Applications of Uni-List Capture-Recapture Methods in Meta-Analysis

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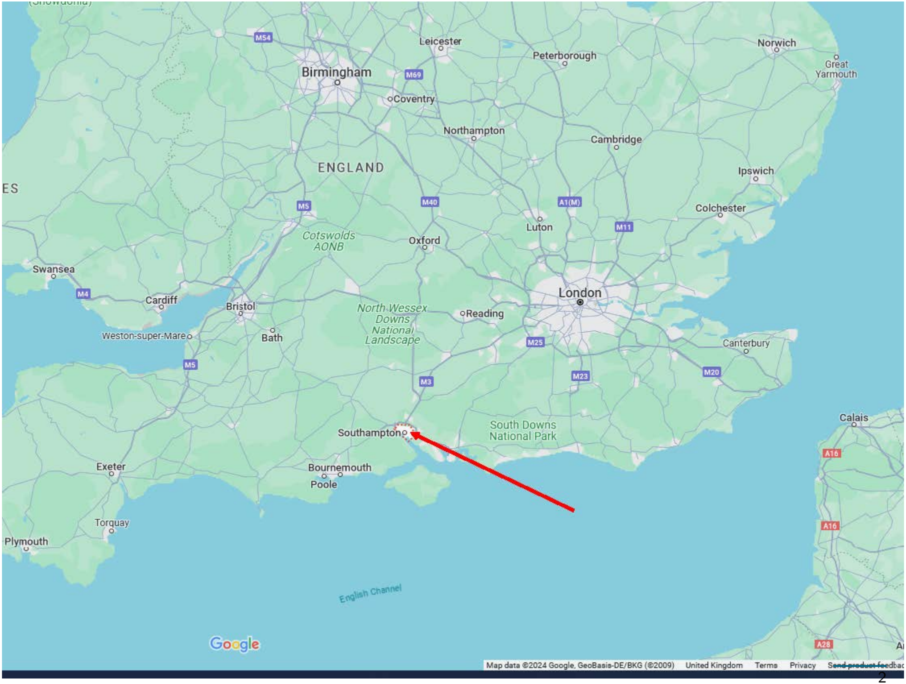
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Key facts

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Duration: up to four years (full-time); up to seven years (part-time); dependent on funding route: Integrated PhD, PhD, 1+3

Start date: September (Integrated PhD, PhD, 1+3); sometimes possible throughout the year (PhD only)

Applying: University application form with transcripts, research proposal and two references

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Assessment: progression from year one to year two of Integrated PhD by taught courses; annual reports, confirmation (for PhD award), thesis and viva*
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Assessment: progression from year one on 1+3 by examination taught courses; annual reports, confirmation (for PhD award), thesis and viva*
Closing date: none, but early application advised
Funding: may be available through University's Vice-Chancellor's Scholarship programme
Additional costs: fieldwork, printing and photocopying, etc.; some help may be provided

* For more information on continued assessment throughout your research programme, see page 19

Carla Azevedo

Research themes

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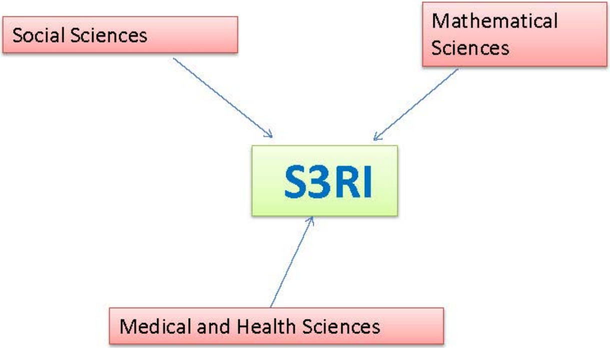
Social Sciences
Mathematical Sciences
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Medical and Health Sciences

obesity reviews

doi: 10.1111/obr.12014

Obesity Treatment

Risk of completed suicide after bariatric surgery: a systematic review

C. Peterhänsel[1,2], D. Petroff[3,4], G. Klinitzke[1,2], A. Kersting[1] and B. Wagner[1,2]

Case-study: Obesity Treatment
Risk of completed suicide after bariatric surgery: a systematic review

- bariatric surgery is one of the most effective treatments for morbid obesity, indicating a significant long-term weight loss
- while overall mortality decreases in patients who received bariatric surgery, risk of suicide is still an issue
- Peterhänsel et al. (2013) undertake a meta-analysis on completed suicide after bariatric surgery
- 27 studies are included in the analysis

Table 2 List of papers included for the estimate of the suicide rate in decreasing order of person-years

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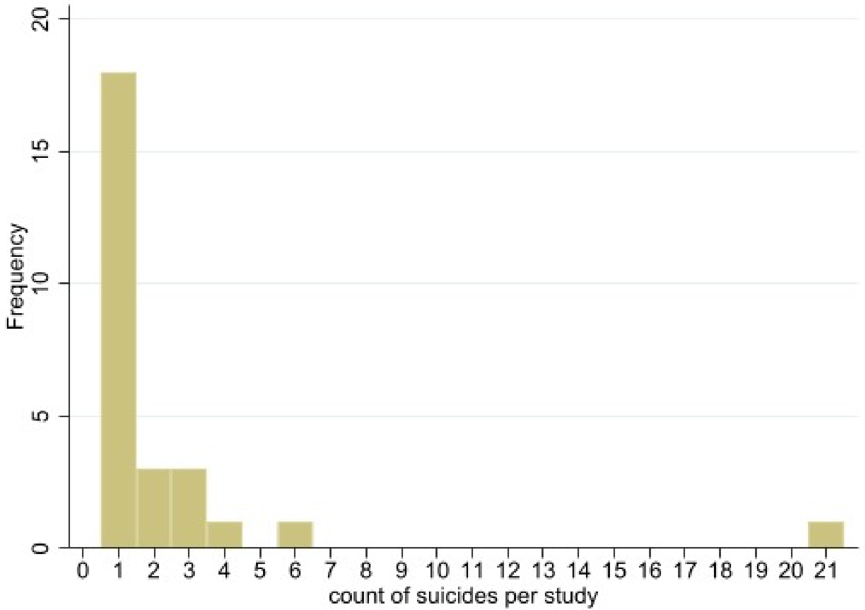
Frequency
0
5
10
15
20
0 1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16 17 18 19 20 21
count of suicides per study

Case-study: Obesity Treatment
Risk of completed suicide after bariatric surgery: a systematic review

- selection bias issue: only studies *with* completed suicide are included
- Peterhänsel et al. (2013):

  > *The most crucial point in the analysis was the proper treatment of the selection bias because of the method of finding papers.*

- hence, suicide rate will be *overestimated* (potentially substantially)

conventional meta-analysis

- in a nutshell, the conventional approach for a meta-analytic analysis (Cooper and Hedges 1994, Egger *et al.* 1995, Stangl and Berry 2000, Borenstein *et al.* 2009:311) proceed as follows:
- let X_i denote the observed count of suicides in study i and $E(X_i) = \mu_i$ its corresponding expected value
- also, let P_i denote the person-years in study i
- Then, in meta-analysis a summary measure as a weighted average of the study-specific rates on log-scale is used:

$$\sum_{i=1}^{n} w_i \log(X_i/P_i) / \sum_{i=1}^{n} w_i$$

where w_i is a proxy estimate of the inverse variance, here $w_i = Y_i$ leading to

$$\sum_{i=1}^{n} Y_i \log(X_i/P_i) / \sum_{i=1}^{n} Y_i$$

conventional meta-analysis

- another approach (Barendregt *et al.* 2013) works on the rate scale
- an attractive choice for w_i in

$$\sum_{i=1}^{n} w_i(X_i/P_i)/\sum_{i=1}^{n} w_i$$

is

$$w_i = P_i$$

- this is in the Mantel-Haenszel philosophy weighting with the denominator (here the person-years) leading to

$$\hat{\lambda} = \sum_{i=1}^{n} X_i/\sum_{i=1}^{n} P_i$$

as a summary estimate of the overall rate λ

conventional meta-analysis

- a benefit of the Mantel-Haenszel approach here is that the variance of $\hat{\lambda}$ is easy to calculate:

$$Var(\hat{\lambda}) = Var(\sum_{i=1}^{n} X_i / \sum_{i=1}^{n} P_i)$$

$$= \sum_{i=1}^{n} \lambda P_i / (\sum_{i=1}^{n} P_i)^2$$

- which is estimated as

$$\hat{\lambda} / \sum_{i=1}^{n} P_i$$

- using this technique we find an overall rate of 44.51 suicides per $100,000$ person years with a 95% CI of $33.60 - 55.42$

problem with the conventional approach

- any of these conventional approaches cope with zero-event studies missing
- hence we need to turn to other ideas

the idea of capture-recapture

- objective is to determine the size N of an elusive target population
- some mechanism (life trapping, register, surveillance system) identifies a unit repeatingly
- there is a count X informing about the number of identifications of each unit in the target population

sample

available: sample

$$X_1, X_2, ..., X_N$$

leading to

Table: Frequency distribution of count X of repeated identifications

x	0	1	2	3	4	...	population size
f_x	f_0	f_1	f_2	f_3	f_4	...	N

problem

if $X_i = 0$ unit is not observed leading to a reduced observable sample

$$X_1, X_2, ..., X_n$$

where – w.l.g. – we assume that

$$X_{n+1} = X_{n+2} = ... = X_N = 0$$

Table: Frequency distribution of count X of repeated identifications

x	0	1	2	3	4	...	observed size
f_x	-	f_1	f_2	f_3	f_4	...	n

hence

$$f_0 = N - n \text{ is unknown}$$

why does data set fit into the capture-recapture setting?

- target population: *studies* on bariatric surgery with or without completed suicide
- identifying mechanism: online web-search including databases PubMed (PM), Web of Knowledge (WK), PsychInfo (PI), ScienceDirect (SD) and Google Scholar (GS)
- X_i number of completed suicides in study i: can be viewed as the count of repeated identifications for study i

modelling

- to cope with missing zeros we need to involve modelling
- $p_x = P(X = x)$ for $x = 0, 1, 2, \cdots$ base model
- for example *Poisson* :

$$p_x = \exp(-\mu)\mu^x/x! = \exp(-\lambda P)(\lambda P)^x/x!$$

λ suicide rate, P person-time, $\mu = \lambda P$

Table: Frequency distribution of count X of repeated identifications

x	0	1	2	3	4	...	m
f_x	-	f_1	f_2	f_3	f_4	...	f_m
p_x	p_0	p_1	p_2	p_3	p_4	...	p_m

modelling

- need to incorporate study-specific person-times
- $p_{ix} = P(X_i = x|P_i)$ probab. for x events in study with person-time P_i
- for example *Poisson* :

$$p_{ix} = \exp(-\lambda P_i)(\lambda P_i)^x / x!$$

  λ suicide rate, P_i person-time in study i, $\mu = \lambda P$

- complete data likelihood

$$\prod_{i=1}^{n} \prod_{x=0}^{m} p_{ix}^{f_{ix}}$$

  where f_{ix} is the frequency of studies with person-time P_i and event count x

- in our case, for given P_i the frequency f_{ix} is zero except for one value of x where it is one

EM philosophy: E-step

f_{i0} is unknown and needs to be replaced by its expected value: $E-step$

there is a general solution for the E-step:

$$e_{i0} := E(f_{i0}|f_{i1}, \cdots, f_{in}; P_i) = N_i p_{i0}$$

where N_i is the population size of studies with person-time P_i

it follows that

$$e_{i0} = N_i p_{i0} = (n_i + e_i) p_{i0}$$

where $n_i = f_{i1} + \cdots + f_{in}$ ($= 1$ in our case)

it follows further that

$$e_{i0} = n_i \frac{p_{i0}}{1 - p_{i0}}$$

which *replaces* f_{i0} in the complete, unobserved likelihood leading to the complete, expected likelihood

EM philosophy: E-step

note the relationship to the $Horvitz - Thompson$ estimator:

$$\hat{N}_i = n_i + e_{i0} = n_i + n_i \frac{p_{i0}}{1 - p_{i0}} = \frac{n_i}{1 - p_{i0}}$$

and

$$\hat{N} = \sum_{i=1}^{n} \hat{N}_i = \sum_{i=1}^{n} \frac{n_i}{1 - p_{i0}}$$

in the case study we have that $n_i = 1$ for $i = 1, \cdots, n$

the E-step provides as $by - product$ the item we are most interested in: the count of studies with no suicides, alternatively, the total number of studies

EM philosophy: M-step

we need to maximize the *complete*, *expected* data likelihood

$$\prod_{i=1}^{n}\prod_{x=1}^{m} p_{ix}^{f_{ix}} p_{i0}^{e_{i0}}$$

the solution will *depend* on the model used: in the *Poisson* case the complete data log-likelihood is

$$\sum_{i=1}^{n}\sum_{x=1}^{m} f_{ix}[-\mu_i + x\log\mu_i] - e_{i0}\mu_i$$

with $\mu_i = \lambda P_i$ which is maximized for

$$\hat{\lambda} = \frac{\sum_{i=1}^{n}\sum_{x=1}^{m} x\ f_{ix}}{\sum_{i=1}^{n}(\sum_{x=1}^{m} P_i f_{ix} + P_i e_{i0})}$$

EM philosophy

now, the EM algorithm toggles between E- and M-step until convergence

$$\text{E-step} \longleftrightarrow \text{M-step}$$

```
start rate MH: 0.0004451183

step:  1 rate:  0.000353999 size:  121.9951
step:  2 rate:  0.000329974 size:  129.6188
step:  3 rate:  0.000321995 size:  132.4051
step:  4 rate:  0.000319157 size:  133.4304
step:  5 rate:  0.000318122 size:  133.8086
...
step:  14 rate:  0.0003175201 size:  134.03
step:  15 rate:  0.0003175201 size:  134.03
```

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The column entitled 'weight' is the fraction of the total number of person-years and is used in the analysis for comparing the estimated suicide rate for patients after a bariatric operation with the rate for an equivalent general population.

EM philosophy: full set of covariates

here an illustration in the Poisson case

$$p_{ix} = P(X_i = x|\beta; \mathbf{z_i}) = \exp(-\mu_i)\mu_i^x/x!$$

and

$$\log \mu_i = \beta^T \mathbf{z_i}$$

if there are *only* person-times

$$\log \mu_i = \log \lambda + \log P_i$$

EM philosophy

complete data likelihood – *with* covariates

$$\prod_{i=1}^{n} \prod_{x=0}^{m} p_{ix}^{f_{ix}}$$

where

- $p_{ix} = P(X_i = x | \beta; \mathbf{z_i})$
- $\mathbf{z_i}$ represents the i-th covariate combination for $i = 1, \cdots, n$
- f_{ix} is the frequency of observed counts equal to x for the i-th covariate combination
- f_{i0} remains unknown

E-step

we have

$$e_{i0} = n_i \frac{p_{i0}}{1 - p_{i0}}$$

with $p_{i0} = P(X_i = 0|\beta; \mathbf{z_i})$

M-step

to maximize

$$\prod_{i=1}^{n} \prod_{x=1}^{m} p_{ix}^{f_{ix}} p_{i0}^{e_{i0}}$$

this is model dependent; in the Poisson case with log-link

$$p_{ix} = P(X_i = x|\beta; \mathbf{z_i}) = \exp(-\mu_i)\mu_i^x / x!,$$

with $\log \mu_i = \beta^T \mathbf{z_i}$

M-step for the Poisson case with only person-times

$$p_{ij} = P(X_i = j|\beta; \mathbf{z_i}) = \exp(-\mu_i)\mu_i^j/j!$$

and

$$\mu_i = \exp(\eta + \underbrace{\log P_i}_{\text{log-person-times become offset}})$$

so, here simply

$$\mu_i = \exp(\beta^T \mathbf{z_i}) = \exp(\eta + \log P_i)$$

where η is the log-rate

alternatives to the EM philosophy

- use the observed, zero-truncated likelihood directly:

$$\prod_{i=1}^{n} \prod_{x=1}^{m} \left(\frac{p_{ix}}{1 - p_{i0}}\right)^{f_{ix}}$$

  where $p_{ix} = P(X_i = x | \beta; \mathbf{z_i})$ as before
- depends on the chosen model (Poisson, geometric, binomial, negative-binomial,...)
- use favorite algorithm such as NR, FS, or GN
- retrieve effect estimate $\hat{\beta}$

population size estimation with Horvitz-Thompson

$Horvitz - Thompson\ estimator$

$$\hat{N} = \sum_{i=1}^{N} I_i / w_i$$

where

- I_i is an indicator if the i-th study of the population of target studies is observed
- $w_i = P(I_i = 1) = 1 - P(I_i = 0) = 1 - p_{i0} = 1 - P(X_i = 0|\hat{\beta}; \mathbf{z_i})$
- under Poisson: $w_i = 1 - \exp(-\mu_i)$ and $\hat{\mu}_i = \exp(\hat{\beta}^T \mathbf{z_i})$

so that

$$\hat{N} = \sum_{i=1}^{n} 1/[1 - \exp(\hat{\beta}^T \mathbf{z_i})]$$

study population size estimation

so, in case we have use only person-times as offset

$$\hat{N} = \sum_{i=1}^{n} 1/[1 - \exp(-\exp(\hat{\eta} + \log PT_i))]$$

for the data

$$\hat{N} = \sum_{i=1}^{n} 1/[1 - \exp(\exp(\hat{\eta} + \log PT_i)] = 134$$

total studies with and *without* completed suicide after bariatric surggery

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The column entitled 'weight' is the fraction of the total number of person-years and is used in the analysis for comparing the estimated suicide rate for patients after a bariatric operation with the rate for an equivalent general population.

practical modelling

Table: Linear predictors considered

Linear predictor	Proportion of women	Country of origin	Interaction	log-person-time as offset
0	No	No	No	No
1	No	No	No	Yes
2	Yes	No	No	Yes
3	No	Yes	No	Yes
4	Yes	Yes	No	Yes
5	Yes	Yes	Yes	Yes

Table: Values of the maximised log-likelihood, number of parameters, and BIC statistic s for models under consideration.

Distribution	LP	Maximised log-likelihood	Number of parameters	BIC
Poisson	5	-22.7	4	58.6
	4	-23.0	3	55.9
	3	-23.0	2	52.6
	2	-23.4	2	53.4
	1	-23.7	1	**50.7**
	0	-68.7	1	139.9
Negative-binomial	5	-22.7	5	61.9
	4	-23.0	4	59.2
	3	-23.0	3	55.9
	2	-23.4	3	56.7
	1	-23.7	2	54.0
	0	-38.7	2	84.0

uncertainty assessment with the bootstrap

- in principle, we have a population of size N
- for each element i we have an indicator I_i telling us if element i has been sampled or not

$$I_i = \begin{cases} 1, & \text{if sampled} \\ 0, & \text{otherwise} \end{cases}$$

  where $i = 1, ..., N$

- the classical nonparametric bootstrap would then consider random samples with replacement from $I_1, ..., I_N$
- problem is that we have *only* observed n out of N
- using the observed sample $I_1, ..., I_n$ for the bootstrap would *underestimate* the variability of $\hat{N}$
- the idea is to impute N using $\hat{N}$

uncertainty assessment with the bootstrap

Horvitz − Thompson estimator

$$\hat{N} = \sum_{i=1}^{N} I_i / \hat{w}_i$$

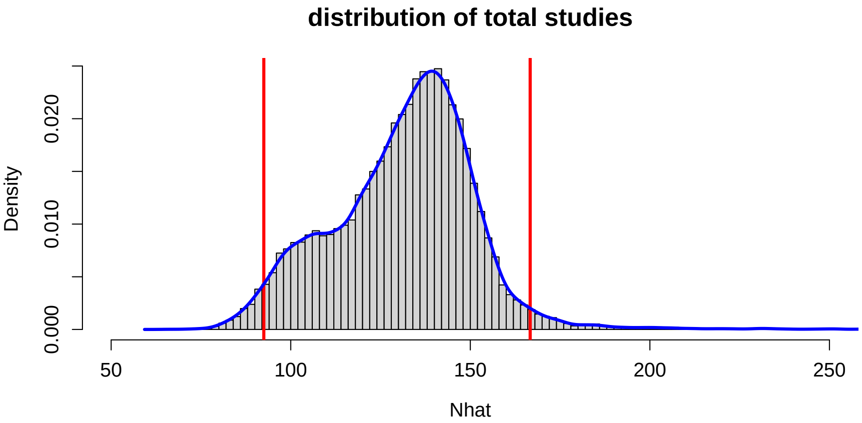
where

- $\hat{w}_i = \hat{P}(I_i = 1) = 1 - \hat{P}(I_i = 0)$
- under Poisson: $\hat{w}_i = 1 - \exp(-\hat{\mu}_i)$ and $\hat{\mu}_i = \exp(\hat{\beta}^T \mathbf{z_i})$
- or $\hat{N} = \sum_{i=1}^{n} 1/[1 - \exp(-\exp(\hat{\beta}^T \mathbf{z_i})]$
- this gives our imputed sample $I_1, ... I_n, ... I_{\hat{N}}$
- note that $I_{n+1}, ... I_{\hat{N}}$ are all zero ($\hat{N}$ needs to be rounded)

uncertainty assessment with the bootstrap

finally

- we can consider bootstrap samples $I_1^*, \ldots I_{\hat{N}}^*$
- note that there is now variability in the observed sample size n
- as all elements in the bootstrap sample with zero counts are truncated, it does not matter that we have *no* covariate information on the truncated counts
- using the zero-truncated bootstrap sample we estimate $\hat{N}^*$
- this process is repeated B times ($B = 25,000$ for example)



- *median* $= 133$ studies on bariatic surgery with or without completed suicide
- 95% *percentile confidence interval*: $93 - 167$ (red vertical bars)

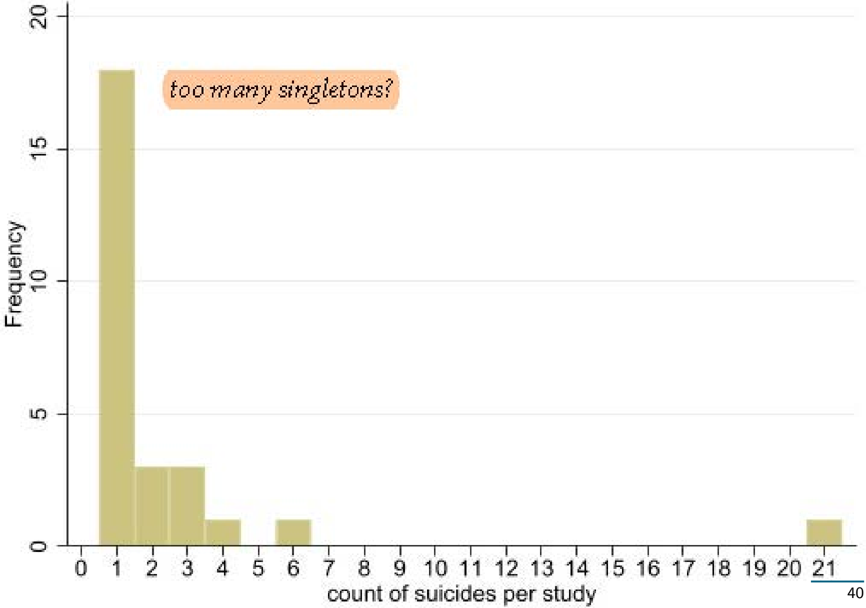
uncertainty assessment with the bootstrap

- in a similar way a 95% percentile confidence interval for the suicide rate is computed
- $24.84 - 49.39$ per $100,000$ person years
- with median rate of 31.86 per $100,000$ person years
- for comparison: the unadjusted rate is 44.51 per $100,000$ person years

acknowledgments

- joint work with Layna Dennett and Antony Overstall (University of Southampton)
- a paper version is available at:
- Layna Charlie Dennett, Antony Overstall, Dankmar Böhning (2023): Zero-Truncated Modelling Meta-Analysis for When Studies with No Events Are Systematically Excluded: Estimating Completed Suicide After Bariatric Surgery. https://arxiv.org/abs/2305.01277

further issues: one-inflation



further issues: one-inflation

Figure: The Guardian 30 Dec 2016: "Thousands of drink-drivers offend again"

drink-driving in Britain

- drink-driving (DD) relates to driving (or attempting to drive) while being above the legal alcohol limit
- according to the Guardian (30/12/16): 219,000 motorist were caught once, 8,068 twice, etc. (see Table below)

Table: Frequency distribution of the count (per person) of DVLA reported drink-driving (DD) in the UK between 2011 and 2015 (figures are based on DR10 endorsements)

count of DD	f_0	f_1	f_2	f_3	f_4	f_5	f_6	n
frequency		219,008	8,068	449	46	5	2	227,578

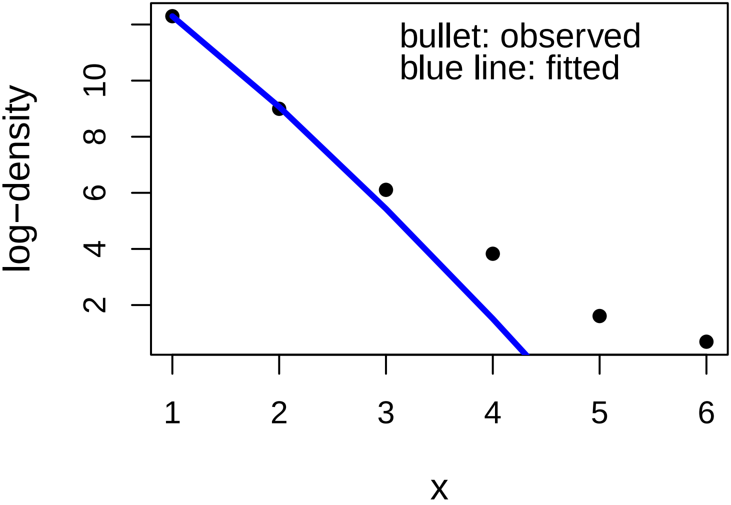


Figure: One-inflation distorts the Poisson fit

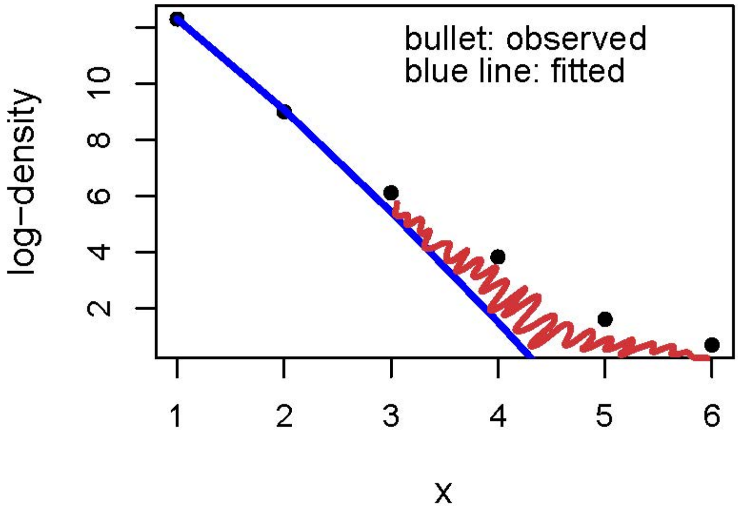


Figure: One-inflation distorts the Poisson fit

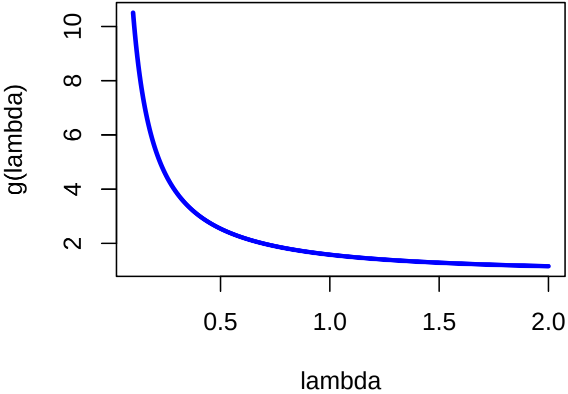
a synthetic example

- 500 counts sampled from $Po(1)$
- 500 extra-counts of 1 so that $N = 1,000$
- $\hat{\lambda} = 0.4091$ and

$$\text{HTE} = \frac{n}{1 - \exp(-\hat{\lambda})} = \frac{824}{1 - \exp(-0.4091)} = 2454$$

Table: one-inflated Poisson data

f_0	f_1	f_2	f_3	f_{4+}	n
176	690	95	32	7	824



- one-inflation leads to $\hat{\lambda} << \lambda$
- Horvitz-Thompson estimator $n \frac{1}{1-\exp(-\hat{\lambda})} >> N$
- as $g(\lambda) = \frac{1}{1-\exp(-\lambda)}$ strictly decreasing

two processes

- do not know the size: $zero - truncation$
- many counts of ones (singletons): $one - inflation$

this can be modelled as

$$(1-w)l_1(x) + \frac{w}{1-\ p(0;\theta)}p(x;\theta)$$

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THE IDENTITY OF THE ZERO-TRUNCATED, ONE-INFLATED LIKELIHOOD AND THE ZERO-ONE-TRUNCATED LIKELIHOOD FOR GENERAL COUNT DENSITIES WITH AN APPLICATION TO DRINK-DRIVING IN BRITAIN

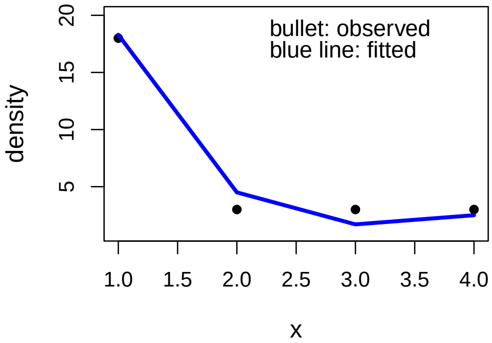
BY DANKMAR BÖHNING AND PETER G. M. VAN DER HEIJDEN

University of Southampton and University of Utrecht

GOF in the case study

Table: Frequency distribution for observed and fitted count of completed suicide under zero-truncated Poisson with offset for person-times; $\chi^2_{(2)} = 1.59$ and $p - \text{value} = 0.45$

count of completed suicide	0	1	2	3	4+
observed frequency f_x	-	18	3	3	3
fitted frequency $\hat{f}_x$	-	18.3	4.5	1.7	2.5



how to present fitted frequency for complex model

suppose a model (here for a Poisson with log-link) the has been fitted leading to

$$\hat{\mu}_i = \exp(\hat{\beta}^T \mathbf{z_i})$$

for unit i in the sample, then:

$$\hat{f}_x = \sum_{i=1}^{n} \exp(-\hat{\mu}_i)\hat{\mu}_i^x / x!$$

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SAGE journals

Article

The covariate-adjusted frequency plot

Heinz Holling[1], Walailuck Böhning[1], Dankmar Böhning[2], and Anton K Formann[3,†]

alternative: Bayes

- posterior $\propto$ likelihood $\times$ prior
- in our case

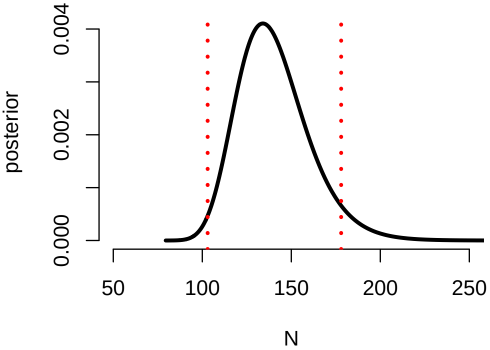
$$\pi(\lambda|x_1,\cdots,x_n) \propto \underbrace{\prod_i \frac{\exp(-\lambda P_i)}{1-\exp(-\lambda P_i)}(\lambda P_i)^{x_i}}_{ZT-Poisson-likelihood} \times \underbrace{\pi(\lambda)}_{prior}$$

- or

$$\pi(\lambda|x_1,\cdots,x_n) = \frac{\prod_i \frac{(\lambda P_i)^{x_i}}{\exp(-\lambda P_i)-1} \times \pi(\lambda)}{\int_\lambda \prod_i \frac{(\lambda P_i)^{x_i}}{\exp(-\lambda P_i)-1} \times \pi(\lambda)\ d\lambda}$$

priors

- non-informative $\pi(\lambda) = 1$
- 95% CI: $23.14 - 43.20$ per $100,000$ person years
- posterior median 31.75 per $100,000$ person years
- more interesting are the population sizes
- 95% CI: $103 - 178$ with posterior median of 134 studies

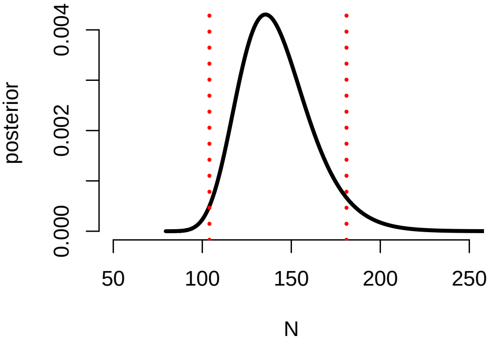


priors

- non-informative but proper $\log \lambda \sim N(0, 1000^2)$
- 95% CI: $23.47 - 43.17$ per $100,000$ person years
- posterior median 31.66 per $100,000$ person years
- more interesting the population sizes
- 95% CI: $103 - 175$ with posterior median of 134 studies
-
- for comparison with $\pi(\lambda) = 1$:
- 95% CI: $103 - \mathbf{178}$ with posterior median of 134 studies

priors

- Jeffreys invariance prior $\pi(\lambda) \propto \sqrt{\text{Fisher information}} = \sqrt{(\sum_i P_i)/\lambda}$
- 95% CI: $104 - 181$ with posterior median of 133 studies
- for comparison with $\pi(\lambda) = 1$:
- 95% CI: $103 - 178$ with posterior median of 134 studies



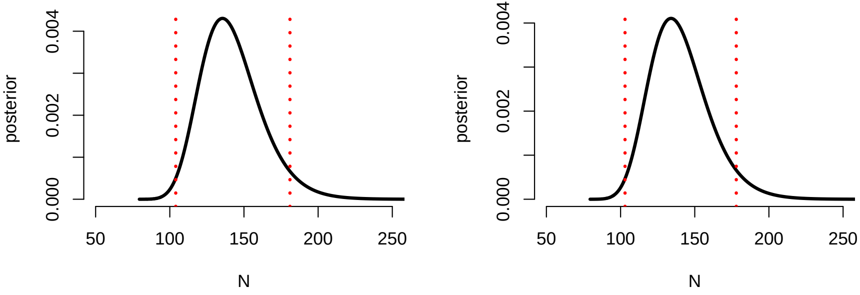


Figure: *left*: Jeffreys invariance prior *right*: non-informative improper prior

overview

Table: **all methods for estimating the total size of studies in a nutshell**

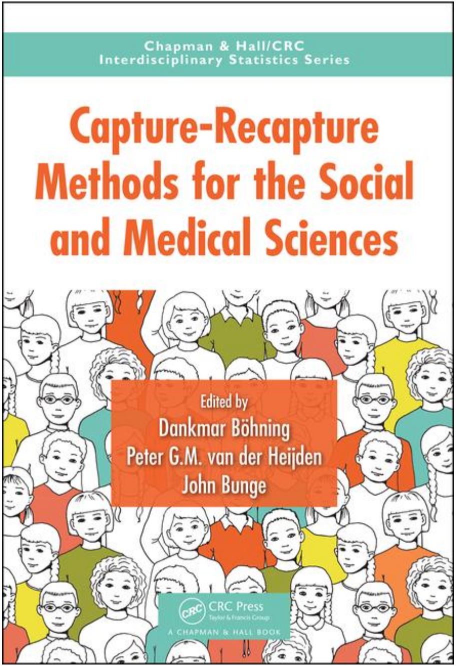
method	median	95% CI
MLE with bootstrap	133	93 – 167
Bayes prior:		
improper non-informative	134	103 – 178
log-normal	134	103 – 175
Jeffreys	133	104 – 181

Table: **a final point: model (likelihood) assessment is essential**

Distribution	LP	BIC	pop size
Poisson	5	58.6	125
	4	55.9	119
	3	52.6	118
	2	53.4	134
	1	50.7	134
	0	139.9	**31**

Table: recall: linear predictors considered

Linear predictor	Proportion of women	Country of origin	Interaction	log-person-time as offset
0	No	No	No	No
1	No	No	No	Yes
2	Yes	No	No	Yes
3	No	Yes	No	Yes
4	Yes	Yes	No	Yes
5	Yes	Yes	Yes	Yes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