

This is the peer reviewed version of the following article:

Craighead, L., W. Gilbert, D. Subasinghe and B. Häsler (2015). "Reconciling surveillance systems with limited resources: an evaluation of passive surveillance for rabies in an endemic setting." *Preventive Veterinary Medicine* 121(3–4): 206-214.

Which is available in final form via <http://dx.doi.org/10.1016/j.prevetmed.2015.06.016>.

This manuscript is made available under the CC-BY-NC-ND 4.0 license  
<http://creativecommons.org/licenses/by-nc-nd/4.0/>.

The full details of the published version of the article are as follows:

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

AUTHORS: Craighead, L., W. Gilbert, D. Subasinghe and B. Häsler

JOURNAL TITLE: Preventive Veterinary Medicine

VOLUME/EDITION: 121/3-4

PUBLICATION DATE: October 2015

DOI: 10.1016/j.prevetmed.2015.06.016

# 1 Reconciling surveillance systems with limited resources: an 2 evaluation of passive surveillance for rabies in an endemic 3 setting

4 *Laura Craighead<sup>a</sup>, William Gilbert<sup>a</sup>, Dynatra Subasinghe<sup>c</sup>, Barbara Häsler<sup>a,b</sup>*

5 <sup>a.</sup> Royal Veterinary College, North Mymms, Hertfordshire, United Kingdom

6 <sup>b.</sup> Leverhulme Centre for Integrative Research on Agriculture and Health, Royal Veterinary College,  
7 North Mymms, Hertfordshire, United Kingdom

8 <sup>c.</sup> Blue Paw Trust, Colombo, Sri Lanka

9 **Corresponding author:** Royal Veterinary College, North Mymms, Hertfordshire, United Kingdom,

10 lcraighead2@rvc.ac.uk, +44 7900817039

## 11 Abstract

12 Surveillance systems for rabies in endemic regions are often subject to severe constraints in terms of  
13 resources. The World Organisation for Animal Health (OIE) and the World Health Organisation  
14 (WHO) propose the use of an active surveillance system to substantiate claims of disease freedom,  
15 including rabies. However, many countries do not have the resources to establish active surveillance  
16 systems for rabies and the testing of dead dogs poses logistical challenges. This paper explores the  
17 potential of using a scenario tree model parameterised with data collected via questionnaires and  
18 interviews to estimate the sensitivity of passive surveillance, assessing its potential as a viable low-  
19 cost alternative to active surveillance systems. The results of this explorative study illustrated that  
20 given a large enough sample size, in this case the entire population of Colombo City, the sensitivity  
21 of passive surveillance can be 100% even at low disease prevalence (0.1%), despite the low  
22 sensitivity of individual surveillance components (mean values in the range  $4.077 \times 10^{-5}$  –  $1.834 \times 10^{-3}$   
23 at 1% prevalence). In addition, logistic regression was used to identify factors associated with  
24 increased recognition of rabies in dogs and reporting of rabies suspect dogs. Increased recognition

25 was observed amongst dog owners (OR 3.8 (1.3 -10.8) ), people previously bitten by dogs(OR 5.9 (2.2  
26 -15.9 )) and people who believed they had seen suspect dogs in the past (OR 4.7 (1.8 – 12.9)).  
27 Increased likelihood of reporting suspect dogs was observed amongst dog owners (OR 5.3 (1.1 -25)).  
28 Further work is required to validate the data collection tool and the assumptions made in the model  
29 with respect to sample size in order to develop a robust methodology for evaluating passive rabies  
30 surveillance.

## 31 **1. Introduction**

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

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

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

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

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

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

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

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

## 103 **2. Materials and methods**

### 104 **2.1 Overview**

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

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

119 **2.2 Primary data collection**

120 **2.2.1 Community questionnaire**

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

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

139 Sampling was done by convenience; the data collected was of a correlated nature with 6  
140 respondents per socioeconomic group in each ward. Seven wards were randomly selected from the  
141 47 wards, assuming a homogenous distribution of ward populations. Stratification by socioeconomic  
142 group was carried out in each ward; the type of housing was used to identify areas of low, middle  
143 and high socioeconomic status. Those in large detached houses enclosed by fencing were classed as

144 high, those in apartment style blocks were classed as middle and those residing in one or two room  
145 residences in large housing schemes were considered in the low category. A central point in each  
146 ward was chosen, then houses in one particular direction visited until six houses had been visited  
147 where someone over the age of 15 was willing to participate. Four pilot questionnaires were carried  
148 out in each socioeconomic strata of the first ward; no changes were deemed necessary after the  
149 pilot.

### 150 *2.2.2 Veterinarian questionnaire*

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

## 159 *2.3 Data analysis*

### 160 *2.3.1 Scenario Tree*

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



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

$$171 \quad CSeU = \sum_{k=1}^4 P_{own} \times P_i \times P_{sym} \times P_{rec} \times P_{rep} \times P_{sub} \times TestSe$$

172

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

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

$$177 \quad CSe = 1 - (1 - CSeU)^n$$

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

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

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

### 189 *2.3.2 Community questionnaire*

190 All data was coded and entered into Microsoft Excel 2011. To obtain a binary recognition score a  
191 logistic regression was performed looking at correct identification of rabies pictures and individual

192 correct symptoms. Identifying a picture was assumed to give a better representation of true  
193 recognition than being prompted by a list of symptoms so this was used as a way to define a cut off  
194 for recognition. From the logistic regression a cut off of two correct symptoms identified correlated  
195 with identifying the picture of a rabid dog. A distribution was then gained by using proportions of  
196 those who identified two correct symptoms and those who identified three as an upper and lower  
197 limit to reflect the probability of recognizing rabies in the scenario tree.

198 Data from questions on reporting behaviour were also combined to give a binary score. Past  
199 reporting behaviour was calculated only in those people who had previously seen a suspect case of  
200 rabies. An open-ended question on their action was then coded as 1 for actions with potential to  
201 lead to diagnosis (e.g. reporting to veterinarian or dog shelter) and 0 for actions that would not  
202 result in diagnosis (e.g. running away or burying the dog). The same technique was used to predict  
203 future reporting. There was a considerable difference in past and future reporting probabilities  
204 (35.5% and 81.8% respectively), so past reporting behaviour was used to define this probability in  
205 the scenario tree.

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

### 211 *2.3.3 Veterinarian questionnaire*

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

## 217        **3. Results**

### 218        **3.1 Community questionnaire**

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

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

232        From the multivariate logistic regression models it was estimated that those owning a pet dog have  
233        3.77 the odds of recognition of rabies signs compared to those who do not own a dog (OR 3.77,  
234        p=0.027, 95% C.I 1.32 – 10.79). Those who had previously seen a suspect case had an increased  
235        likelihood of recognition, compared to those who had never seen a suspect case before, (OR 4.74,  
236        p=0.002, 95% C.I 1.75 – 12.85). Stepwise logistic regression with a likelihood ratio test showed that  
237        knowing someone who had contracted or been treated for rabies did not show significance when  
238        added to the model.

239 When considering reporting behaviour the model showed that those who owned a pet dog had an  
240 increased likelihood of reporting compared to people who did not own a dog, (OR 5.26,  $p = 0.041$ ,  
241 95% C.I 1.06 – 25).

### 242 3.2 Veterinarian questionnaire

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

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

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

### 255 3.3 Scenario Tree

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

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

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

#### 267 **4. Discussion**

268 The results of this study indicate that a passive surveillance system, as parameterized within this  
269 scenario tree model, is able to achieve 100% sensitivity even at very low levels of design prevalence  
270 (0.1%). This is a reflection of the ability of a passive surveillance system to sample a much larger  
271 population than would be economically or technically feasible within many societies where rabies is  
272 endemic using an active system.

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

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

288 model could be parameterised further to reflect the relative incidence of the two forms with a  
289 probability of observation adjusted accordingly and tested for effect on the system sensitivity.

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

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

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

309 The model was initially run with the same design prevalence for both branches. However, since the  
310 vaccination of street dogs is currently suspended, vaccination coverage is expected to decrease over  
311 time, which may lead to a higher risk of infection in the street dog population. When the model was

312 run at a lower design prevalence to reflect this, the unit sensitivity was reduced from its already low  
313 level; however the overall system sensitivity remained at unity. Further sensitivity analysis is  
314 required to reflect an expected decline in public awareness and recognition of rabies as control  
315 activities have decreased, and prevalence of rabid dogs declined.

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

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

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

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

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

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

352 Respondents in both questionnaires showed little awareness of or intention to communicate with  
353 the staff of the dog shelter. Only 14.3% of vets and 30% of the community said they would contact  
354 them if they suspected a dog to have rabies. As the body responsible for canine rabies control, the  
355 dog shelter should be the contact point for anybody who suspects a dog to have rabies. Resources  
356 channelled into enhancing public use of this service would be required if it became necessary to  
357 achieve a certain sample size for a disease freedom claim. It was also indicated within the data that  
358 potential information was lost through people reporting to their veterinarian rather than to the dog  
359 shelter. Submission rates by private veterinarians were variable, with a large proportion (57.1%) of



360 the sample reporting that they advised people to tie a suspect dog at home rather than taking any  
361 action. However, the small sample size of veterinarians means the figures derived from this source  
362 must be treated with caution with respect to representativeness.

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

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

379 When designating a probability for reporting behaviour it was decided to use the past-reporting  
380 behaviour score. While this represented a more reliable estimate, it considerably reduced the  
381 sample size from 137 to 30 people. The overestimation of future behaviour was most likely a result  
382 of the social desirability bias introduced. 'Social desirability reflects the tendency on behalf of the  
383 subjects to deny socially undesirable traits and to claim sociably desirable ones' (Nederhof, 1985).

384 While efforts were made to try to minimise this effect, this was inhibited by the face-to face format

385 of the questionnaire. Another major limitation in data collection was introduced by response bias.  
386 No information was available on those who refused to partake or those who were not at home at  
387 time of interviewing. Interviews were carried out during the weekdays and meant that there was an  
388 over representation of those who were at home during these times, such as housewives, the retired  
389 and the unemployed. The use of convenience sampling instead of formal random sampling in the  
390 questionnaires is likely to have introduced further bias, however in this setting a conventional  
391 random sampling would have proved unworkable.

392 Data to parameterize the submission rate from the dog shelter was limited. It was reported during  
393 verbal interview that no dogs had been held as suspect cases over the last year but no records were  
394 available to corroborate this information. As a result, the probability was determined to be between  
395 80-90% from the verbal information gained when asking the dog shelter manager what proportion  
396 he believed to have been submitted in the past.

## 397 **5. Conclusion**

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

## 410 **Conflict of interest**

411 No conflict of interest.

412

## 413 **Role of funding source**

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

417

## 418 **Acknowledgements**

419 The authors wish to thank all the staff at the Blue Paw Trust, Colombo City, for their advice and

420 assistance in collecting field data. Thanks are given to Dr. Kanthi Nanayakkara of the Medical

421 Research Institute and Dr. Mohammed Ijaz from the dog shelter for their time and sharing of data,

422 and to all the respondents in the field.

## 423 **References**

- 424 CORNER, L., MELVILLE, L., MCCUBBIN, K., SMALL, K. J., MCCORMICK, B. S., WOOD, P. R. &  
425 ROTHEL, J. S. 1990. Efficiency of inspection procedures for the detection of tuberculous lesions in  
426 cattle. *Aust Vet J*, 67, 389-92.
- 427 DOHERR, M. G. & AUDIGE, L. 2001. Monitoring and surveillance for rare health-related events: a review  
428 from the veterinary perspective. *Philos Trans R Soc Lond B Biol Sci*, 356, 1097-106.
- 429 GILBERT, W. H., HÄSLER, B. N. & RUSHTON, J. 2014. Influences of farmer and veterinarian behaviour on  
430 emerging disease surveillance in England and Wales. *Epidemiol Infect*, 142, 172-86.
- 431 GONGAL, G. & WRIGHT, A. E. 2011. Human Rabies in the WHO Southeast Asia Region: Forward Steps for  
432 Elimination. *Adv Prev Med*, 2011, 383870.
- 433 HADORN, D. C. & STARK, K. D. 2008. Evaluation and optimization of surveillance systems for rare and  
434 emerging infectious diseases. *Vet Res*, 39, 57.
- 435 HAMPSON, K., CLEAVELAND, S., & BRIGGS, D. (2011). Evaluation of Cost-Effective Strategies for Rabies  
436 Post-Exposure Vaccination in Low-Income Countries. *PLoS Neglected Tropical Diseases*, 5(3), 11.  
437 Retrieved from <http://dx.doi.org/10.1371/journal.pntd.0000982>
- 438 HAMPSON, K., DUSHOFF, J., CLEAVELAND, S., HAYDON, D. T., KAARE, M., PACKER, C., &  
439 DOBSON, A. (2009). Transmission dynamics and prospects for the elimination of canine rabies. *PLoS*  
440 *Biology*, 7(3), e53. doi:10.1371/journal.pbio.1000053
- 441 HÄSLER, B., HIBY, E., GILBERT, W., OBEYESEKERE, N., BENNANI, H. & RUSHTON, J. 2014. A one  
442 health framework for the evaluation of rabies control programmes: a case study from colombo city, sri  
443 lanka. *PLoS Negl Trop Dis*, 8, e3270.
- 444 HÄSLER, B., HOWE, K. S. & STARK, K. D. 2011. Conceptualising the technical relationship of animal  
445 disease surveillance to intervention and mitigation as a basis for economic analysis. *BMC Health Serv*  
446 *Res*, 11, 225.
- 447 HOWE, K. S., HÄSLER, B. & STARK, K. D. 2013. Economic principles for resource allocation decisions at  
448 national level to mitigate the effects of disease in farm animal populations. *Epidemiol Infect*, 141, 91-  
449 101.
- 450 KNIGHT-JONES, T. J., HAUSER, R., MATTHES, D. & STARK, K. D. 2010. Evaluation of effectiveness and  
451 efficiency of wild bird surveillance for avian influenza. *Vet Res*, 41, 50.

- 452 KNOBEL, D. L., CLEAVELAND, S., COLEMAN, P. G., FEVRE, E. M., MELTZER, M. I., MIRANDA, M.  
453 E., SHAW, A., ZINSSTAG, J. & MESLIN, F. X. 2005. Re-evaluating the burden of rabies in Africa  
454 and Asia. *Bull World Health Organ*, 83, 360-8.
- 455 MARTIN, P. A., CAMERON, A. R. & GREINER, M. 2007. Demonstrating freedom from disease using  
456 multiple complex data sources 1: a new methodology based on scenario trees. *Prev Vet Med*, 79, 71-97. A  
457 technique for the measurement of attitudes (Book, 1932) . (n.d.). Retrieved April 28, 2015, from  
458 [http://www.worldcat.org/title/technique-for-the-measurement-of-](http://www.worldcat.org/title/technique-for-the-measurement-of-attitudes/oclc/812060&referer=brief_results)  
459 [attitudes/oclc/812060&referer=brief\\_results](http://www.worldcat.org/title/technique-for-the-measurement-of-attitudes/oclc/812060&referer=brief_results)
- 460 Hampson, K., Cleaveland, S., & Briggs, D. (2011). Evaluation of Cost-Effective Strategies for Rabies Post-  
461 Exposure Vaccination in Low-Income Countries. *PLoS Neglected Tropical Diseases*, 5(3), 11. Retrieved  
462 from <http://dx.doi.org/10.1371/journal.pntd.0000982>
- 463 Hampson, K., Dushoff, J., Cleaveland, S., Haydon, D. T., Kaare, M., Packer, C., & Dobson, A. (2009).  
464 Transmission dynamics and prospects for the elimination of canine rabies. *PLoS Biology*, 7(3), e53.  
465 <http://doi.org/10.1371/journal.pbio.1000053>
- 466 Knight-Jones, T. J. D., Hauser, R., Matthes, D., & Stärk, K. D. C. (2010). Evaluation of effectiveness and  
467 efficiency of wild bird surveillance for avian influenza. *Veterinary Research*, 41(4), 50.  
468 <http://doi.org/10.1051/vetres/2010023>
- 469 Mani, R. S., & Madhusudana, S. N. (2013). Laboratory diagnosis of human rabies: recent advances.  
470 *TheScientificWorldJournal*, 2013, 569712. <http://doi.org/10.1155/2013/569712>
- 471 Ministry of Health. (2007). Annual Health Statistics, Sri Lanka.
- 472 Nagle, B. J., Usita, P. M., & Edland, S. D. (2013). United States medical students' knowledge of Alzheimer  
473 disease. *Journal of Educational Evaluation for Health Professions*, 10(4), 4.  
474 <http://doi.org/10.3352/jeehp.2013.10.4>
- 475 Townsend, S. E., Lembo, T., Cleaveland, S., Meslin, F. X., Miranda, M. E., Putra, A. A. G., ... Hampson, K.  
476 (2013). Surveillance guidelines for disease elimination: a case study of canine rabies. *Comparative*  
477 *Immunology, Microbiology and Infectious Diseases*, 36(3), 249-61.  
478 <http://doi.org/10.1016/j.cimid.2012.10.008>
- 479 WHO. (2004). WHO EXPERT CONSULTATION ON RABIES, 1.
- 480 WHO. (2013). WHO Expert Consultation on Rabies.
- 481 NAGLE, B. J., USITA, P. M. & EDLAND, S. D. 2013. United States medical students' knowledge of  
482 Alzheimer disease. *J Educ Eval Health Prof*, 10, 4.
- 483 NEDERHOF, A. J. 1985. Methods of coping with social desirability bias: A review. *European Journal of Social*  
484 *Psychology*, 15, 263-280.
- 485 OIE 2011. *Terrestrial animal health code Volume II: recommendations applicable to OIE listed diseases and*  
486 *other diseases of importance to international trade*, Paris, OIE.  
487 [http://www.oie.int/index.php?id=169&L=0&htmfile=chapitre\\_surveillance\\_general.htm#chapitre\\_sur-](http://www.oie.int/index.php?id=169&L=0&htmfile=chapitre_surveillance_general.htm#chapitre_surveillance_general)  
488 [veillance\\_general accessed July 2013](http://www.oie.int/index.php?id=169&L=0&htmfile=chapitre_surveillance_general.htm#chapitre_surveillance_general)
- 489 THULKE, H.-H., EISINGER, D., FREULING, C., FRÖHLICH, A., GLOBIG, A., GRIMM, V., MÜLLER, T.,  
490 SELHORST, T., STAUBACH, C. & ZIPS, S. 2009. Situation-based surveillance: Adapting  
491 investigations to actual epidemic situations. *Journal of Wildlife Diseases*, 45, 1089-1103.
- 492 TOWNSEND, S. E., LEMBO, T., CLEAVELAND, S., MESLIN, F. X., MIRANDA, M. E., PUTRA, A. A.,  
493 HAYDON, D. T. & HAMPSON, K. 2013. Surveillance guidelines for disease elimination: a case study  
494 of canine rabies. *Comp Immunol Microbiol Infect Dis*, 36, 249-61.
- 495 WORLD HEALTH ORGANIZATION. 2013. *RE: WHO expert consultation on rabies. Second report. World*  
496 *Health Organization Technical Report Series*, (982).

498

499 **Table 1**

500

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

501

502 **Table 2**

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

503

504

505

506 **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
<b>Socioeconomic group</b>			0.52		0.6
	<b>Low</b>	1		1	
	<b>Middle</b>	0.811 (0.23-2.83)		1.81 (0.6-5.47)	
	<b>High</b>	0.47 (0.12-1.77)		1.14 (0.42-3.14)	
<b>Gender</b>			0.08		0.06
	<b>Male</b>	1		1	
	<b>Female</b>	0.44(0.17-1.11)		2.38 (0.99-5.75)	
<b>Religion (12 missing values)</b>	-	-	-	-	-
<b>Age group</b>			0.88	5.88 (0.72 – 50)	0.3
	<b>15-35yrs</b>	1		1	
	<b>36-65yrs</b>	1.14 (0.41-3.18)		3.5 (0.35-35-38)	
	<b>66+yrs</b>	0.82 (0.2-3.34)		9 (0.56-143.95)	
<b>Children &lt;5yr in household</b>			0.4		0.48
	<b>Yes</b>	1		1	
	<b>No</b>	1.63 (0.52-5.09)		1.93 (0.32-11.74)	
<b>Children &lt;15yr in household</b>			0.36		0.15
	<b>Yes</b>	1		1	
	<b>No</b>	1.54 (0.62-3.83)		3.26 (0.66-16.03)	
<b>Education level reached</b>	-	-	-	-	-
<b>Dog owner</b>			0.01		0.04
	<b>Yes</b>	1		1	
	<b>No</b>	0.27 (0.09-0.76)		0.19 (0.04-0.94)	
<b>No. stray dogs in neighbourhood</b>			0.65		1.00
	<b>None</b>	1		1	
	<b>1-5</b>	0.67 (0.2-2.21)		0.95 (0.14-6.28)	
	<b>6-10</b>	1.89 (0.48-7.55)		1 (0.13-7.57)	
	<b>11-15</b>	-		-	
	<b>20-30</b>	1.95 (0.12.-31.32)		-	
<b>Previously bitten by a dog</b>			0.0005		0.81
	<b>Yes</b>	1		1	
	<b>No</b>	0.17 (0.06-0.46)		0.83 (0.19-3.64)	

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

507



508

509 **Figure and table legends**

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

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

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

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

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

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

523

524 Figure 1.

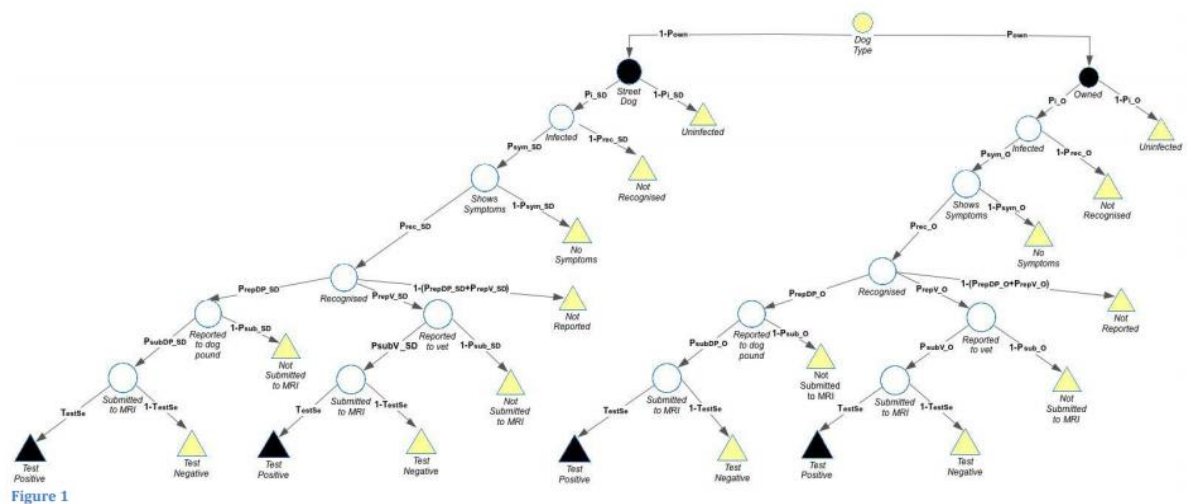


Figure 1

525

526

527 Figure 2:

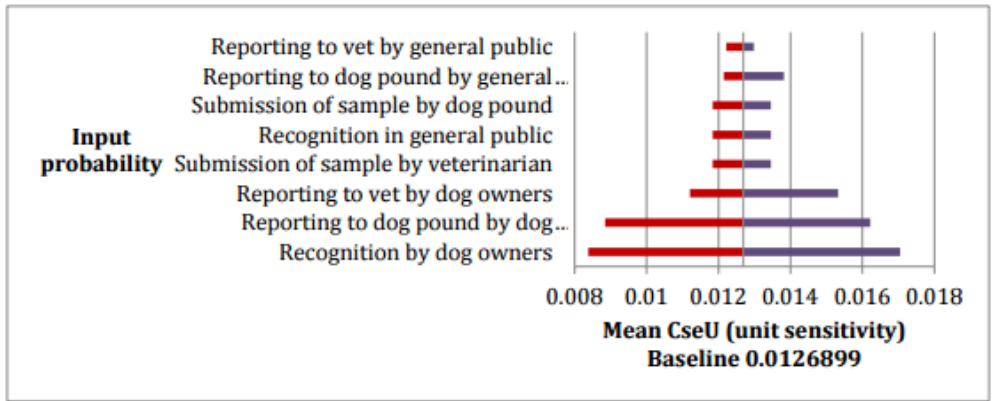


Figure 2

528

529 Figure 3:

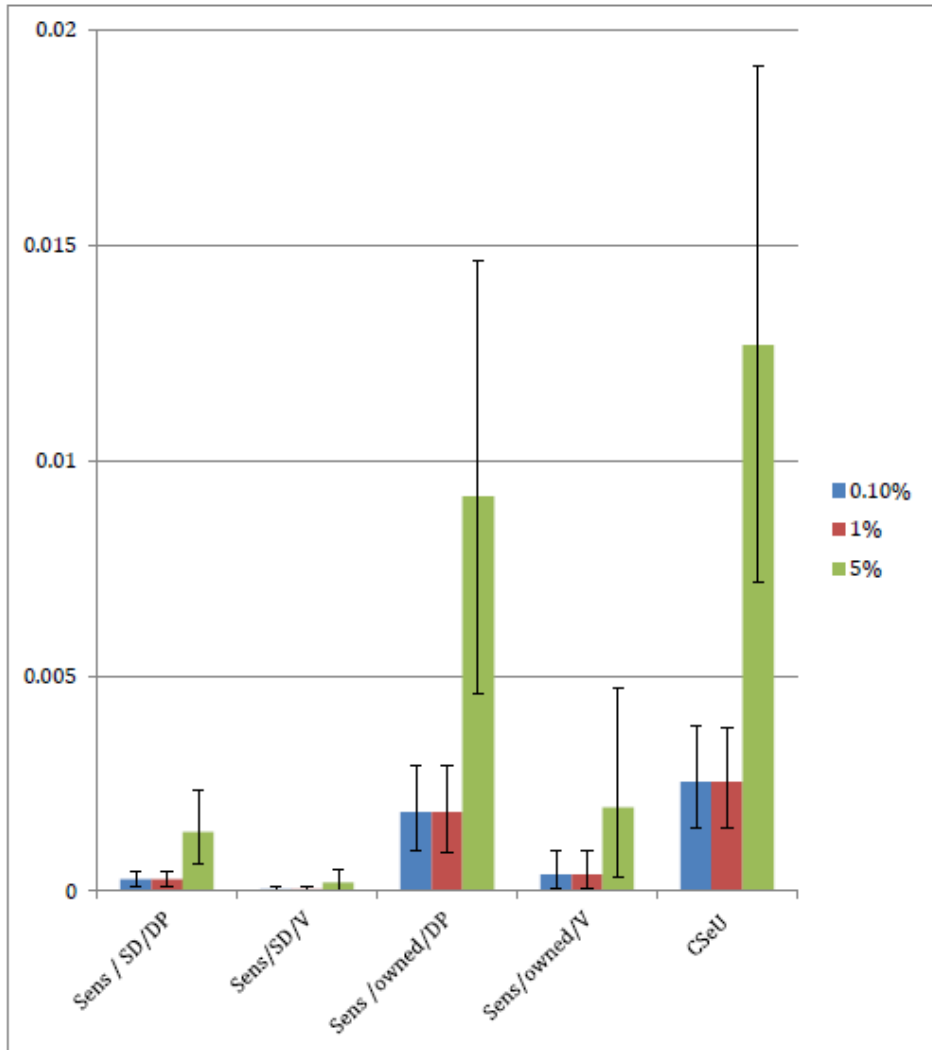


Figure 3

530