

## PREVALENCE AND TREND ESTIMATION FROM OBSERVATIONAL DATA WITH HIGHLY VARIABLE POST-STRATIFICATION WEIGHTS<sup>1</sup>

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In observational surveys, post-stratification is used to reduce bias resulting from differences between the survey population and the population under investigation. However, this can lead to inflated post-stratification weights and, therefore, appropriate methods are required to obtain less variable estimates. Proposed methods include collapsing post-strata, trimming post-stratification weights, generalized regression estimators (GREG) and weight smoothing models, the latter defined by random-effects models that induce shrinkage across post-stratum means. Here, we first describe the weight-smoothing model for prevalence estimation from binary survey outcomes in observational surveys. Second, we propose an extension of this method for trend estimation. And, third, a method is provided such that the GREG can be used for prevalence and trend estimation for observational surveys. Variance estimates of all methods are described. A simulation study is performed to compare the proposed methods with other established methods. The performance of the nonparametric GREG is consistent over all simulation conditions and therefore serves as a valuable solution for prevalence and trend estimation from observational surveys. The method is applied to the estimation of the prevalence and incidence trend of influenza-like illness using the 2010/2011 Great Influenza Survey in Flanders, Belgium.

**1. Introduction.** Frequently, researchers are interested in estimating the overall mean or time trend of an outcome in the study population based on a survey. To obtain valid estimates, the sample must be representative for the population. Stratified sampling provides samples that are representative for the population and can reduce the variance of the estimators. In observational surveys, though, it is not possible to perform stratified sampling. However, when auxiliary information is available, post-stratification can be used to correct for known differences between

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the obtained sample and the population under investigation. This is done by equating the sample distribution of a secondary variable with its distribution in the population, and adjusting estimates using appropriate weighting techniques. When the secondary variable is related with the survey outcome variable, post-stratification can improve both the accuracy and precision of estimators [Little (1993)]. In this article we restrict ourselves to the setting where a binary survey outcome is of interest and an ordinal (or interval-scaled) post-stratifying variable is available.

Post-stratification weights play important, but different roles in design-based and model-based inference. In design-based inference, it is assumed that the sample inclusion indicator is random and the survey variables are fixed. Popular design-based estimators are the Horvitz–Thompson (HT) estimator and its extensions which weigh observations by the inverse of their probability of inclusion [Horvitz and Thompson (1952)]. Although these estimators are design consistent [Isaki and Fuller (1982)], they can be very inefficient when highly variable post-stratification weights are present, as illustrated by Basu’s (1971) famous elephant example.

In contrast, in model-based inference, distributional assumptions are made about the survey outcome and a model is used to predict the outcome for the nonsampled units [Little (2004)]. Such model-based prediction estimators are consistent, but are subject to bias when the underlying model is misspecified. Weight smoothing models treat the strata of the post-stratifying variable as random effects in the model. In this manner, information between strata is borrowed through shrinkage related to the sample size in each stratum and, consequently, post-stratification weights are implicitly smoothed [Elliott and Little (2000)]. In the literature, these models are extensively described for Gaussian data within the empirical Bayesian or full Bayesian framework [Elliott and Little (2000), Gelman (2007), Ghosh and Meeden (1986), Lazzeroni and Little (1998), Little (1983, 1991, 1993), Zheng and Little (2004)]. In the full Bayesian framework, generalized linear regression estimators for non-Gaussian data have been discussed [Elliott (2007)].

Weight smoothing models are similar to model-based prediction models widely used in the field of small area estimation (SAE) [Rao (2003)]. In SAE, generalized linear mixed models are used for the prediction of small area proportions, with random effects introduced to capture area specific differences. Because of the similarity between these models and weight smoothing models, several results from SAE are also applicable in the context of weight smoothing models.

Lehtonen and Veijanen (1998) proposed the generalized regression estimator (GREG), which is a design-based model-assisted estimator where predictions based on a suitable model and the HT estimator for the model residuals are combined. This GREG estimator was extended by Chen, Elliott and Little (2010) who introduce a Bayesian penalized spline predictive estimator for probability-proportional-to-size sampling. Their predictive model includes the weights as covariates.

In this paper, focus is on estimating the prevalence of a survey outcome when one or more post-stratification weights are large. A weight smoothing model based on a generalized linear mixed model is used for this purpose. This weight smoothing model for binary data is a natural extension of the weight smoothing model for continuous data. The predicted values from the model are used in the GREG estimator. The two major additions of this article to the existing literature are as follows: (i) the adjustment of the weights used in the GREG estimator such that the GREG estimator can be used for post-stratification weights obtained from an observational survey; and (ii) a weight smoothing model for estimating a time trend of a binary survey outcome.

The outline of this paper is as follows. Section 2 describes a motivating data example that concerns the estimation of the overall prevalence and weekly incidence trend of influenza-like illness from the Great Influenza Survey (GIS) in Flanders, Belgium. Notation and conventional method of analysis are given in Section 3. Section 4 describes our modeling approaches, and in Section 5 these methods are compared with standard alternatives in an elaborate simulation study. We return to the GIS data example in Section 6, and conclude the paper with a discussion in Section 7.

**2. Motivating example.** The Great Influenza Survey (GIS) is an observational survey based on the voluntary participation of individuals via the internet aiming at monitoring influenza-like illness (ILI) in the Netherlands and Belgium [Friesema et al. (2009), Marquet et al. (2006), Vandendijck, Faes and Hens (2013)]. Participants of the GIS receive a weekly email with a link to a questionnaire (it is not mandatory to complete this questionnaire). Based on the questionnaire, well-defined criteria are used to determine whether or not the participant has experienced an ILI episode in the preceding week. At the start of the flu-season (or at registration), an additional questionnaire needs to be completed to obtain information on several demographic characteristics. In this paper, we focus on the GIS in Flanders, Belgium, during the 2010–2011 influenza season. Interest is in the estimation of the overall mean prevalence and the weekly incidence trend of ILI. Data from the Flemish GIS can be obtained upon request via [www.influenzanel.com](http://www.influenzanel.com).

The GIS recorded information of 27 weeks during the 2010–2011 influenza season (from 1/11/2010 up to 8/5/2011). In total, there were 4551 participants, with a total of 83,500 records. The highest number of respondents in a week was 3467 and the minimum was 2402.

The age of the participants ranged from 1 to 88 years. From Figure 1 it is observed that the age distribution of the survey population is very dissimilar to the age distribution of the overall Flemish population and, therefore, we post-stratify the survey according to 18 five-year age intervals. The population age distribution of the overall Flemish population is obtained from Statistics Belgium ([www.statbel.fgov.be](http://www.statbel.fgov.be)). Some post-strata are seriously underrepresented and therefore high post-stratification weights are present, with weights ranging from 0.46 up to 35.70 (in Section 3 a definition of post-stratification weights is presented).

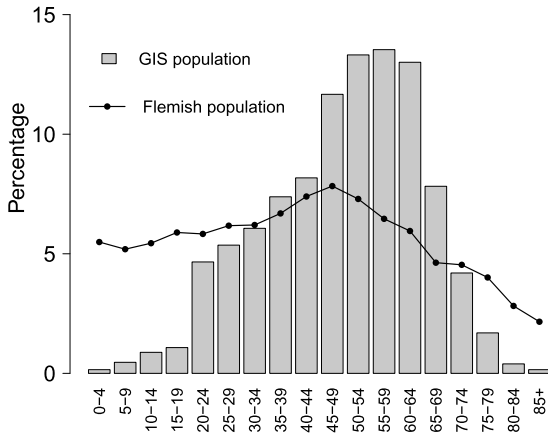


FIG. 1. Age distribution of the Great Influenza Survey (GIS) population and the overall Flemish population stratified by 18 age intervals of five years.

In Section 6, results for this survey are given to illustrate the methods described in this paper. In this analysis, we will assume no response bias is present. Because this data example is only used for illustration purposes, we do not correct for gender post-stratification, though the method allows easy extension to take this into account.

### 3. Notation and the conventional methods of analysis.

3.1. *Notation.* Let  $Y$  denote a binary survey outcome variable and  $X$  an ordinal post-stratifying variable with  $H$  levels and known population distribution. Let  $N_h$  denote the population size and  $n_h$  the sample size in post-stratum  $h$ , for  $h = 1, \dots, H$ . We assume that  $N_h$  is obtained from auxiliary data and  $n_h > 0$  ( $\forall h = 1, \dots, H$ ). The total population and sample size are denoted by  $N = \sum_{h=1}^H N_h$  and  $n = \sum_{h=1}^H n_h$ , respectively. It is assumed that the respondents in a post-stratum can be treated as a random sample. Interest is in the estimation of (or inference about) the population prevalence, namely,  $\bar{Y} = \frac{1}{N} \sum_{i=1}^N Y_i = \sum_{h=1}^H P_h \bar{Y}_h$ , where  $\bar{Y}_h$  is the population mean and  $P_h = N_h/N$  is the population proportion of post-stratum  $h$ .

3.2. *Design-based methods.* Standard design-based approaches to estimate  $\bar{Y}$  consider estimates of the form  $\bar{y} = \frac{1}{n} \sum_{i=1}^n w_{i(h)} y_i$ , where  $w_{i(h)}$  is the weight of observation  $i$  belonging to post-stratum  $h$  with  $\sum_{i=1}^n w_{i(h)} = n$ , and  $y_i$  is the corresponding observed survey outcome. Some special cases are discussed.

(1) The *unweighted sample prevalence*,  $\bar{y}_{unw}$ , is obtained when  $w_{i(h)} = 1$  ( $\forall i = 1, \dots, n$ ). It can be written as  $\bar{y}_{unw} = \sum_{h=1}^H p_h \bar{y}_h$ , with  $p_h = n_h/n$  the sample proportion and  $\bar{y}_h$  the sample prevalence in post-stratum  $h$ . It is an unbiased estimate whenever  $Y$  and  $X$  are unrelated or as long as the probability of inclusion

does not depend on  $X$ . However, if sampling variability is apparent or systematic bias in the sampling procedure is present,  $p_h$  deviates from its population counterpart  $P_h$  and the unweighted sample prevalence is a biased estimator for  $\bar{Y}$ .

(2) The *post-stratified prevalence*,  $\bar{y}_{ps}$ , is obtained when  $w_{i(h)} = P_h/p_h$  ( $\forall i = 1, \dots, n$ ) and can be written as  $\bar{y}_{ps} = \sum_{h=1}^H P_h \bar{y}_h$ . The  $w_{i(h)}$  are called post-stratification weights in this case. This is an unbiased estimate of  $\bar{Y}$ . However, the post-stratified prevalence can be unstable because the sample prevalence in post-stratum  $h$  is used in  $\bar{y}_{ps}$  regardless of the number of respondents in post-stratum  $h$ . Second, when a post-stratum contains only few observations, the variance is inflated. This inflated variance can overwhelm the bias reduction, so that the post-stratified prevalence yields an increased mean-squared error.

(3) In order to solve the instability and inflated variance of  $\bar{y}_{ps}$ , a *weight-trimming prevalence* estimator,  $\bar{y}_{trim}$ , obtained by trimming all weights larger than a prespecified cutoff value  $w_0$ , can be used. The other weights are adjusted to maintain the same weighted sample size. The trimmed prevalence estimate can be written as  $\bar{y}_{trim} = \sum_{h=1}^{l-1} \gamma \frac{N_h}{N} \bar{y}_h + \sum_{h=l}^H w_0 \frac{n_h}{n} \bar{y}_h$ , with normalizing constant  $\gamma = \frac{n - \sum_i \zeta_i w_0}{\sum_i (1 - \zeta_i) w_{i(h)}}$ , where  $\zeta_i$  equals 1 when  $w_{i(h)} > w_0$  and 0 otherwise. The choice of the cutoff value  $w_0$  is mostly done *ad hoc*, although Potter (1990) presented several systematic methods for choosing  $w_0$  based on the data.

Other methods to solve the instability and inflated variance of  $\bar{y}_{ps}$  include pooling or collapsing small post-strata that contribute excessively to the variance [Little (1993), Tremblay (1986)]. Weight trimming can be seen as a special case of collapsing several post-strata. Elliott and Little (2000) proposed an extension of the method, the compound weight pooling method, by conducting the pooling at every possible level of  $w_0$  and computing a Bayesian weighted average.

## 4. Methodology.

4.1. *Model-based inference for prevalence estimation.* Design-based methods ignore the ordinal nature of the post-stratifying variable, whereas a model-based approach reflects the intrinsic order and allows to borrow strength from neighboring strata with more information. Weight smoothing models directly model the means in the weight strata making use of random effects [Lazzeroni and Little (1998)]. The weight smoothing model for a binary survey outcome is

$$(1) \quad y_{i(h)} | \mu_h \sim \text{Bern}(\mu_h) \quad \text{and} \quad \delta^* \sim \mathcal{N}_H(\delta, \mathbf{D}),$$

where  $g(\mu_h) = \delta_h^*$ ,  $\delta^* = (\delta_1^*, \dots, \delta_H^*)^T$  and  $\delta = (\delta_1, \dots, \delta_H)^T$  are vectors of unknown parameters,  $\mathbf{D}$  is an unknown  $H \times H$  variance-covariance matrix and  $g(\cdot)$  is the logit-link function. The following choices for  $\delta$  and  $\mathbf{D}$  have been considered in model (1):

(a) *Exchangeable random effects (XRE)*:  $\delta_h = \beta$  for all  $h$  and  $\mathbf{D} = \sigma^2 \mathbf{I}_H$  [Ghosh and Meeden (1986), Holt and Smith (1979), Little (1983, 1991)].

(b) *Linear model (LIN)*:  $\delta_h = \beta_0 + \beta_1 X_h$  for all  $h$  and  $\mathbf{D} = \sigma^2 \mathbf{I}_H$  [Lazzeroni and Little (1998)].

(c) *Nonparametric (NPAR)*  $\delta_h = f(X_h)$  for all  $h$  and  $\mathbf{D} = \sigma^2 \mathbf{I}_H$ , where  $f$  is a nonparametric spline function [Elliott and Little (2000), Zheng and Little (2004)]. We use the approximating thin plate spline family for constructing  $f$ . This choice was made because this spline family is readily available in the procedure *GLIMMIX* in the software package SAS. Simulations indicate that the approximating thin plate splines perform as well as linear truncated splines or cubic B-splines (see Supplementary Material [Vandendijck, Faes and Hens (2015)]).

All the above models can be cast in the generalized linear mixed model (GLMM) framework:  $g(E(\mathbf{y}|\mathbf{b})) = \boldsymbol{\eta} \equiv \mathbf{N}\mathbf{X}\boldsymbol{\beta} + \mathbf{N}\mathbf{Z}\mathbf{b}$ , where  $\mathbf{N}$  is an  $n \times H$  matrix indicating to which stratum an observation belongs [ $(\mathbf{N})_{ih} = 1$  if  $y_i$  belongs to stratum  $h$  and 0 otherwise],  $\mathbf{X}$  is an  $H \times p$  fixed-effect design matrix and  $\boldsymbol{\beta}$  is a  $p \times 1$  vector of unknown fixed-effect parameters,  $\mathbf{Z}$  is an  $H \times q$  random-effect design matrix and  $\mathbf{b}$  is a  $q \times 1$  vector of unknown random effects for which it is assumed that  $\mathbf{b} \sim \mathcal{N}_q(\mathbf{0}, \mathbf{G})$ . In Appendix A of the Supplementary Material [Vandendijck, Faes and Hens (2015)], it is shown how models (a)–(c) are formulated in the GLMM framework. Once the model has been cast in the GLMM framework, the model is fit by pseudo-restricted maximum likelihood estimation based on linearization [Wolfinger and O’Connell (1993)]. This method is doubly iterative and employs Taylor series expansions to approximate the model by a linear mixed model using pseudo-data. The details of the estimation method are given in Appendix B of the Supplementary Material [Vandendijck, Faes and Hens (2015)]. In practice, the weight smoothing models (a)–(c) can be implemented in, for example, the procedure *GLIMMIX* in the software package SAS. See Appendix C of the Supplementary Material for example code [Vandendijck, Faes and Hens (2015)].

A predictive model-based approach, by predicting the outcome of the nonsampled individuals based on model (1), by Royall (1970) is used to specify an estimator of  $\bar{Y}$ . The estimator is

$$(2) \quad \bar{y}_{ws} = E(\bar{Y}|\mathbf{y}) = \frac{1}{N} \sum_{h=1}^H \{n_h \bar{y}_h + (N_h - n_h) \hat{\mu}_h\},$$

where  $\hat{\mu}_h = E(\bar{Y}_h|\mathbf{y}) = g^{-1}(\delta_h^*)$ . The unweighted and post-stratified prevalences are special cases of (2), and are obtained if  $\mathbf{D} \rightarrow 0$  and  $\mathbf{D} \rightarrow \infty$  in (1), respectively.

Note that in (1) and (2) there is no indication that the post-stratification weights are actually smoothed. However, the post-stratification weights are indirectly smoothed by the shrinkage associated with model (1). For Gaussian data, explicit analytical formulas for the smoothed weights are available [Lazzeroni and Little (1998)]. For binary data, on the contrary, these formulas are not available due to the lack of the hat matrix from model (1).

Essentially, estimator (2) estimates the finite population mean by using a GLMM to predict the unobserved values. These estimators are also in common use for prediction of small area proportions in SAE [see, e.g., Farrell (2000)].

With regard to a variance estimator of (2), both an analytical formula and a re-sampling method are presented. The analytical approach is attractive because it only involves matrix calculations, but it is only an approximation. As an alternative, a more computer intensive bootstrap approach is proposed.

*Analytical approach.* Because the weight smoothing model (1) can be cast into a GLMM, the estimation of the variance can be derived within this framework. To derive an approximate expression for the variance of  $\bar{y}_{ws}$ , a first order Taylor series expansion of (2) is taken with respect to the fixed and random effects. This leads to

$$(3) \quad \text{Var}(\bar{y}_{ws}) \approx \frac{1}{N^2} (\mathbf{N} - \mathbf{n})^T \boldsymbol{\Theta} (\mathbf{N} - \mathbf{n}),$$

with  $(\mathbf{N} - \mathbf{n}) = (N_1 - n_1, \dots, N_H - n_H)^T$  and  $\boldsymbol{\Theta} = \boldsymbol{\Delta}_* \mathbf{C} (\mathbf{C}^T \boldsymbol{\Sigma}_p^{-1} \mathbf{C} + \mathbf{B})^{-1} \mathbf{C}^T \boldsymbol{\Delta}_*$ . Definitions of the matrices  $\boldsymbol{\Delta}_*$ ,  $\mathbf{C}$ ,  $\boldsymbol{\Sigma}_p$  and  $\mathbf{B}$  are given in Appendix B of the Supplementary Material [Vandendijck, Faes and Hens (2015)]. Finally, the variance is estimated by replacing  $\boldsymbol{\Delta}_*$ ,  $\boldsymbol{\Sigma}_p$  and  $\mathbf{B}$  by their estimates  $\hat{\boldsymbol{\Delta}}_*$ ,  $\hat{\boldsymbol{\Sigma}}_p$  and  $\hat{\mathbf{B}}$ . Similar variance estimates as in (3) are also obtained in model-based approaches in SAE [Farrell (2000)]. Note that the variance estimator (3) does not include a term that accounts for the uncertainty of the estimated variance components of model (1). Hence, confidence intervals based on (3) are often too narrow to achieve the desired level of coverage. A number of analytical methods for addressing this shortcoming are available [see, e.g., Prasad and Rao (1990)]. We, however, opt to use a parametric bootstrap approach because it is a conceptually simple alternative.

*Bootstrap approach.* The described bootstrap approach was proposed in SAE [González-Manteiga et al. (2007), Laird and Louis (1987)] and is easily adjusted to the case of weight smoothing models. The approach consists of two procedures.

#### PROCEDURE I.

(I.1) From the original survey, obtain estimates  $\hat{\boldsymbol{\beta}}$  and  $\hat{\mathbf{D}}$  of  $\boldsymbol{\beta}$  and  $\mathbf{D}$ , respectively. For the NPAR model, also obtain the empirical Bayes prediction  $\hat{\mathbf{b}}_u$  of the spline coefficients  $\mathbf{b}_u$ .

(I.2) Generate a random vector  $\tilde{\mathbf{u}}$  from  $\tilde{\mathbf{u}} \sim \mathcal{N}(\mathbf{0}, \hat{\mathbf{D}})$ .

(I.3) Generate a population  $\tilde{P}$  of size  $N$  by generating values from a binomial distribution with sizes  $N_h$  and probabilities  $\tilde{p}_h$ , for  $h = 1, \dots, H$ , according to the bootstrap superpopulation model  $\tilde{p}_h = \text{expit}(\tilde{\delta}_h^*)$ , where  $\tilde{\boldsymbol{\delta}}^* = (\tilde{\delta}_1^*, \dots, \tilde{\delta}_H^*)^T = \mathbf{X}\hat{\boldsymbol{\beta}} + \mathbf{Z}\tilde{\mathbf{u}}$  for the XRE and LIN model, and  $\tilde{\boldsymbol{\delta}}^* = (\tilde{\delta}_1^*, \dots, \tilde{\delta}_H^*)^T = \mathbf{X}\hat{\boldsymbol{\beta}} + \mathbf{Z}_1\hat{\mathbf{b}}_u + \mathbf{Z}_2\tilde{\mathbf{u}}$  for the NPAR model (see Appendix B of the Supplementary Material [Vandendijck, Faes and Hens (2015)]).



PROCEDURE II.

(II.1) Use Procedure I to generate  $B$  independent bootstrap populations  $\tilde{P}^{(b)}$  of size  $N$ , and calculate the bootstrap population mean  $\tilde{y}^{(b)}$ .

(II.2) Extract a sample  $\tilde{s}^{(b)}$  of size  $n$  from each  $\tilde{P}^{(b)}$ , taking into account the sample sizes in the post-strata. Fit model (1) to the sample data  $\tilde{s}^{(b)}$  and calculate the bootstrap predictor  $\hat{y}^{(b)}$  using (2).

(II.3) The bootstrap variance of  $\bar{y}_{ws}$  is  $\frac{1}{B} \sum_{b=1}^B (\hat{y}^{(b)} - \tilde{y}^{(b)})^2$ .

From our simulation results, we recommend to take  $B$  at least larger than 100. The computing time necessary for this bootstrap method largely depends on the computing time of the model to be estimated in step (II.2). For the prevalence example shown in Appendix C of the Supplementary Material [Vandendijck, Faes and Hens (2015)], we performed a bootstrap variance calculation with  $B = 250$ . For this example, step (II.2) took about 10 seconds in total for all bootstrap samples on a laptop personal computer with Intel Core i5-2540M processor.

4.2. *Model-assisted design-based inference.* The GREG estimator for binary outcomes, proposed by Lehtonen and Veijanen (1998), is a popular model-assisted design-based estimator that combines predicted values  $\hat{y}_i$  based on a suitable model and design-weighted residuals  $r_i = y_i - \hat{y}_i$  of the sampled units. The estimator is given by

$$(4) \quad \bar{y}_{GREG} = \frac{1}{N} \left( \sum_{i=1}^N \hat{y}_i + \sum_{i \in s} \frac{r_i}{\pi_i} \right),$$

where  $\pi_i$  is the probability of inclusion in the sample for unit  $i$ . The second term on the right-hand side is a bias correction factor that eliminates possible bias due to model misspecification. Lehtonen and Veijanen (1998) used a design-weighted logistic regression model on other covariates as the assisting model to predict  $\hat{y}_i$ . Based on estimator (4), we propose a model-assisted design-based estimator  $\bar{y}_{ws,GREG}$  that uses a weight smoothing model as assisting model and that can be used for observational surveys.

Define  $r_i = y_i - \hat{\mu}_h$ , where  $\hat{\mu}_h$  is the predicted value from (1) of the post-stratum  $h$  to which unit  $i$  belongs. The estimator  $\bar{y}_{ws,GREG}$  is

$$(5) \quad \bar{y}_{ws,GREG} = \frac{1}{N} \left( \sum_{h=1}^H \sum_{i=1}^{N_h} \hat{\mu}_h + \sum_{i \in s} \frac{r_i}{\tilde{\pi}_i} \right),$$

where  $\tilde{\pi}_i$  will be defined later. Assuming that the value of  $\tilde{\pi}_i$  is the same for all respondents in stratum  $h$ , the estimator can be written as

$$(6) \quad \bar{y}_{ws,GREG} = \frac{1}{N} \sum_{h=1}^H \left\{ \frac{n_h}{\tilde{\pi}_h} \bar{y}_h + \left( N_h - \frac{n_h}{\tilde{\pi}_h} \right) \hat{\mu}_h \right\}.$$



Because we are dealing with self-selected samples in observational surveys, no inclusion probabilities  $\pi_i$  are available. Additionally, setting  $\tilde{\pi}_h = n_h/N_h$  would just return the post-stratified mean. We propose to set the values of  $\tilde{\pi}_h$  using the idea of weight trimming, namely,

$$\tilde{\pi}_h = \frac{\hat{n}_h}{N_h} \quad \text{where } \hat{n}_h = \begin{cases} N_h/N, & \text{if } w_h > w_0, \\ w_0/n, & \text{if } w_h \leq w_0 \end{cases} \quad \text{and}$$

$$\gamma = \frac{n - \sum_{h:w_h > w_0} \hat{n}_h}{\sum_{h:w_h \leq w_0} n_h}.$$

Note that for the calculation of  $\gamma$ , the values of  $\hat{n}_h$  for those strata where  $w_h > w_0$  need to be calculated first. In summary, our proposed estimator  $\bar{y}_{ws,GREG}$  combines predicted values based on a weight smoothing model and a weighted sum of residuals of the sampled units where the weights  $\tilde{\pi}_h^{-1}$  resemble trimmed weights.

For the variance estimator of  $\bar{y}_{ws,GREG}$ , we use a jackknife procedure described in Zheng and Little (2005) and Chen, Elliott and Little (2010). A jackknife procedure is used because it resembles the concept of design-based inference, in which inference is based on the sampling distribution. The jackknife approach is given in Procedure III.

### PROCEDURE III.

(III.1) Sort the original sample  $s$  according to the post-stratifying variable  $X$ . Stratify the sample into  $n/G$  strata, each of size  $G$  with similar values of the post-stratifying variable  $X$ . Construct  $G$  subgroups by selecting one element at a time from each stratum without replacement.

(III.2) For each  $g = 1, \dots, G$ , construct the reduced sample  $s^{(g)}$  from  $s$  without the elements in the  $g$ th subgroup. Fit model (1) to  $s^{(g)}$  and calculate  $\bar{y}_{ws,GREG}^{(g)}$  using (6). Note that in this step the weights  $\tilde{\pi}_h^{-1}$  calculated on the original sample  $s$  are still used.

(III.3) Define  $\bar{y}_{ws,GREG}^* = G^{-1} \sum_{g=1}^G \bar{y}_{ws,GREG}^{(g)}$ . The jackknife variance estimator of  $\bar{y}_{ws,GREG}$  is

$$\frac{G-1}{G} \sum_{g=1}^G (\bar{y}_{ws,GREG}^{(g)} - \bar{y}_{ws,GREG}^*)^2.$$

4.3. *Extension toward trend estimation.* So far, focus was on estimating the overall prevalence. Here, a method is proposed to estimate the overall time trend of a survey outcome measured at different time points  $t = 1, \dots, T$ . Interest is in estimating  $\bar{Y}_t = \frac{1}{N_t} \sum_{i=1}^{N_t} Y_{it}$ , for  $t = 1, \dots, T$ , from a sample which is available at each time point  $t$ . The sample at time  $t$  can be used to estimate  $\bar{Y}_t$  via the

unweighted, post-stratified, trimmed or weight smoothed mean. However, we propose to extend model (1) to a weight smoothing model that exploits the time trend. The general form of the weight smoothing model with smooth time trend is

$$(7) \quad y_{i(h),t} | \mu_{h,t} \sim \text{Bern}(\mu_{h,t}) \quad \text{and} \quad \boldsymbol{\delta}^* \sim \mathcal{N}_H(\boldsymbol{\delta}, \mathbf{D}),$$

where  $g(\mu_{h,t}) = \delta_t + \delta_h^*$ ,  $\boldsymbol{\delta}^* = (\delta_1^*, \dots, \delta_H^*)^T$  and  $\boldsymbol{\delta} = (\delta_1, \dots, \delta_H)^T$  as before. For  $\boldsymbol{\delta}$  and  $\mathbf{D}$  we can again assume a XRE, LIN or NPAR model as in Section 4.1. The parameter  $\delta_t$  corresponds to the time trend and is modeled by a nonparametric function,  $f_t(\cdot)$ , which is specified by the approximating thin plate spline family. Model (7) can again be cast in the GLMM framework. After model fitting, the fitted post-strata means,  $\hat{\boldsymbol{\mu}}_t = (\hat{\mu}_{1,t}, \dots, \hat{\mu}_{H,t})^T$ , are obtained at each time point  $t$ , and an estimator of  $\bar{Y}_t$  is

$$(8) \quad \bar{y}_{ws,t} = \frac{1}{N_t} \sum_{h=1}^H \{n_{h,t} \bar{y}_{h,t} + (N_{h,t} - n_{h,t}) \hat{\mu}_{h,t}\}.$$

A GREG adjusted estimator,  $\bar{y}_{ws,t,GREG}$ , is constructed in the same manner as in Section 4.2,

$$(9) \quad \bar{y}_{ws,t,GREG} = \frac{1}{N_t} \sum_{h=1}^H \left\{ \frac{n_{h,t}}{\tilde{\pi}_{h,t}} \bar{y}_{h,t} + \left( N_{h,t} - \frac{n_{h,t}}{\tilde{\pi}_{h,t}} \right) \hat{\mu}_{h,t} \right\}.$$

Variance estimation of  $\bar{y}_{ws,t}$  can be obtained by an analytical formula or a bootstrap approach. The approximate analytical variance is given by

$$(10) \quad \text{Var}(\bar{y}_{ws,t}) \approx \frac{1}{N_t^2} (\mathbf{N}_t - \mathbf{n}_t)^T \boldsymbol{\Theta}_t (\mathbf{N}_t - \mathbf{n}_t),$$

where  $(\mathbf{N}_t - \mathbf{n}_t) = (N_{1,t} - n_{1,t}, \dots, N_{H,t} - n_{H,t})^T$  and  $\boldsymbol{\Theta}_t$ , for  $t = 1, \dots, T$ , are the subsequent  $H \times H$  block matrices along the main diagonal of the  $HT \times HT$  covariance matrix  $\boldsymbol{\Theta}$  of the fitted post-stratum means, calculated, as before, by a first order Taylor series expansion. For the bootstrap approach, Procedure I and Procedure II proceed analogously with the adjustment that bootstrap data is now sampled from model (7).

For the jackknife variance calculation of  $\bar{y}_{ws,t,GREG}$ , some technical adjustments need to be made to Procedure III. The jackknife procedure is provided in Procedure IV.

PROCEDURE IV.

(IV.1) Sort the original sample  $s$  first according to time and second to the post-stratifying variable  $X$ . At each time point, stratify the sample into  $n_t/G$  strata each of size  $G$  with similar values of the post-stratifying variable  $X$ . Construct  $G$  subgroups by selecting one element at a time from each stratum without replacement.

(IV.2) For each  $g = 1, \dots, G$ , construct the reduced sample  $s^{(g)}$  from  $s$  where at each time point the elements in the  $g$ th subgroup are removed. Fit model (7) to  $s^{(g)}$  and calculate  $\bar{y}_{ws,t,GREG}^{(g)}$  using (9).

(IV.3) Similar as in (III.3).

**5. Simulation study.**

5.1. *Simulation settings for overall prevalence estimation.* In total,  $8 \times 2 \times 2 = 32$  simulation conditions are evaluated for the estimation of the overall prevalence. This is done by crossing eight population types with two population sizes and two sample sizes for each population size. For the first population a total size  $N^{(1)} = 6,000,000$  is considered and for the second size  $N^{(2)} = 150,000$ . It is assumed that both populations consist of 18 strata with strata population sizes given in Table 1.

The population values are generated as in model (1) with the following choices:

- (1) NULL:  $\delta_h = 0 (\forall h = 1, \dots, 18)$  and  $\mathbf{D} = \mathbf{0}$ .
- (2) XRE:  $\delta_h = 0 (\forall h = 1, \dots, 18)$  and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .
- (3) LIN<sub>0</sub>:  $\delta_h = -2 + 0.2h (\forall h = 1, \dots, 18)$  and  $\mathbf{D} = \mathbf{0}$ .
- (4) LIN<sub>1</sub>:  $\delta_h = -2 + 0.2h (\forall h = 1, \dots, 18)$  and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .

TABLE 1  
Population and sample sizes in the 18 strata for the large population ( $N^{(1)} = 6,000,000$ ) and small population ( $N^{(2)} = 150,000$ ) used in the simulation study.

Stratum $h$	1	2	3	4	5	6	7	8	9
$N_h^{(1)}$	300,000	300,000	320,000	320,000	340,000	340,000	360,000	360,000	360,000
$n_{1,h}^{(1)}$	50	150	300	750	1250	1500	2000	2750	3750
$n_{2,h}^{(1)}$	10	30	60	150	250	300	400	550	750
$N_h^{(2)}$	7500	7500	8000	8000	8500	8500	9000	9000	9000
$n_{1,h}^{(2)}$	5	15	30	75	125	150	200	275	375
$n_{2,h}^{(2)}$	1	3	6	15	25	30	40	55	75

Stratum $h$	10	11	12	13	14	15	16	17	18
$N_h^{(1)}$	360,000	360,000	360,000	340,000	340,000	320,000	320,000	300,000	300,000
$n_{1,h}^{(1)}$	3750	2750	2000	1500	1000	800	400	200	100
$n_{2,h}^{(1)}$	750	550	400	300	200	160	80	40	20
$N_h^{(2)}$	9000	9000	9000	8500	8500	8000	8000	7500	7500
$n_{1,h}^{(2)}$	375	275	200	150	100	80	40	20	10
$n_{2,h}^{(2)}$	75	55	40	30	20	16	8	4	2

- (5) QUAD<sub>0</sub>:  $\delta_h = 1 - 0.25h + 0.01h^2$  ( $\forall h = 1, \dots, 18$ ) and  $\mathbf{D} = \mathbf{0}$ .  
 (6) QUAD<sub>1</sub>:  $\delta_h = 1 - 0.25h + 0.01h^2$  ( $\forall h = 1, \dots, 18$ ) and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .  
 (7) EXP<sub>0</sub>:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$  ( $\forall h = 1, \dots, 18$ ) and  $\mathbf{D} = \mathbf{0}$ .  
 (8) EXP<sub>1</sub>:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$  ( $\forall h = 1, \dots, 18$ ) and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .

The parameters are chosen such that the overall mean prevalence in the population is about 0.5. For each of the above population models (1)–(8), 25 populations are randomly generated and this for both population sizes  $N^{(1)}$  and  $N^{(2)}$ . In each of these populations, ten samples of fixed sample size are generated. This procedure yields a total of 250 replications for each combination of population size, sample size and population model. For the large population size ( $N^{(1)}$ ) we consider samples of size 25,000 and size 5000. For the small population size ( $N^{(2)}$ ) we consider samples of sizes 2500 and 500, respectively. The sample sizes per stratum are given in Table 1. In all of the settings the normalized post-stratification weights of the strata are similar, ranging from 0.4 to 25.

The following nine estimators for estimating the overall population mean from the generated samples are compared: (*psm*) the post-stratified mean,  $\bar{y}_{ps}$ ; (*unw*) the unweighted mean,  $\bar{y}_{unw}$ ; (*trim*) the trimmed mean,  $\bar{y}_{trim}$ , with a cutoff value of  $w_0 = 3$ ; (*xre*)  $\bar{y}_{ws}$ , using the XRE assumption; (*xre-greg*)  $\bar{y}_{ws,greg}$ , using the XRE assumption; (*lin*)  $\bar{y}_{ws}$ , using the LIN assumption; (*lin-greg*)  $\bar{y}_{ws,greg}$ , using the LIN assumption; (*npar*)  $\bar{y}_{ws}$ , using the NPAR assumption; (*npar-greg*)  $\bar{y}_{ws,greg}$ , using the NPAR assumption. For all GREG estimators we use  $w_0 = 3$ . For the *npar* estimator we place knots at each value of  $h$ , for a total of 18 knots. A sensitivity analysis with ten equally spaced knots revealed no meaningful differences.

For each of these estimators we calculate the average bias, variability, mean squared error (MSE), the coverage and average length of the 95% confidence interval (CI). To calculate the MSE, we first calculate the mean squared error within each of the 25 populations. Denote by  $\theta^{(p)}$  the true population proportion of population  $p$ . For each population, we obtain ten estimates  $\hat{\theta}^{(p)} = (\hat{\theta}_1^{(p)}, \dots, \hat{\theta}_{10}^{(p)})$  of  $\theta^{(p)}$ . The mean squared error in population  $p$  is estimated by  $MSE^{(p)} = \text{Var}(\hat{\theta}^{(p)}) + (\text{Bias}(\theta^{(p)}, \hat{\theta}^{(p)}))^2$ . The overall mean squared error is then calculated by averaging over the 25  $MSE^{(p)}$  values. For the bootstrap and jackknife variance procedure we used  $B = 250$  and  $G = 250$ , respectively. The CIs are first calculated on the linear scale. Next, these CIs are back-transformed to yield CIs between 0 and 1. A normal reference distribution is used for the CIs. For *psm*, *unw* and *trim* we use the variance formulas as given in Table 1 in Little (1991).

5.2. *Results for overall prevalence estimation.* Only part of the simulation results are presented here. For more results we refer to Appendix D of the Supplementary Material [Vandendijck, Faes and Hens (2015)]. Table 2 summarizes the MSE and Figure 2 shows the bias for  $N^{(1)}$  with a sample size of 5000. When the

TABLE 2  
*Mean squared error ( $\times 10^4$ ) of nine estimators and eight population models for the large population  $N^{(1)} = 6,000,000$  with a sample size of 5000. †Based on 249 simulated data sets*

Estimator	NULL	XRE	LIN <sub>0</sub> <sup>†</sup>	LIN <sub>1</sub>	QUAD <sub>0</sub>	QUAD <sub>1</sub>	EXP <sub>0</sub>	EXP <sub>1</sub>
<i>psm</i>	1.70	2.85	1.16	1.73	1.88	2.92	1.82	2.90
<i>unw</i>	0.46	1.83	0.43	1.54	16.09	17.52	4.16	5.42
<i>trim</i>	0.68	1.92	0.86	1.68	5.61	6.50	2.41	3.26
<i>xre</i>	0.47	0.61	1.01	1.04	4.64	4.11	1.83	1.75
<i>xre-greg</i>	0.67	0.84	1.06	1.09	3.14	2.90	1.73	1.68
<i>lin</i>	0.48	0.61	0.30	0.35	8.35	5.59	2.97	2.04
<i>lin-greg</i>	0.67	0.83	0.50	0.53	3.66	2.96	1.49	1.32
<i>npar</i>	0.65	0.90	0.48	0.57	1.63	1.71	1.74	1.49
<i>npar-greg</i>	0.85	1.08	0.66	0.71	1.52	1.57	1.49	1.40

underlying population has a constant mean (NULL and XRE), all estimators yield unbiased estimates. For the QUAD and EXP populations, the GREG adjusted estimators show less bias than their nonadjusted counterparts. The *psm* estimator

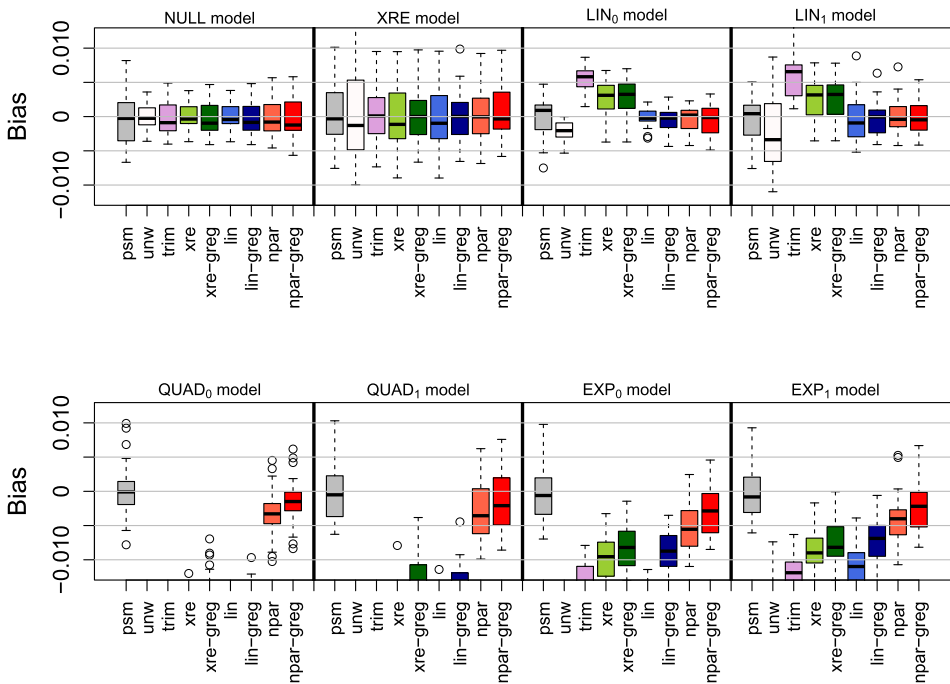


FIG. 2. *Bias results of nine estimators and eight population models for the large population  $N^{(1)} = 6,000,000$  with a sample size of 5000. Boxplots of the average bias associated with the 25 simulated populations are shown. The bias of some estimators exceeds the range of the y-axis used and are therefore not depicted on the figure.*

remains unbiased in all simulation settings. In general, *psm* does not perform well in terms of MSE, due to the overwhelming increase in variability over the other estimators (see Appendix D of the Supplementary Material [Vandendijck, Faes and Hens (2015)]). For a population with more structure (LIN, QUAD and EXP), the *unw* and *trim* estimators show a high MSE, due to the bias of the estimates (see Figure 2). For the LIN populations, as expected, the estimators *lin* and *lin-greg* performed the best in terms of MSE. The *npar* estimator is second best with a slight increase in MSE. The *npar* estimators perform the best for the QUAD populations. For the EXP populations, the *lin-greg* and both *npar* estimators perform well. The *npar* estimator has a higher MSE than *xre* and *lin* for the NULL and XRE populations. This is due to the higher variance associated with the *npar* estimator. For the QUAD and EXP underlying populations, it is observed that the estimators *xre-greg*, *lin-greg* and *npar-greg* have lower MSE when compared with the nonadjusted estimators. In the QUAD and EXP scenarios, the *npar-greg* yields smaller MSE values than the *npar*, which could suggest that the NPAR weight smoothing model does not fit the data generated by the QUAD and EXP models well. Investigating the model fits reveals no problems with the fits of the NPAR weight smoothing model (see Appendix E of the Supplementary Material [Vandendijck, Faes and Hens (2015)]). The bias reduction of the *npar-greg* achieved by the bias reduction term is greater than the increase in variability of this estimator, which leads to smaller MSE values for the *npar-greg* in the QUAD and EXP scenarios. Overall, the *npar* and *npar-greg* estimators perform the most consistent in the simulation study. Results for other population and sample sizes do not differ qualitatively (see Appendix D of the Supplementary Material [Vandendijck, Faes and Hens (2015)]).

Table 3 shows the nominal coverage and average length of the 95% CI for  $N^{(1)}$  with a sample size of 5000. The *psm* estimator attains a good coverage over all simulation conditions. Due to the bias, the nominal coverage of the *unw* and *trim* estimators deteriorates when the underlying model has more structure. For the underlying models with nonlinear mean (QUAD and EXP), the *xre* and *lin* estimators do not obtain good coverage results. Overall, the *npar-greg* estimator yields consistent nominal coverage results over all the simulation conditions. Only for the QUAD and EXP populations, the *npar-greg* underestimates the actual coverage slightly. The average length of the CIs is also smaller for the *npar-greg* method when compared to *psm*.

5.3. *Simulation settings for mean trend estimation.* For trend estimation,  $2 \times 2 \times 4 = 16$  simulation conditions are considered. This is done by crossing four population types with the same population and sample sizes as in Section 5.1. We assume that the survey is available at 30 time points which are equally spaced in time. Further, it is assumed that the post-strata population and sample sizes remain constant over time. Similarly, as in Section 5.1, 25 populations are randomly generated and ten samples are generated in each population. The population values

TABLE 3  
*Nominal coverage of the 95% CI (average length  $\times 100$  of the CI) of nine estimators for the large population [ $N^{(1)} = 6,000,000$ ] with a sample size of 5000*

<b>Estimator</b>	<b>NULL</b>	<b>XRE</b>	<b>LIN<sub>0</sub><sup>†</sup></b>	<b>LIN<sub>1</sub></b>	<b>QUAD<sub>0</sub></b>	<b>QUAD<sub>1</sub></b>	<b>EXP<sub>0</sub></b>	<b>EXP<sub>1</sub></b>
<i>psm</i>	95.2 (5.3)	95.2 (5.3)	93.6 (4.1)	92.8 (4.1)	93.2 (5.1)	94.8 (5.1)	93.6 (5.0)	92.4 (5.0)
<i>unw</i>	96.8 (2.8)	87.2 (2.8)	95.6 (2.7)	88.8 (2.7)	0.0 (2.7)	0.0 (2.7)	22.0 (2.7)	26.4 (2.7)
<i>trim</i>	95.6 (3.3)	92.8 (3.3)	88.4 (2.9)	84.4 (2.9)	24.0 (3.2)	28.0 (3.2)	64.4 (3.2)	65.2 (3.2)
<i>xre</i> (analytical)	96.8 (2.8)	94.0 (3.2)	96.8 (4.2)	96.4 (4.2)	51.2 (3.9)	58.0 (4.0)	87.2 (4.1)	86.8 (4.1)
<i>xre</i> (bootstrap)	97.2 (2.9)	94.0 (3.3)	98.8 (4.5)	98.8 (4.5)	50.4 (3.9)	58.0 (4.0)	86.4 (4.1)	88.8 (4.1)
<i>xre-greg</i> (jackknife)	98.0 (3.4)	96.0 (3.7)	96.4 (3.9)	95.6 (3.9)	58.8 (4.5)	64.0 (4.5)	85.6 (4.0)	84.4 (4.2)
<i>lin</i> (analytical)	96.8 (2.8)	94.8 (3.2)	96.8 (2.4)	95.2 (2.6)	19.6 (3.2)	36.0 (3.5)	40.4 (2.8)	69.2 (3.2)
<i>lin</i> (bootstrap)	97.2 (2.9)	94.4 (3.3)	96.8 (2.4)	95.6 (2.6)	19.2 (3.2)	37.2 (3.5)	41.2 (2.8)	69.6 (3.2)
<i>lin-greg</i> (jackknife)	98.0 (3.4)	96.8 (3.5)	98.8 (2.9)	96.4 (2.9)	25.6 (3.8)	42.4 (3.9)	60.0 (3.3)	73.2 (3.5)
<i>npar</i> (analytical)	94.8 (3.0)	93.2 (3.5)	95.6 (2.6)	94.0 (2.8)	91.2 (4.4)	89.2 (4.4)	77.6 (3.7)	86.4 (3.8)
<i>npar</i> (bootstrap)	97.6 (3.6)	96.0 (3.6)	98.4 (2.9)	95.2 (3.0)	94.4 (4.9)	92.0 (4.8)	84.8 (4.2)	87.6 (4.1)
<i>npar-greg</i> (jackknife)	98.0 (4.0)	96.8 (4.1)	98.8 (3.4)	96.8 (3.4)	93.2 (4.6)	92.4 (4.7)	87.6 (4.4)	90.0 (4.4)

<sup>†</sup> Based on 249 simulated data sets.



are generated according to model (7) with the following choices ( $\forall h = 1, \dots, 18$  and  $\forall t = 1, \dots, 30$ ):

F1:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$ ,  $\delta_t = -2 + 3 \exp(-\frac{(t-15)^2}{50})$  and  $\mathbf{D} = \mathbf{0}$ .

F2:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$ ,  $\delta_t = -2 + 3 \exp(-\frac{(t-15)^2}{50})$  and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .

F3:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$ ,  $\delta_t = -2 + 3 \exp(-\frac{(t-15)^2}{50}) - \exp(-(t - 15)^2)$  and  $\mathbf{D} = \mathbf{0}$ .

F4:  $\delta_h = -1 + 2 \exp(-\frac{h}{9})$ ,  $\delta_t = -2 + 3 \exp(-\frac{(t-15)^2}{50}) - \exp(-(t - 15)^2)$  and  $\mathbf{D} = \sigma^2 \mathbf{I}$  with  $\sigma^2 = 0.02$ .

Figure 3 shows the time trends of a population that is randomly generated from F2 and F4. We consider population types F3 and F4 to investigate the robustness of the different estimation methods to a sharp valley in the overall trend. The non-parametric time function,  $f_t(\cdot)$ , is not able to perfectly describe this valley. We shall investigate whether the GREG adjusted estimators offer a useful solution in such case.

The following five estimators are used to obtain the time trend in the simulation study: (*psm*) the post-stratified mean is calculated at each time point; (*unw*) the unweighted mean is calculated at each time point; (*trim*) the trimmed mean ( $w_0 = 3$ ) is calculated at each time point; (*npar*) estimator (8) is used where the NPAR model is assumed for  $\delta$  and  $\mathbf{D}$ ; (*npar-greg*) estimator (9) with  $w_0 = 3$  is used with a NPAR model assumption for  $\delta$  and  $\mathbf{D}$ .

For each of the above five estimators, we calculate the average bias, variability and mean squared error from the obtained estimates at each of the 30 time points. For each time point, this is done in a similar manner as done for the prevalence estimation described in Section 5.1. The nominal coverage and average length of the 95% point-wise confidence interval are calculated. For the nonparametric time function, we set knots at all time points to ensure enough flexibility.

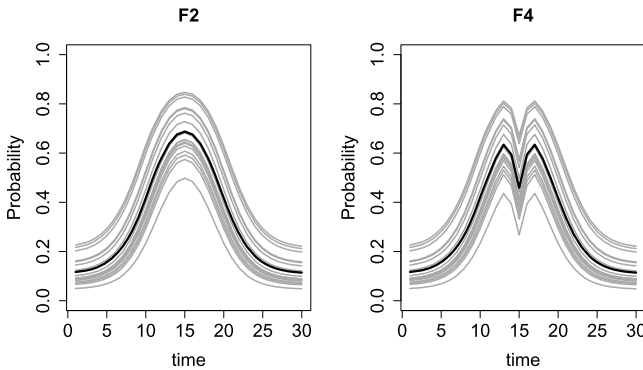


FIG. 3. Mean time trends of the separate post-strata (grey lines) and total population (black line) for a random population generated according to population models F2 and F4.

5.4. *Results for mean trend estimation.* A part of the results is summarized in Figure 4. The mean squared errors  $\times 10^4$  are presented for a population size of 150,000 and a sample size of 2500. More figures on the simulation study are given in Appendix D of the Supplementary Material [Vandendijck, Faes and Hens (2015)]. The *unw* and *trim* estimators are biased for all population models. The *npar* estimators are unbiased for F1 and F2. For F3 and F4, however, they are unbiased at the start and end of the time trend, but show some severe bias in the region of the sharp valley. No bias is present for the *psm* and *npar-greg* estimators. The variance of *psm* is larger when compared to the other estimators. The *npar-greg* estimator exhibits more variability than *unw* and *npar*, and has a comparable variability as the *trim* estimator.

In terms of MSE, this leads to a better performance of *npar* and *npar-greg* when compared to the other methods. For all time points, the *npar* estimator has lower MSE values for F1 and F2 when compared with *npar-greg*. For the functions with the sharp valley, F3 and F4, the mean squared error of *npar* is smaller than the mean squared error of *npar-greg*, except at those time points where the sharp valley occurs. The MSE of the *npar* estimator shows a steep increase in that region. Thus, for cases such as scenarios F3 and F4, the *npar* estimator is not preferred, but the *npar-greg* estimator is recommended. Under varying simulation conditions — increasing and decreasing population and sample sizes — the same general results are observed.

Results of the nominal coverage and average length of the 95% point-wise confidence intervals are presented in Figure 5 (see Appendix D for additional results).

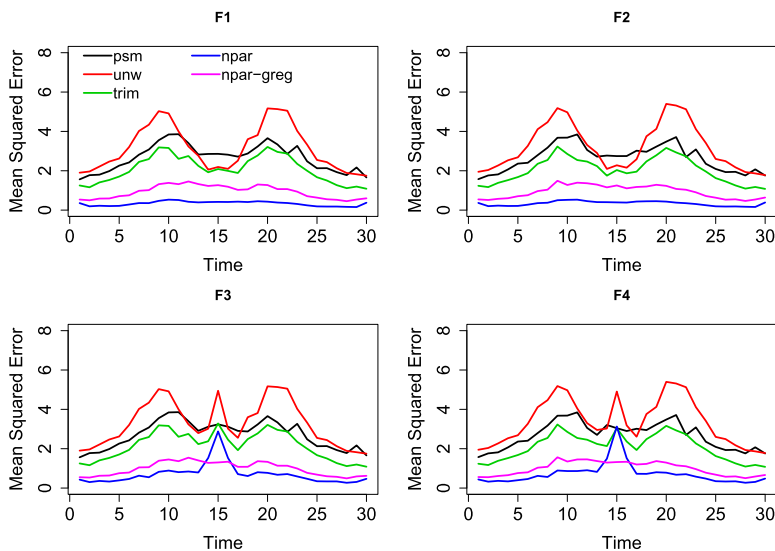


FIG. 4. Mean squared errors  $\times 10^4$  at each time point for population models F1, F2, F3 and F4 with population size  $N^{(2)} = 150,000$  and sample size 2500.

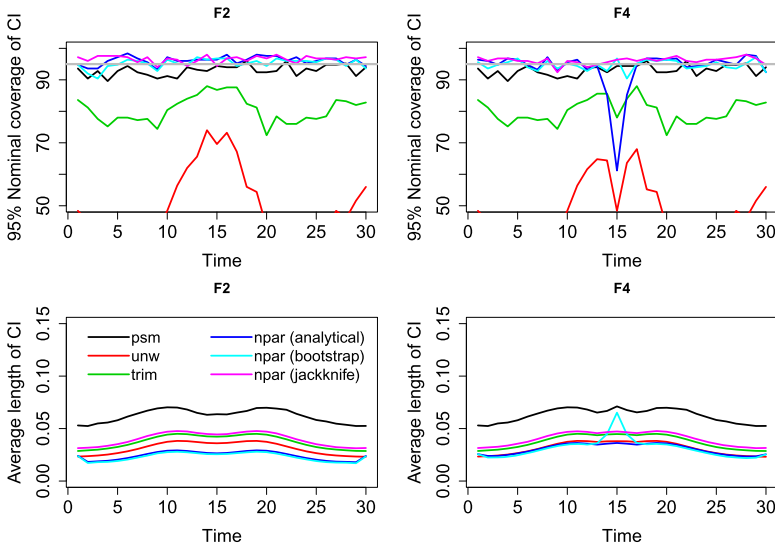


FIG. 5. Nominal coverage and average length of the 95% point-wise confidence intervals at each time point for population models F2 and F4 with population size  $N^{(2)} = 150,000$  and sample size 2500. The nominal coverage of some estimators is smaller than the range of the y-axis used and are therefore not depicted on the figure.

The *unw* and *trim* estimators have very poor coverage resulting from the bias associated with the estimates. The *psm* slightly underestimates the true coverage at almost all time points. For population model F2, both the analytical and bootstrap based CIs of the *npar* estimator have good coverage values. For F4, the coverage of the *npar* CIs based on the analytical variance shows a drop in the sharp valley area. The bootstrap variance-based CIs attain a good nominal coverage. However, for F4 this comes at the cost of wider CIs in the sharp valley area. The jackknife based CIs of the *npar-greg* estimator achieve good coverage for both F2 and F4 for all time points. The average lengths of the CIs of the *npar-greg* are comparable with those of *trim* and are substantially smaller when compared to *psm*.

**6. Motivating example revisited.** We apply the methods developed in Section 4 to the Great Influenza Survey of 2010–2011 introduced in Section 2. The same nine estimators as in Section 5.1 are used to calculate the overall ILI prevalence. The results are presented in Table 4. To calculate the CIs for *psm*, *unw* and *trim*, we use the variance formulas given in Table 1 in Little (1991). Both the analytical and bootstrap (with  $B = 250$ ) based CIs are calculated for *xre*, *lin* and *npar*. The jackknife procedure (with  $G = 250$ ) is used for the variance of the GREG-adjusted estimates.

The post-stratified mean yields the largest estimated prevalence. This is because the younger age groups, which have the highest ILI prevalence, receive high post-

TABLE 4

Estimates and 95% confidence intervals (CI) of the overall prevalence of ILI during the 2010–2011 influenza season based on nine estimators.

Method	Estimate	Analytical CI	Bootstrap CI	Jackknife CI
<i>psm</i>	7.10%	[5.31–9.45%]	–	–
<i>unw</i>	5.12%	[4.52–5.80%]	–	–
<i>trim</i>	5.61%	[4.88–6.46%]	–	–
<i>xre</i>	5.96%	[4.95–7.17%]	[5.00–7.10%]	–
<i>xre-greg</i>	6.16%	–	–	[4.92–7.68%]
<i>lin</i>	6.88%	[5.69–8.30%]	[5.76–8.21%]	–
<i>lin-greg</i>	6.82%	–	–	[5.66–8.20%]
<i>npar</i>	6.88%	[5.69–8.30%]	[5.61–8.41%]	–
<i>npar-greg</i>	6.82%	–	–	[5.66–8.20%]

stratification weights. The *xre* estimate yields an estimate in between the post-stratified and unweighted mean. The difference between the *xre* and *xre-greg* estimates is larger than the difference between the estimates of *lin* and *lin-greg*, and *npar* and *npar-greg*. This indicates that the XRE model assumption is most likely a misspecification for this data. The point estimates *lin* and *npar* are similar because the nonparametric function in the NPAR model is estimated to be essentially linear. The point estimates based on the LIN and NPAR model assumptions are closer to the post-stratified mean than to the unweighted and trimmed mean. The  $\sigma^2$  parameter was estimated to be 0.068 in the LIN and NPAR models. The post-stratified mean has the widest confidence interval associated with its point estimate. Confidence intervals of *lin* and *npar* are less wide. Reductions of 30–40% are observed when compared with *psm*.

The same five estimators as in Section 5.3 are used to obtain the overall time trend (the time trend represents the ILI incidence in this data example). We take both  $B = 250$  and  $G = 250$  for the bootstrap and jackknife procedures. Twenty equally spaced knots are used for the nonparametric time function. A sensitivity analysis with 10 knots and knots at all 27 time points revealed no meaningful differences. Figure 6 presents the estimated trends with the corresponding point-wise 95% confidence intervals. It is observed that the post-stratified mean yields a very wiggly curve. This trend estimate is not useful for practical usage. The unweighted and trimmed mean trend are more stable but most likely produce biased estimates as was observed in the simulation study. The *npar-greg* yields a trend that is less smooth than *npar*. The width of the confidence intervals of both *npar* and *npar-greg* yield noticeable reductions in length when compared to the post-stratified mean trend.

**7. Discussion.** In this article we examined methods that can deal with ordinal post-stratifiers that yield high post-stratification weights in observational survey

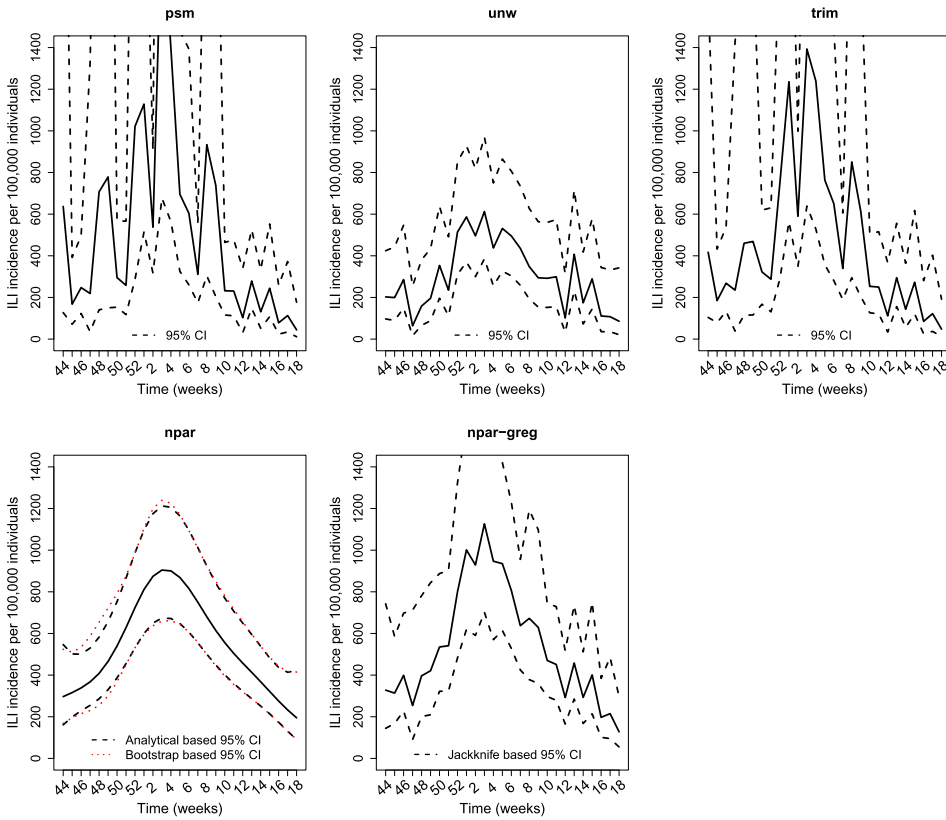


FIG. 6. Estimated incidence trends with corresponding 95% point-wise confidence intervals from the 2010/2011 Great Influenza Survey (GIS) using five estimation methods.

data. Standard methods such as the post-stratified, unweighted and trimmed mean break down in this situation due to either substantial bias or high variability of the obtained estimates. Weight smoothing models modify the standard methodology by imposing a random-effects structure on the post-stratum means. By predicting the unobserved values based on the weight smoothing model, an estimate is obtained. The post-stratification weights are implicitly smoothed in this manner. We described two extensions on the existing literature of weight smoothing models for binary data. First, we proposed a GREG-adjusted weight smoothed estimator that can be used for observational data for which no inclusion probabilities are available. Second, an extension for trend estimation was considered. XRE, LIN and NPAR model assumptions for the weight smoothing models were considered.

The construction of the weights  $\tilde{\pi}_h^{-1}$  for the GREG-adjusted estimator is based on weight trimming. Whereas the GREG estimator of Lehtonen and Veijanen (1998) in (4) is unbiased, the proposed estimator in (6) is biased. A related approach was also presented by Beaumont and Alavi (2006) in the context of miss-

ing survey data where the inclusion probabilities (design weights) are known. They proposed a robust-GREG estimator by truncating large weights to reduce the effect of widely dispersed design weights. In the simulation study in Section 5, we observed that the GREG-adjusted estimator (6) is more efficient than the post-stratified mean in all scenarios and more efficient than estimator (2) in some scenarios. [Beaumont and Alavi \(2006\)](#) also show that their estimator is more efficient than the standard GREG estimator when widely dispersed design probabilities are present.

In the simulation study in Section 5 and the data example in Section 6, we used  $w_0 = 3$  for the GREG-adjusted estimates. To investigate the impact of this cutoff value, we performed some extra simulations and investigated the GIS with other values of  $w_0$ . These results can be found in Appendix E of the Supplementary Material [[Vandendijck, Faes and Hens \(2015\)](#)]. As expected, smaller values of  $w_0$  yield estimates that have a larger bias, but smaller variance; the opposite is true for larger values of  $w_0$ . Developing systematic methods to choose the value of  $w_0$  for the GREG-adjusted estimates is an important future research endeavor. To this purpose, existing approaches to estimate the optimal cutoff value can be of help [see, e.g., [Cox and McGrath \(1981\)](#), [Little \(1993\)](#), [Potter \(1990\)](#)].

Simulation results (see Appendix E of the Supplementary Material) suggest that for smaller sample size ( $n = 150$ ) the estimators *npar* and *npar-greg* still perform adequately but do not yield superior MSE results. For smaller sample sizes it is much harder to fit a nonparametric regression model to the data and this results in an increase in the variability of the *npar* and *npar-greg* estimators. Therefore, care needs to be taken when using the proposed methods with small sample sizes.

Variance estimates of the proposed methods were described. The analytical formula to calculate the variance ignores the uncertainty in estimating the variance parameters. The bootstrap procedure for the weight smoothed estimates and the jackknife resampling approach for the GREG-adjusted estimates were proposed as alternatives. Another approach is turning to Bayesian methods. A *t*-based correction to account for the uncertainty can also be considered in the analytical case [[Lazzeroni and Little \(1998\)](#)]. In the simulation study, we observed that a normal reference distribution yielded confidence intervals with appropriate coverages.

Computationally, weight smoothed estimates are more complex than the post-stratified, unweighted and trimmed means. This complexity has two main sources: (i) Weight smoothed estimates are specific to each separate survey outcome. For the analysis of another response from the same survey, new computations are necessary. On the other hand, the unweighted, post-stratified and trimmed mean have fixed weights for each response in the same survey. As stated by [Lazzeroni and Little \(1998\)](#), any procedure directed at reducing variance must tailor the weights, depending on the degree of association of the post-stratifier with the outcome. (ii) To calculate a weight smoothed estimate, a GLMM must be fit which is computationally hard. A computer intensive resampling procedure must be used to obtain

variance estimates. However, modern computing power has made these computations practicable.

This article focused on binary survey outcomes. Nonetheless, because the GLMM framework is used to estimate the models, extensions to other distributions in the exponential family of distributions can be performed without much extra work. The formulae and methods proposed in our paper are still valid for outcomes with nonbinary distributions. Only one post-stratifying variable was considered in this article. Extending the weight smoothing models to more than one post-stratifier is possible without much effort and is a topic of further research.

Model (7) used for the trend estimation is possibly not flexible enough for the application example. There is evidence in the influenza literature that the timings and lengths of influenza-like illness peaks vary by age groups [see, e.g., Adler et al. (2014)]. Model (7) assumes additive effects of time and age groups and would thus not allow for this possible interaction between age and time. The inclusion of such an interaction term falls out of the scope of this paper. Nevertheless, in general, model (7) can be extended to allow for interaction effects.

From the results in our simulation study, we recommend the use of the GREG-adjusted weight smoothed estimate with a NPAR model assumption. It performed the most consistent over all simulation conditions. This robustness comes, however, with a cost of efficiency when the true underlying model is less complex.

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## SUPPLEMENTARY MATERIAL

**Additional details and results** (DOI: [10.1214/15-AOAS874SUPP](https://doi.org/10.1214/15-AOAS874SUPP); .pdf). The reader is referred to the online Supplementary Material for more information on how the models can be cast in the GLMM framework (Appendix A), for more details on the estimation method (Appendix B), for annotated SAS and R programs (Appendix C), for additional simulation results (Appendix D), and for additional results for different values of  $w_0$ , additional results for smaller sample size and results on model fits and other spline basis functions (Appendix E).

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