is provided as supplementary material.

Sampling was done by convenience; the data collected was of a correlated nature with 6 respondents per socioeconomic group in each ward. Seven wards were randomly selected from the 47 wards, assuming a homogenous distribution of ward populations. Stratification by socioeconomic group was carried out in each ward; the type of housing was used to identify areas of low, middle and high socioeconomic status. Those in large detached houses enclosed by fencing were classed as high, those in apartment style blocks were classed as middle and those residing in one or two room residences in large housing schemes were considered in the low category. A central point in each ward was chosen, then houses in one particular direction visited until six houses had been visited where someone over the age of 15 was willing to participate. Four pilot questionnaires were carried out in each socioeconomic strata of the first ward; no changes were deemed necessary after the pilot.

2.2.2 Veterinarian questionnaire

A questionnaire was developed to assess the likelihood of disease recognition and reporting behaviour of private veterinarians. All of the thirteen private veterinary clinics within Colombo City were approached; seven agreed to participate in face-to-face interviews conducted in English. Questions covered the size of the practice in terms of number of veterinarians employed, number of clients seen per week and the proportion of clients who undertook rabies vaccination of their pets. Respondents were asked to identify symptoms of rabies as per the community questionnaire. Open-ended questions covered past behaviour with regard to rabies cases. The final section of the questionnaire used Likert scale questions to assess opinions on the ease of reporting to officials.

2.3 Data analysis

2.3.1 Scenario Tree

Surveillance sensitivity was estimated using the scenario tree methodology developed by Martin et al (2007). Probabilities were derived from primary and secondary data. These, and their respective notations, are detailed in Table 1 with the distributions assigned to each probability. The scenario tree (Figure 1) was built in Microsoft Excel 2011; distributions were defined in @Risk 6 (Palisade Corporation, Ithaca NY). The tree was designed to model surveillance of the whole dog population of Colombo City. Two branches represented those dogs that were owned and those not owned (labelled street dogs). This definition was made as to allow separate allocation of design prevalence in these two groups, as well as to reflect the hypothesized difference in a dog owner’s knowledge.

The unit sensitivity (CSeU), that is, the probability that an individual infected dog in Colombo City will be diagnosed under the surveillance system was estimated as follows:

$$CSeU = \sum_{k=1}^{4} P_{own} \times P_{i} \times P_{sym} \times P_{rec} \times P_{rep} \times P_{sub} \times TestSe$$

This combines the sensitivity calculated at each of k terminal ends of the tree.

The overall sensitivity of the system (CSe), i.e. the probability that the surveillance system will detect at least 1 dog as positive if rabies is present in the population at the design prevalence stated, was calculated as follows, where *n* is the dog population of Colombo City:

$$CSe = 1 - (1 - CSeU)^{n}$$

The model therefore assumes that the duration of an iteration is the timeframe in which every dog within the city is observed by a person, that is, the proportion of dogs sampled is 1.

Outputs were defined for the terminal sensitivities, the combined unit sensitivity and the overall sensitivity for the system in @Risk. The model was run stochastically for 10,000 iterations and an output detailing the mean, 5th and 95th percentiles was obtained.

A sensitivity analysis was then performed using @Risk. The output was given as a tornado plot reflecting the uncertainty in measurement of input variables for their effects on the mean of the output parameters. This was compared at different design prevalences (0.1%, 1% and 5%) designating the same prevalence in both owned and street dogs. To simulate what may happen in the future if vaccination coverage in street dogs reduces, the model was then run with 0.1% prevalence in owned dogs and 5% prevalence in street dogs.

2.3.2 Community questionnaire

All data was coded and entered into Microsoft Excel 2011. To obtain a binary recognition score a logistic regression was performed looking at correct identification of rabies pictures and individual correct symptoms. Identifying a picture was assumed to give a better representation of true recognition than being prompted by a list of symptoms so this was used as a way to define a cut off for recognition. From the logistic regression a cut off of two correct symptoms identified correlated with identifying the picture of a rabid dog. A distribution was then gained by using proportions of those who identified two correct symptoms and those who identified three as an upper and lower limit to reflect the probability of recognizing rabies in the scenario tree.

Data from questions on reporting behaviour were also combined to give a binary score. Past reporting behaviour was calculated only in those people who had previously seen a suspect case of rabies. An open-ended question on their action was then coded as 1 for actions with potential to lead to diagnosis (e.g. reporting to veterinarian or dog shelter) and 0 for actions that would not result in diagnosis (e.g. running away or burying the dog). The same technique was used to predict future reporting. There was a considerable difference in past and future reporting probabilities (35.5% and 81.8% respectively), so past reporting behaviour was used to define this probability in the scenario tree.

Statistical analysis was then performed using Stata 12.1. Descriptive statistics were obtained before carrying out univariate logistic regression with each variable for effect on the dependent variables of recognition and reporting score, to assess for any associations. After identifying variables showing significant evidence of association, forward stepwise regression was carried out with addition of significant variables into a multivariate model.

2.3.3 Veterinarian questionnaire

Answers were coded and input to Excel then analysed using Stata 12.1 to give descriptive statistics of the sample. The probability of submission of suspect samples to the MRI was calculated from this sample by coding answers as to actions taken in the past when seeing a rabies suspect dog. The number of respondents who reported they submitted samples to MRI in the past was then modelled as a beta distribution in the scenario tree to give the probability of submission.

3. Results

3.1 Community questionnaire

A total of 137 responses with equal proportions coming from each of the three socioeconomic groupings were obtained using the community questionnaires. Table 2 provides descriptive statistics of the respondents. The most common occupation listed by respondents was 'housewife' (47%), followed by 'retired' (13%). The remaining 40% reported varying occupations. 27% of respondents owned pet dogs, of which 50% let their dogs roam freely on the street.

The logistic regression to identify a cut off for recognition score identified hydrophobia, salivation and convulsions as the symptoms that were associated with the correct identification of pictures. Table 3 shows the results of univariate logistic analysis for both recognition and reporting score. When considering factors that might be associated with people's recognition score, three variables showed strong evidence of association:-owning a pet dog (p-value 0.01), having been previously bitten (p-value <0.01) and having seen a suspect case in the past (p-value <0.01). When looking for factors that may be associated with reporting score only owning a pet dog had statistical evidence of association (p-value 0.04).

From the multivariate logistic regression models it was estimated that those owning a pet dog have 3.77 the odds of recognition of rabies signs compared to those who do not own a dog (OR 3.77, p=0.027, 95% C.I 1.32 – 10.79). Those who had previously seen a suspect case had an increased likelihood of recognition, compared to those who had never seen a suspect case before, (OR 4.74, p=0.002, 95% C.I 1.75 – 12.85). Stepwise logistic regression with a likelihood ratio test showed that knowing someone who had contracted or been treated for rabies did not show significance when added to the model.

When considering reporting behaviour the model showed that those who owned a pet dog had an increased likelihood of reporting compared to people who did not own a dog, (OR 5.26, p = 0.041, 95% C.I 1.06 – 25).

3.2 Veterinarian questionnaire

There were seven respondents from seven of the thirteen private vet practices within Colombo City. Four out of seven veterinarians questioned were over 56 years old and two respondents were female. The largest practice employed fourteen vets, while four practices employed three or fewer vets. The largest reported client number seen per week was 600 and the smallest 20 clients.

When asked what action they took upon seeing suspect rabies cases, four respondents said they advised owners to tie the dog up at home, three respondents reported sending the dog's head for testing following death. Only one respondent had sent the suspect dog for isolation and monitoring at the municipality dog shelter. One practice had isolation facilities where they had isolated suspect cases; this was the only practice that reported euthanizing the dog when symptoms progressed.

Of the rabies symptoms, six respondents correctly identified six or more out of the eight correct symptoms presented. Six veterinarians reported always getting results and feedback from the laboratory. One respondent said they would report a suspect case to the dog shelter.

3.3 Scenario Tree

The overall sensitivity of the system (CSe), representing the probability of the system to detect at least one positive dog at a given infection prevalence, was found to be one (100%) throughout each prevalence estimation (**Error! Reference source not found.**). The model showed the unit sensitivity (CSeU) increased as design prevalence was increased above 1% (0.00254 at 1%, 0.0127 at 5%).

The probability of recognition in dog owners was the most influential factor on the unit sensitivity and the probability of reporting to the dog shelter by dog owners also had a profound effect on unit sensitivity, when both branches of the tree were set at the same design prevalence (Figure 2).

However, when the design prevalence was set at 5% in street dogs and 0.1% in owned dogs the mean unit sensitivity was 0.00180 (0.000936 – 0.00292). The sensitivity analysis on this model showed recognition in all members of the community to be the most influential input followed by the probability of reporting to the dog shelter.

4. Discussion

The results of this study indicate that a passive surveillance system, as parameterized within this scenario tree model, is able to achieve 100% sensitivity even at very low levels of design prevalence (0.1%). This is a reflection of the ability of a passive surveillance system to sample a much larger population than would be economically or technically feasible within many societies where rabies is endemic using an active system.

Past use of scenario tree models has focused on active surveillance systems with well-defined sample sizes. The model designed within this study assumes a timeframe per iteration in which the entire dog population is observed prior to the step at which rabies is either recognized or not recognized by the observer.

The validity of the assumption that the entire dog population of the city is sampled, or observed by a person, at each time step is worth further consideration as it is crucial to the generation of the unity of sensitivity observed in the model. In a city where approximately one quarter of the dog population is estimated to be ownerless, it is conceivable that a number of these stray dogs are not observed frequently and with sufficient scrutiny by members of the public to evaluate their health status. The human to dog contact rate for example, may be different in areas with varying ratio of human to dog population densities, meaning that contact and therefore observation of signs becomes increasingly or decreasingly likely. The form of the disease itself, and the behaviour change associated, is likely to affect the likelihood of infected animals being observed. In furious form dogs in the clinical stage are thought to be more likely to be observed. However the paralytic or dumb form is likely to reduce potential for observation since it is characterised by ataxia and paralysis. The model could be parameterised further to reflect the relative incidence of the two forms with a probability of observation adjusted accordingly and tested for effect on the system sensitivity.

Results from the scenario tree suggest that the component sensitivities for the current surveillance system are very low, this is common in rabies endemic countries (Townsend et al., 2013). This is attributable to the passive nature of the surveillance, where the onus is on the public to report.

The highest component sensitivity in the model was seen in owned dogs after being reported to the dog shelter. This is a reflection of the higher proportion of dogs being owned than street dogs, meaning more pass down this branch, combined with the fact that owners have a higher probability of recognition of rabies. Dog owners were found to have a mean probability of 0.49 of recognizing rabies compared to the public as a whole, who only had a mean 0.37 probability. This is also reflected when considering a dog owner's mean probability of reporting to the correct authority, which was estimated at 0.75, compared to the public's mean probability of 0.38. These large differences suggest that there are sectors of the public with very little knowledge of the disease. It is possible that these people may not seek out adequate treatment if they were to come into contact with rabies. Public health campaigns to increase knowledge about the disease would be beneficial to highlight symptoms of rabies. In addition, dog owners showed evidence of being highly motivated to report, but only half of them were able to recognise the disease. Hence, this group would therefore be a candidate target if it were necessary to encourage increased reporting of suspect dogs.

Not all suspect cases underwent laboratory confirmation of disease (represented by 1-Psub in the scenario tree), the proportion undergoing laboratory testing was a conservative estimate from only one expert in the field and would need further investigation to substantiate the figure.

The model was initially run with the same design prevalence for both branches. However, since the vaccination of street dogs is currently suspended, vaccination coverage is expected to decrease over time, which may lead to a higher risk of infection in the street dog population. When the model was run at a lower design prevalence to reflect this, the unit sensitivity was reduced from its already low level; however the overall system sensitivity remained at unity. Further sensitivity analysis is required to reflect an expected decline in public awareness and recognition of rabies as control activities have decreased, and prevalence of rabid dogs declined.

If the rabies campaign were to be maintained or widened such that it became realistic to consider a claim of rabies freedom two options are to be considered. Previous studies looked at targeted or risk based surveillance components and found these to give much higher sensitivities and often prove to be more cost effective (Knight-Jones, Hauser, Matthes, & Stärk, 2010). As rabies is a disease with high mortality the probability of detecting a positive case may be increased by targeting only dead or suspect animals as has been documented in rabies surveillance in wildlife (Thulke et al., 2009). It is also likely that geographical location may be a risk factor, with those wards on the outskirts of the city at higher risk of reintroduction of rabies. At present active surveillance is required to meet the standards set by the OIE to prove freedom from disease.

As is widely acknowledged (Townsend et al., 2013), the funding of rabies surveillance systems in many endemic countries is severely limited. As a result, a passive system such as the one evaluated here, if shown to be sufficiently sensitive, is likely to provide the most cost-effective and feasible for implementation in these settings. Therefore, refining the methodology proposed herein for passive surveillance systems and validating data collection instruments and model output would allow a case to be made for passive surveillance to be sufficient to substantiate a claim of rabies freedom without a prohibitively costly active surveillance component being required.

Within Sri Lanka rabies control is a public health concern rather than being of importance to trade. Proving disease freedom from rabies may not have the same economic incentive as proving freedom from certain livestock diseases, which affects trade with other countries.

It could be argued that disease free status would be beneficial for the tourist trade as tourists may be more inclined to travel to areas known to be rabies free. Since the end of the civil war in 2009, tourism in Sri Lanka has grown rapidly (visitor numbers from western Europe have more than doubled in the period 2009 to 2013 (Sri Lanka tourism development authority figures)). The need to control rabies then might become more significant in Sri Lanka as tourism becomes more important to the economy.

Another major limitation on the ability to declare freedom is the free movement of animals both into Colombo city from neighbouring areas and indeed into Sri Lanka. Being an island nation, however, countrywide control of imported canines is more plausible than in landlocked nations.

The questionnaire surveys highlighted various areas where peoples' behaviour limited the unit sensitivity. In the survey only 25% of respondents said they would contact someone if they found a dead dog and, while the cost of testing is covered by the government health service, the laboratory will only accept decapitated heads. At present then, the cost of travel to the laboratory and any costs associated with decapitation must be covered by the individual submitting the sample. In the community questionnaire 70.8% of people said they would not be willing to transport a dog head to the laboratory. If the veterinary department were designated the responsibility of decapitation and transport of samples to the laboratory, submissions of suspect cases may increase.

Respondents in both questionnaires showed little awareness of or intention to communicate with the staff of the dog shelter. Only 14.3% of vets and 30% of the community said they would contact them if they suspected a dog to have rabies. As the body responsible for canine rabies control, the dog shelter should be the contact point for anybody who suspects a dog to have rabies. Resources channelled into enhancing public use of this service would be required if it became necessary to achieve a certain sample size for a disease freedom claim. It was also indicated within the data that potential information was lost through people reporting to their veterinarian rather than to the dog shelter. Submission rates by private veterinarians were variable, with a large proportion (57.1%) of the sample reporting that they advised people to tie a suspect dog at home rather than taking any action. However, the small sample size of veterinarians means the figures derived from this source must be treated with caution with respect to representativeness.

The passive surveillance system is dependent on the general public's ability to recognise and report cases of rabies. The only factors found to be associated with recognition of rabies symptoms in the sample were the owning of a dog and having been previously bitten. This association is expected to be due to these groups being exposed to a higher volume of risk information than the general public, either through contact with veterinarians or medical services. When it came to factors that may be associated with reporting, owning a pet dog was the only variable that showed significance. Again in this situation this may reflect greater awareness through exposure to information. Alternatively, it is conceivable that dog owners feel a greater concern for canine welfare in general and are more motivated to report than the general public

For the derivation and analysis of recognition and reporting behaviour, various assumptions were made. Both scores were formulated to give a binary outcome; this was more straightforward for reporting scores where self-reported past actions defined the outcome. For recognition scores a representation of a standard rabies case had to be defined. In reality the presentation would very often vary from those portrayed. Nevertheless, the approach captures the situation in the community and allows scoring recognition ability rather than knowledge about the disease per se (Nagle et al., 2013)(Nagle, Usita, & Edland, 2013)(Nagle, Usita, & Edland, 2013).

When designating a probability for reporting behaviour it was decided to use the past-reporting behaviour score. While this represented a more reliable estimate, it considerably reduced the sample size from 137 to 30 people. The overestimation of future behaviour was most likely a result of the social desirability bias introduced. 'Social desirability reflects the tendency on behalf of the subjects to deny socially undesirable traits and to claim sociably desirable ones' (Nederhof, 1985). While efforts were made to try to minimise this effect, this was inhibited by the face-to face format of the questionnaire. Another major limitation in data collection was introduced by response bias. No information was available on those who refused to partake or those who were not at home at time of interviewing. Interviews were carried out during the weekdays and meant that there was an over representation of those who were at home during these times, such as housewives, the retired and the unemployed. The use of convenience sampling instead of formal random sampling in the questionnaires is likely to have introduced further bias, however in this setting a conventional random sampling would have proved unworkable.

Data to parameterize the submission rate from the dog shelter was limited. It was reported during verbal interview that no dogs had been held as suspect cases over the last year but no records were available to corroborate this information. As a result, the probability was determined to be between 80-90% from the verbal information gained when asking the dog shelter manager what proportion he believed to have been submitted in the past.

5. Conclusion

The dedication of different agencies within Colombo City to control rabies is encouraging; a lot of progress has been made towards eradication within the city. This is a novel setting for the use of the scenario tree model, but the results obtained in this research highlight its functionality. While the passive surveillance system currently in place in Colombo City has low unit sensitivity, the model indicates that with sufficient sample size this need not inhibit overall system sensitivity. Further work however is needed to validate the sample size assumptions used in these calculations. The passive surveillance system seen in Colombo City in Sri Lanka and other similar systems are commonplace in most rabies endemic countries where more costly active surveillance systems are difficult to implement. This project therefore provides a framework that could be widely utilised to evaluate surveillance in countries battling to control the rabies burden. Implementation of cost-effective monitoring of control efforts and facilitating the ability to substantiate freedom from disease using a passive system provide incentives for rabies control.

Conflict of interest

No conflict of interest.

Role of funding source

RVC MSc course funded research. Barbara Häsler acknowledges financial support from the Leverhulme Centre for Integrative Research on Agriculture and Health (LCIRAH).

Acknowledgements

The authors wish to thank all the staff at the Blue Paw Trust, Colombo City, for their advice and assistance in collecting field data. Thanks are given to Dr. Kanthi Nanayakkara of the Medical Research Institute and Dr. Mohammed Ijaz from the dog shelter for their time and sharing of data, and to all the respondents in the field.

References

CORNER, L., MELVILLE, L., MCCUBBIN, K., SMALL, K. J., MCCORMICK, B. S., WOOD, P. R. & ROTHEL, J. S. 1990. Efficiency of inspection procedures for the detection of tuberculous lesions in cattle. *Aust Vet J,* 67**,** 389-92.

DOHERR, M. G. & AUDIGE, L. 2001. Monitoring and surveillance for rare health-related events: a review from the veterinary perspective. *Philos Trans R Soc Lond B Biol Sci,* 356**,** 1097-106.

GILBERT, W. H., HÄSLER, B. N. & RUSHTON, J. 2014. Influences of farmer and veterinarian behaviour on emerging disease surveillance in England and Wales. *Epidemiol Infect,* 142**,** 172-86.

GONGAL, G. & WRIGHT, A. E. 2011. Human Rabies in the WHO Southeast Asia Region: Forward Steps for Elimination. *Adv Prev Med,* 2011**,** 383870.

HADORN, D. C. & STARK, K. D. 2008. Evaluation and optimization of surveillance systems for rare and emerging infectious diseases. *Vet Res,* 39**,** 57.

HAMPSON, K., CLEAVELAND, S., & BRIGGS, D. (2011). Evaluation of Cost-Effective Strategies for Rabies Post-Exposure Vaccination in Low-Income Countries. *PLoS Neglected Tropical Diseases*, *5*(3), 11. Retrieved from http://dx.doi.org/10.1371/journal.pntd.0000982

HAMPSON, K., DUSHOFF, J., CLEAVELAND, S., HAYDON, D. T., KAARE, M., PACKER, C., & DOBSON, A. (2009). Transmission dynamics and prospects for the elimination of canine rabies. *PLoS Biology*, *7*(3), e53. doi:10.1371/journal.pbio.1000053

HÄSLER, B., HIBY, E., GILBERT, W., OBEYESEKERE, N., BENNANI, H. & RUSHTON, J. 2014. A one health framework for the evaluation of rabies control programmes: a case study from colombo city, sri lanka. *PLoS Negl Trop Dis,* 8**,** e3270.

HÄSLER, B., HOWE, K. S. & STARK, K. D. 2011. Conceptualising the technical relationship of animal disease surveillance to intervention and mitigation as a basis for economic analysis. *BMC Health Serv Res,* 11**,** 225.

HOWE, K. S., HÄSLER, B. & STARK, K. D. 2013. Economic principles for resource allocation decisions at national level to mitigate the effects of disease in farm animal populations. *Epidemiol Infect,* 141**,** 91-101.

KNIGHT-JONES, T. J., HAUSER, R., MATTHES, D. & STARK, K. D. 2010. Evaluation of effectiveness and efficiency of wild bird surveillance for avian influenza. *Vet Res,* 41**,** 50.

KNOBEL, D. L., CLEAVELAND, S., COLEMAN, P. G., FEVRE, E. M., MELTZER, M. I., MIRANDA, M. E., SHAW, A., ZINSSTAG, J. & MESLIN, F. X. 2005. Re-evaluating the burden of rabies in Africa and Asia. *Bull World Health Organ,* 83**,** 360-8.

MARTIN, P. A., CAMERON, A. R. & GREINER, M. 2007. Demonstrating freedom from disease using multiple complex data sources 1: a new methodology based on scenario trees. *Prev Vet Med,* 79**,** 71-97. A technique for the measurement of attitudes (Book, 1932) . (n.d.). Retrieved April 28, 2015, from http://www.worldcat.org/title/technique-for-the-measurement-of-attitudes/oclc/812060&referer=brief_results

Hampson, K., Cleaveland, S., & Briggs, D. (2011). Evaluation of Cost-Effective Strategies for Rabies Post-Exposure Vaccination in Low-Income Countries. *PLoS Neglected Tropical Diseases*, *5*(3), 11. Retrieved from http://dx.doi.org/10.1371/journal.pntd.0000982

Hampson, K., Dushoff, J., Cleaveland, S., Haydon, D. T., Kaare, M., Packer, C., & Dobson, A. (2009). Transmission dynamics and prospects for the elimination of canine rabies. *PLoS Biology*, *7*(3), e53. http://doi.org/10.1371/journal.pbio.1000053

Knight-Jones, T. J. D., Hauser, R., Matthes, D., & Stärk, K. D. C. (2010). Evaluation of effectiveness and efficiency of wild bird surveillance for avian influenza. *Veterinary Research*, *41*(4), 50. http://doi.org/10.1051/vetres/2010023

Mani, R. S., & Madhusudana, S. N. (2013). Laboratory diagnosis of human rabies: recent advances. *TheScientificWorldJournal*, *2013*, 569712. http://doi.org/10.1155/2013/569712

Ministry of Health. (2007). Annual Health Statisitics, Sri Lanka.

Nagle, B. J., Usita, P. M., & Edland, S. D. (2013). United States medical students' knowledge of Alzheimer disease. *Journal of Educational Evaluation for Health Professions*, *10*(4), 4. http://doi.org/10.3352/jeehp.2013.10.4

Townsend, S. E., Lembo, T., Cleaveland, S., Meslin, F. X., Miranda, M. E., Putra, A. A. G., … Hampson, K. (2013). Surveillance guidelines for disease elimination: a case study of canine rabies. *Comparative Immunology, Microbiology and Infectious Diseases*, *36*(3), 249–61. http://doi.org/10.1016/j.cimid.2012.10.008

WHO. (2004). WHO EXPERT CONSULTATION ON RABIES, *1*.

WHO. (2013). WHO Expert Consultation on Rabies.

NAGLE, B. J., USITA, P. M. & EDLAND, S. D. 2013. United States medical students' knowledge of Alzheimer disease. *J Educ Eval Health Prof,* 10**,** 4.

NEDERHOF, A. J. 1985. Methods of coping with social desirability bias: A review. *European Journal of Social Psychology,* 15**,** 263-280.

OIE 2011. *Terrestrial animal health code Volime II: recommendations applicable to OIE listed diseases and other diseases of importance to international trade,* Paris, OIE. *http://www.oie.int/index.php?id=169&L=0&htmfile=chapitre_surveillance_general.htm#chapitre_surveillance_general accessed July 2013*

THULKE, H.-H., EISINGER, D., FREULING, C., FRÖHLICH, A., GLOBIG, A., GRIMM, V., MÜLLER, T., SELHORST, T., STAUBACH, C. & ZIPS, S. 2009. Situation-based surveillance: Adapting investigations to actual epidemic situations. *Journal of Wildlife Diseases,* 45**,** 1089-1103.

TOWNSEND, S. E., LEMBO, T., CLEAVELAND, S., MESLIN, F. X., MIRANDA, M. E., PUTRA, A. A., HAYDON, D. T. & HAMPSON, K. 2013. Surveillance guidelines for disease elimination: a case study of canine rabies. *Comp Immunol Microbiol Infect Dis,* 36**,** 249-61.

WORLD HEALTH ORGANIZATION. 2013. *RE: WHO expert consultation on rabies. Second report. World Health Organization Technical Report Series, (982).*

Table 1

Abbreviation	Value or distribution	Description and data source
recognition	Beta(24+1,37-24+1)	Upper estimate of recognition ability from community questionnaire. (Probability of recognising two of the three symptoms)
recognition2	Beta(12+1,37-12+1)	Lower estimate of recognition ability from community questionnaire. (Probability of recognising all three symptoms)
Pown	Beta(15640+1,21377-15640+1)	The probability that a dog is owned, taken from the 2007 dog census of Colombo City.
Pi	0.01	The design prevalence, assumption based on past prevalence data from the Blue Paw Trust.
Psym	1	The probability that a dog infected with rabies will show clinical symptoms.
Prec_O	Uniform (recognition2(O),recognition(O))	The probability that a dog owner recognises the signs of rabies, calculated from the community survey.
Prec_SD	Uniform (recognition2(SD),recognition(SD))	The probability that any person (dog owner or not)recognises the signs of rabies, calculated from the community survey.
PrepDP_O	Beta(6+1,10-6+1)	The probability that a dog owner would report to the dog shelter if they suspect a dog has rabies, calculated form the community survey.
PrepV_O	Beta(1+1,10-1+1)	The probability that a dog owner would report to the veterinarian if they suspect a dog has rabies, calculated form the community survey.
PrepDP_SD	Beta(9+1,30-9+1))	The probability that a person would report to the dog shelter if they suspect a street dog has rabies, calculated form the community survey.
PrepV_SD	Beta(1+1,30-1+1))	The probability that a person would report to the veterinarian if they suspect a street dog has rabies, calculated form the community survey.
Psub	Pert(0.8,0.9,1)	The probability that the veterinarians at the dog shelter will submit the head for rabies testing.
PsubV	Beta(5+1,7-5+1)	The probability that the veterinarian will submit the head for rabies testing, calculated from the survey amongst private vets.
Testse	0.98	The diagnostic sensitivity of the laboratory diagnosis from expert opinion.
n	21,377	Dog population of Colombo City from 2007 census data provided by Blue Paw Trust.

Table 2

Variable	Category	Frequency
Gender	Male	48 (35.0%)
	Female	89 (65.0%)
Religion (12 missing values)	Buddhist	66 (52.8%)
	Christian	27 (21.6%)
	Hindu	20 (16.0%)
	Muslim	9 (7.2%)
	None	3 (2.4%)
Age group	15-36 years	42 (30.7%)
	36-65 years	70 (51.1%)
	66+ years	25 (18.3%)
Children <5yr in household	Yes	35 (25.6%)
	No	102 (74.5%)
Children <15yr in household	Yes	77 (56.2%)
	No	60 (43.8%)
Education level reached	No formal education	11 (0.7%)
	Primary	34 (24.8%)
	Secondary	83 (60.6%)
	Undergraduate	16 (11.7%)
	Postgraduate	3 (2.2%)
Dog Owner	Yes	37 (27.0%)
	No	100 (73.0%)
Number of stray dogs in neighbourhood	0	35 (25.9%)
	1-5	60 (44.4%)
	6-10	32 (23.7%)
	11-15	4 (3.0%)
	15+	4 (3.0%)
Previously bitten by dog	Yes	50 (36.5%)
	No	87 (63.5%)
Known anyone who has contracted or been treated for rabies	Yes	19 (13.9%)
	No	118 (86.1%)
Seen a suspect rabid dog in the past	Yes	30 (21.9%)
	No	107 (78.1%)

Table 3

Factors potentially associated with recognition and reporting of rabies	Univariate logistic regression with recognition score		Univariate logistic regression with reporting score	
	Odds Ratio (95% C.I)	**P value**	**Odds Ratio (95% C.I)**	**P value**
Socioeconomic group		0.52		0.6
Low	1		1	
Middle	0.811 (0.23-2.83)		1.81 (0.6-5.47)	
High	0.47 (0.12-1.77)		1.14 (0.42-3.14)	
Gender		0.08		0.06
Male	1		1	
Female	0.44(0.17-1.11)		2.38 (0.99-5.75)	
Religion (12 missing values)	-	-	-	-
Age group		0.88	5.88 (0.72 – 50)	0.3
15-35yrs	1		1	
36-65yrs	1.14 (0.41-3.18)		3.5 (0.35-35-38)	
66+yrs	0.82 (0.2-3.34)		9 (0.56-143.95)	
Children <5yr in household		0.4		0.48
Yes	1		1	
No	1.63 (0.52-5.09)		1.93 (0.32-11.74)	
Children <15yr in household		0.36		0.15
Yes	1		1	
No	1.54 (0.62-3.83)		3.26 (0.66-16.03)	
Education level reached	-	-	-	-
Dog owner		0.01		0.04
Yes	1		1	
No	0.27 (0.09-0.76)		0.19 (0.04-0.94)	
No. stray dogs in neighbourhood		0.65		1.00
None	1		1	
1-5	0.67 (0.2-2.21)		0.95 (0.14-6.28)	
6-10	1.89 (0.48-7.55)		1 (0.13-7.57)	
11-15	-		-	
20-30	1.95 (012.-31.32)		-	
Previously bitten by a dog		0.0005		0.81
Yes	1		1	
No	0.17 (0.06-0.46)		0.83 (0.19-3.64)	

Known anyone contracted or treated for rabies		0.27		0.64
Yes	1		1	
No	0.5 (0.14-1.74)		0.67 (0.12-3.73)	
Seen suspect rabid dog in past		0.002		0.43
Yes	1		1	
No	0.21 (0.08-0.57)		0.63 (0.2-2.0)	

Figure and table legends

Figure 1. Scenario tree for the passive surveillance of rabies in the dog population of Colombo City, Sri Lanka.

Figure 2. Sensitivity analysis of scenario tree parameters, exploring influence on mean unit sensitivity of passive surveillance for rabies in the dog population of Colombo City, Sri Lanka.

Figure 3. Output of the stochastic scenario tree model. Mean sensitivity for each terminal, as well as unit and overall sensitivity is given with 5% and 95% percentiles Model output at 0.1%, 1% and 5% design prevalence in both branches are given. Se = component sensitivity, SD = street dog, O = owned dog, CSeU = unit sensitivity, Pound = reporting to dog shelter, Vet = reporting to vet.

Table 1. Parameters used in the scenario tree model, distributions applied and data sources.

Table 2. Frequency of demographic and descriptive categorical data amongst respondents to the community survey.

Table 3. Univariate logistic regression results from the community survey for factors associated with increased or decreased recognition and reporting of rabies suspect animals.

Figure 1.

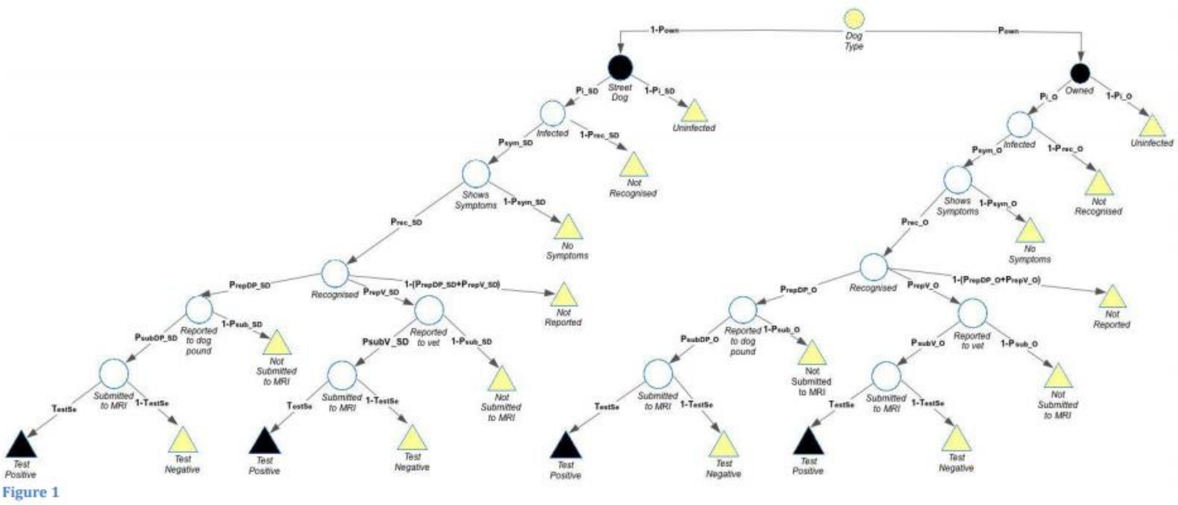

Figure 2:

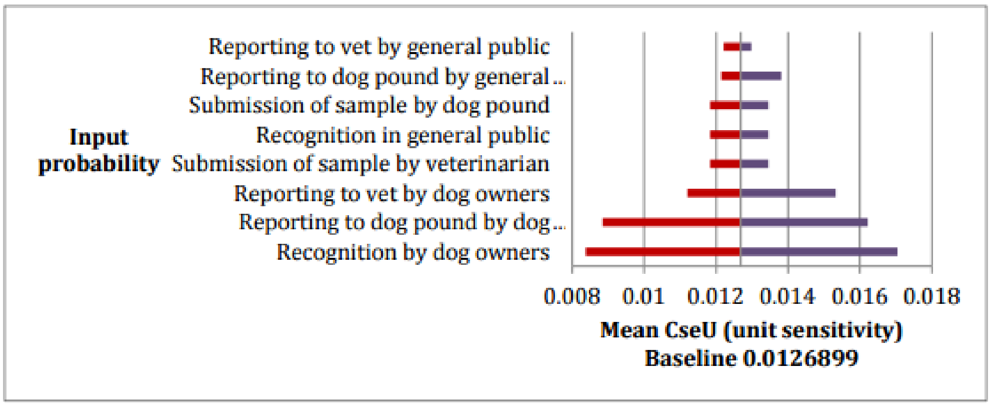

Figure 2

Figure 3:

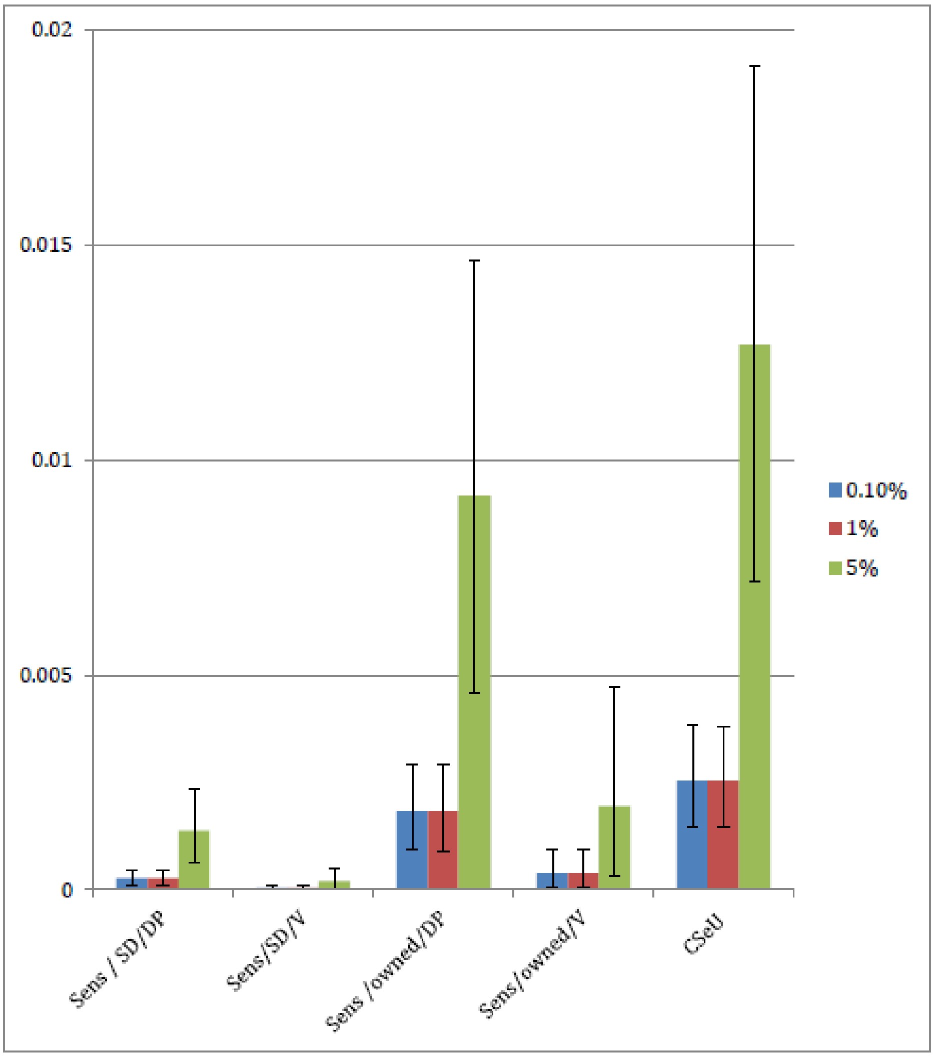

Figure 3