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RESEARCH ARTICLE

Statistical Modelling

A comparison of model parameter estimation methods for complex survey survival data

IT Jayamanne*, A Ramanayake and RV Jayatillake
Department of Statistics, Faculty of Science,University of Colombo, Colombo 03, Sri Lanka.

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Abstract: When estimation of model parameters is the target in survey data, the inference on the model can be based on a pure model-based approach or a model-assisted approach which is a hybrid approach combining design-based and model-based methods. This study aims to compare these methods for survival data that are gathered from a stratified random sampling design. The Accelerated Failure Time (AFT) model was fitted to describe the relationship between the censored response variable and the explanatory variables. Resampling methods with different sample sizes and different sampling designs from a real dataset were considered in the study to generate samples. The AFT models were fitted to each of the samples using, firstly a pure model-based method ignoring the survey design and weights, secondly a model-based method with survey weights, and finally a model-assisted method considering both survey design and weights. Squared bias, variance, and mean squared error (MSE) were used to compare the three approaches. The AFT model, with all covariates and the best AFT model with the best set of covariates and distribution, were analyzed. Even though it was challenging to select the best method for all cases, the second and third approaches worked better for small samples than the first approach.

Keywords: Analytical studies, model-assisted, post-stratification, survey design, survival data.

INTRODUCTION

In analytical studies, groups of subjects are compared to estimate the magnitude of the association between exposures and outcomes. Statistical models play a vital role in identifying this association. In general, the primary focus of the statistical model is on estimating and evaluating the model and its associations. Thus, data analysts often neglect to consider the complex sampling design features, such as stratification, clustering, and unequal probability of selection when performing analytical studies. Data are frequently analyzed as if they are from a simple random sample, perhaps due to convenience or lack of proper guidance. The inference on the model can be performed based on a pure model-based approach or a model-assisted approach. The latter is a hybrid approach combining both design-based and model-based methods. While the model-based framework, relies only on modelling, particularly the model distributional assumptions, the model-assisted framework incorporates the random sampling mechanism (Opsomer, 2009; Sterba, 2009; Pfeffermann, 2011).

Survey design weights play a vital role in incorporating survey design features of the complex survey data into the model. In the model-assisted approach, the sampling weights are used to estimate the parameters, and besides the sample design is used to determine the variances of the estimates (Lohr, 2010). Complex survey designs namely, stratification, clustering, and multistage sampling lead to estimates having standard errors different from data obtained using simple random sampling (SRS). Besides, post-stratification and nonresponse adjustments also affect the variance. The design effects (deff) are used to quantify the effect of the design on the variance of the point estimates (Lumley, 2010; Lohr, 2010). Several methods for estimating variances for regression estimates for complex survey data were introduced, namely Taylor linearization and resampling methods as Balanced Repeated Replication, Jackknife, and Bootstrap. These methods were implemented in

* Corresponding author (imali@stat.cmb.ac.lk; https://orcid.org/0009-0002-0314-5021)

some software packages to correct the standard error in complex survey data. In the absence of the model-assisted method implementation in the statistical software, Lohr (2010) advised running the programs with design weights to acquire accurate parameter estimates. She further mentioned several reviews about the effect of survey weights on regression estimates. She recommends performing the analysis with and without design weights to perceive the difference in the model parameter estimates.

Quite a few theoretical pieces of evidence (Sarndal *et al.*, 2003; Binder & Roberts, 2009; Lohr, 2010; Lumley, 2010) explained the pros and cons of model-based and model-assisted inference on regression parameters. However, there is a lack of substantial evidence to evaluate the inference framework for analytical studies for complex surveys on survival data. Most of the simulation studies focused on generating hypothetical data and comparing the performances of linear or logistic regression models. A few authors (DuMouchel & Duncan, 1983; Binder *et al*., 2005), have used real data to distinguish the differences. Even in these studies, only a few aspects of the model are evaluated and none of the studies was based on survival data. Hence, the main focus of this study is to reduce this gap by designing a study based on a real dataset to distinguish the inference methods for complex surveys of survival data.

Even though pure model-based approaches were well developed in almost all statistical packages many years ago, model-assisted inferences were only implemented recently in some statistical software. Different mechanisms were used to integrate the complex design structure of the data into different models during the model estimation procedure. The focus of this research solely will be on the Accelerated Failure Time (AFT) model. The AFT model is a popular model used to explain the relationship between a survival response (time to an event) and other explanatory variables. A typical feature of the survival response is that the time to an event is not always observed, and observations are subject to censoring. The AFT model relates the logarithm of the survival time linearly to covariates. Specific parametric error distribution for the AFT model is assumed and hence a particular survival distribution. If the survival data is from a simple random sampling, the pure model-based inferences can be used to estimate the model parameters. In the presence of complex survey data, one can either follow a pure model-based approach or a model-assisted approach. Howbeit, most of the pure model-based software packages allow the user to include survey weight for the analysis. Thus, one can perform a study based on these three approaches. Though selecting the best approach prior to analysis is a dilemma, after analyzing data using these three approaches, the analyst may be able to select the best model based on the Akaike information criterion (AIC) or the Bayesian information criterion (BIC) and he validation of model assumption. However, it would be beneficial if proper guidelines can be given to the analyst prior to the analysis to select the best approach on available data. Furthermore, it is also necessary to assess the impact of the survey design on the conclusion from the analysis.

In consequence, this study aims to compare the three approaches mentioned above for survival data that are gathered from a complex survey design with proportionate and disproportionate stratified random samples. It will describe the strengths and weaknesses of the three approaches via a simulation study. The difference in the model performances for the full model, the best fitted model (best set of covariates), changes in the distribution will also be investigated. Besides, the effect of post-stratification on model parameters will also be studied.

MATERIALS AND METHODS

As discussed in the introduction, a simulation study was carried out to achieve the objective of the study. First, a real dataset was obtained to generate data for the simulation study. Then the theoretical difference between the three methods of estimation is described in section 2.2. Next, the simulation framework was designed to achieve the objectives of the study.

Data for the simulation study

A survey was conducted in the year 2016 on a stratified random sample of all Arts graduates from Sri Lankan universities who had graduated in 2012 to identify the changes in their employment over time. The waiting time for the first employment is the response variable, and the qualitative explanatory variables considered are university, gender, ethnicity, area lived, type of school attended, parent's highest education, civil status at the time of employment (civilStatusAtFE), English grade at GCE O/L, English grade at GCE A/L (ALEnglish), the class received (Class) and the type of degree (DegreeType). There were 386 complete records that consisted of 15 interval-censored and 10 right-censored data (Jayamanne & Ramanayake, 2018). Almost all the past studies on finding factors associated with the waiting time for the first employment had followed a model-based approach (Grilli *et al.*, 2001; Moore, 2006; Salas-Velasco, 2007). The log-logistic AFT model was chosen as the best fitted model to explain the relationship between the time to first employment and the explanatory variables (Jayamanne & Ramanayake, 2017). The best set of covariates and their coefficient were estimated using the survival package in R following the stepwise backward selection method having the lowest AIC and BIC. Only the Class, ALEnglish, Gender, civilStatusAtFE, DegreeType, and Ethnicity variables were significant, and these values are depicted in Table 1. Since all the variables are qualitative, coefficient values are depicted with respect to the reference level.

Table 1: Parameters of the AFT model

	Variable	Coefficient	Parameter value
1		(Intercept)	1.5312
2	Class	General	0.6166
3		Second Lower	0.5183
4		Second Upper	0.511
5	ALEnglish	Absent	1.0476
6		B	0.8313
7		C	1.1059
8		F/W	1.0021
9		S	1.2355
10	Gender	Male	0.3529
11	civilStatusAtFE	Single	-0.2416
12	DegreeType	Special	-0.2487
13	Ethnicity	Sinhala	-0.6972
14		Tamil	-0.0792
15		Log(scale)	-0.7545

To compare the estimation methods precisely, the parameters of the population AFT model are required. But since population survival data are unavailable, the dataset itself was considered to be the population data. Hence the above model coefficients were assumed to be regression parameters.

The theoretical concepts

In AFT models, the direct effect of the explanatory variables on the survival time is measured. Assume survival data are the realization of n random variables $T_1, ..., T_n$, depending on the covariate vector, $X_i = (X_{i1}, ..., X_{ip})'$. The AFT model is denoted as,

$$log\, T_i \ = X'\beta + \varepsilon \qquad ...(1)$$

where $\beta = (\ \beta_1, ..., \beta_p)$ represents unknown regression parameters and $\varepsilon = (\varepsilon_1, \ldots, \varepsilon_n)$ are independent and identically distributed with density $g(e)$. Under the parametric approach, error density consists of μ, the location parameter (intercept) and σ, the scale parameter.
Hence the equation can be rewritten as,

$$log\, T_i \ = \mu + X'\beta + \sigma\varepsilon^* \qquad \ldots (2)$$

where ε^* is the standardized error term with a particular survival time distribution. The most commonly used survival distributions for AFT are exponential, Weibull, log-logistic, lognormal, and generalized gamma.

The maximum likelihood estimation method can be used to estimate *p*+2 parameters in the pure model-based approach using the likelihood L. When $L_i(\theta)$ corresponds to the parametric model for independent observations with density $f_i(t;\theta)$ and survival function $S_i(t;\theta)$,

$$L = \prod_{i=1}^{n} [L_i(\theta)] = \prod_{i=1}^{n} [f_i(t_i;\theta)]^{\delta i}[S_i(t_i;\theta)]^{1-\delta i} \quad \ldots(3)$$

where δ_i is the event indicator. δi =1, if the i^{th} observation is an event and δi =0, if the i^{th} observation is censored.

When the AFT model is extended to complex survey data, the first fundamental difference in the likelihood function is to incorporate sampling weight w_i for $i = 1, \ldots, n$ as follows. This likelihood is called pseudo-likelihood

$$PL = \prod_{i=1}^{n} [L_i(\theta)]^{w_i}. \quad \ldots(4)$$

Hence, both model-based with sampling weight and model-assisted approaches use sampling weights to estimate *p*+2 parameters with the use of pseudo-likelihood. Parameter estimations are obtained using the Newton Raphson method.

The standard errors of the model-assisted parameter estimates were obtained using Taylor series linearization (Lohr, 2010; Lumley, 2020). This is the corrected standard error which incorporates both sampling weight and design. In the model-based with sampling weight approach, only sampling weights are incorporated to get the corrected standard deviation. It uses robust variance estimation.

A simulation study was carried out to achieve the objectives of this study as follows.

The Framework of the simulation study

The extensive simulation study is discussed in three steps;

Step 1: Generating data
Step 2: Fitting AFT Models under the three approaches described earlier
Step 3: Comparing the estimates

Step 1: Generating data

Considering the survey data as the population $(N = 386)$, 1000 samples were drawn by resampling with replacements (bootstrap replicates) for each different sample size and survey design.

Sample sizes: 50, 100, 200

Sampling design:

- Stratified sampling with equal weights by taking the gender as strata (*e.g.*, when $n = 100, Female = 78\ and\ Male = 23$)
- Stratified sampling with unequal weights by considering the gender as strata
  - Higher weights for females (when $n = 100, Female = 50\ and\ Male = 50$)
  - Higher weights for males (when $n = 100, Female = 85\ and\ Male = 15$)
- Stratified sampling with equal weights by choosing the degree as strata
- Stratified sampling with unequal weights by taking the degree as strata
  - Higher weights for general degree holders
  - Higher weights for special degree holders

Altogether 18 $(= 3\ inferences \times 6\ different\ designs)$ different cases with each having 1,000 stratified random sample data were considered in this study.

Step 2: Fitting AFT models:

AFT models were fitted for each dataset using three approaches namely
- Pure model-based approach
- Model-based with sampling weights
- Model-assisted approach

The parameter estimates of the full model with all covariates were obtained under the three inference approaches. However, the best fitted model for the sample data may be different from the population model. Hence it would be improper to consider an insignificant explanatory variable in the AFT model. Thus, the best fitted model with only significant covariates (variables in Table 1) was considered for the model comparisons under the three approaches, by following the stepwise backward selection method having the lowest AIC and BIC with the best fitted distribution suited for the sample data. The best fitted distribution was selected by selecting the model which gives minimum AIC values out of the models fitting all available distributions. Furthermore, the best fitted model parameter estimates with post-stratified weights were also compared.

These models were fitted using the Survival package in R to perform pure model-based and model-based with survey weights approaches, while the Survey package is used to perform a model-assisted method considering design features such as stratification and survey design weights (Bogaerts *et al*, 2017; Lumley, 2020).

Step 3: Compare estimates

A variety of measures such as bias, variance and Mean Square Error (MSE) were considered for comparing the different approaches. The paramenter β_{ij} represents the j^{th} coefficient of i^{th} sample.

Average parameter estimates of $\beta_j = \sum_{i=1}^{1000} \frac{\widehat{\beta_{ij}}}{1000}$ for $j = 1, \ldots, p$ …(5)

where p is the number of coefficients present in the best fitted model, and $\widehat{\beta_{ij}}$ is the estimate of parameter β_{ij}.

Individual parameter estimates

Relative bias of $\beta_{ij} = relB(\widehat{\beta}_{ij}) = \frac{(\widehat{\beta}_{ij} - \beta_j)}{\beta_j}$ …(6)

Squared bias of $\beta_{ij} = SB(\widehat{\beta}_{ij}) = \left(\widehat{\beta}_{ij} - \beta_j\right)^2$ …(7)

MSE of $\widehat{\beta}_{ij} = MSE(\widehat{\beta}_{ij}) = SB(\widehat{\beta}_{ij}) + var(\widehat{\beta}_{ij})$ …(8)

Total parameter estimates for full model

Total squared bias $= TSB(\widehat{\beta}_i) = \sum_{j=1}^{p} \left(\widehat{\beta}_{ij} - \beta_j\right)^2$ …(9)

Total variance $= Tvar(\widehat{\beta}_i) = \sum_{j=1}^{p} var(\widehat{\beta}_{ij})$ …(10)

Total MSE$= TMSE(\widehat{\beta}_i) = \sum_{j=1}^{p} MSE(\widehat{\beta}_{ij})$ …(11)

Average parameter estimates for the best model

Let $\propto_{ij} = \begin{cases} 1 & if\ \widehat{\beta}_{ij}\ is\ in\ the\ best\ fitted\ model \\ 0 & otherwise \end{cases}$

Average squared bias $= ASB(\widehat{\beta}_i) = \frac{\sum_{j=1}^{p} \propto_{ij} * \left(\widehat{\beta}_{ij} - \beta_j\right)^2}{\sum_{j=1}^{p} \propto_{ij}}$ …(12)

Average variance $=AVar(\widehat{\beta}_i) = \frac{\sum_{j=1}^{p} \propto_{ij} * \text{var}(\widehat{\beta}_{ij})}{\sum_{j=1}^{p} \propto_{ij}}$ ...(13)

Average $MSE = AMSE(\widehat{\beta}_i) = \frac{\sum_{j=1}^{p} \propto_{ij} * \text{MSE}(\widehat{\beta}_{ij})}{\sum_{j=1}^{p} \propto_{ij}}$ ...(14)

RESULTS AND DISCUSSION

The core findings of the simulation study are depicted in this section using the statistics described in the methodology.

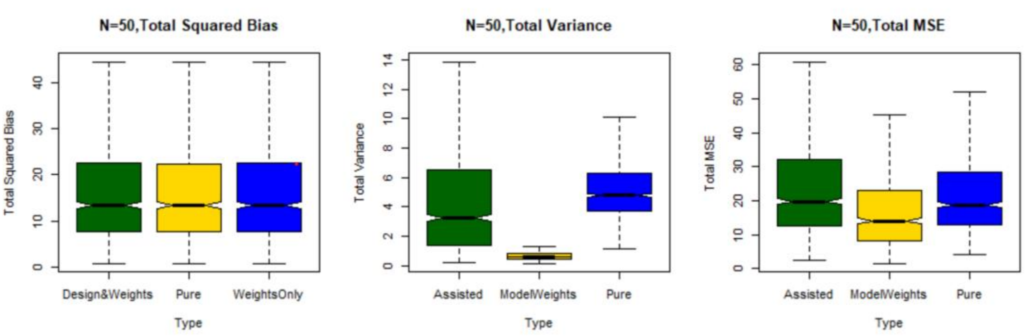

Figure 1: Total parameter estimates of the full model

Full model

Figure1 depicts the total parameter estimates of the full model under three inference approaches for the 1000 sample data. Even though the overall relative bias is quite similar for the three methods, total variance and MSE are quite varied for the three approaches. The total variance is lowest for the model-based method with weight (yellow). Even though the variability of the total variance of the model-assisted method (green) is higher than that of the pure model-based approach (blue), the median value is lesser for the model-assisted method. A similar trend is shown by total MSE as well. The model-based approach with weight does not give the correct variance estimates as it only takes weights but not the design into consideration when estimating variance.

It is essential to explore the individual differences of the beta coefficient estimates under the three approaches. For almost all cases, there is not a considerable difference in the squared bias among the three methods. However, as illustrated in Figure1, the estimated variance varied significantly. Regardless of the approach used, variances are higher for the intercept, ALEnglish and class coefficients. For almost all the coefficients, the variance is the highest for pure model-based (green) followed by model-based with weights. This indicates that the model-assisted approach has given rise to parameter estimates with smaller variance by incorporating both the design features such as stratification as well as unequal weights. Similar trends are shown for the other 17 cases and the MSE as well.

The best fitted model

As seen in Figure 2, the main reason for the higher variances for A/L English is the insignificance of the coefficients in the full model in the simulated data. Therefore, it is essential to consider only the explanatory variables that are significant in the model in order to select the best fitted AFT model for each sample. At first, the best fitted distribution of the AFT model was selected among a set of distributions using minimum AIC value, and then the best fitted collection of covariates was also chosen using minimum AIC values. It was noticed that the best fitted distribution for the AFT model is the same for all three approaches for all cases. It may be because the best fitted distribution is selected first for the waiting time variable, and it is not affected by the parameter estimation approaches. Thus, the variation in the best fitted distribution is depicted in Figure 3 by the sample size. It can be seen that with the increase in sample size, sample data attain population distribution.

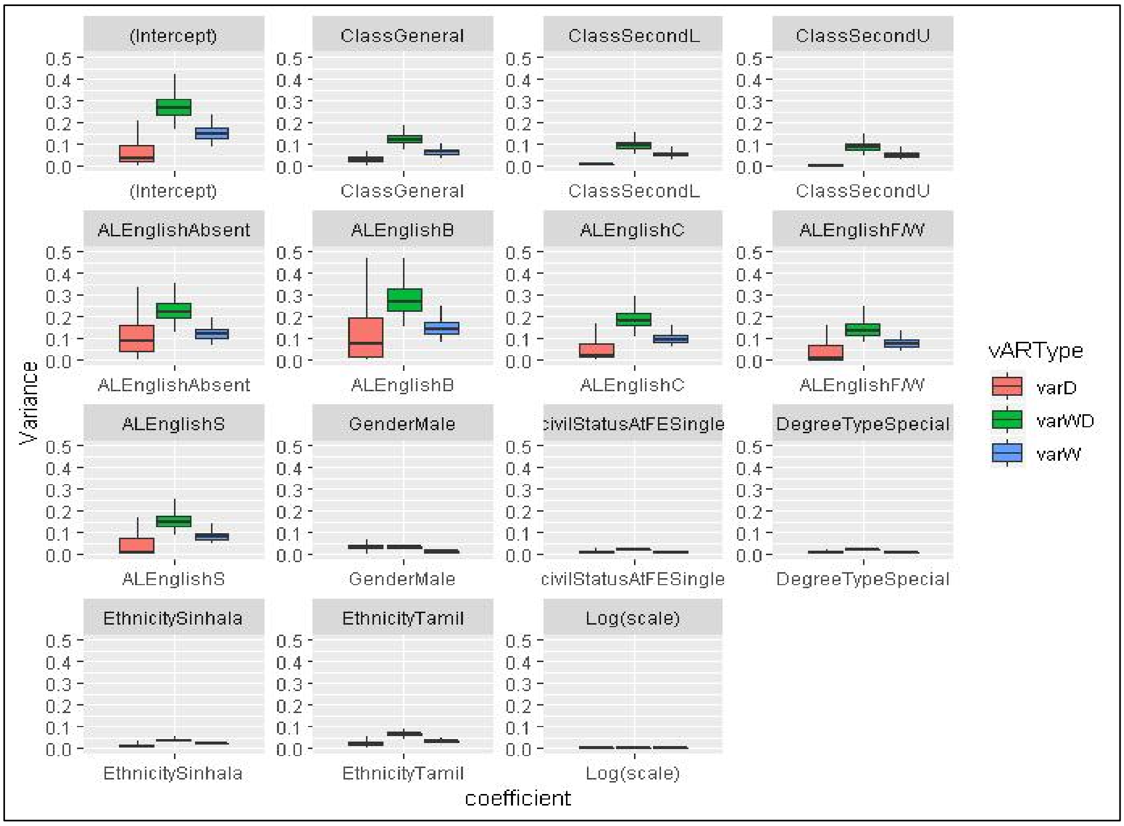

Figure 2: Estimated variances in beta coefficients by inference approach (n = 200, unequal weights, gender)

(Note: varD - variance from Model-assisted approach; varWD - variance from Pure Model-based approach; and varW - variance from Model-based with weights approach.)

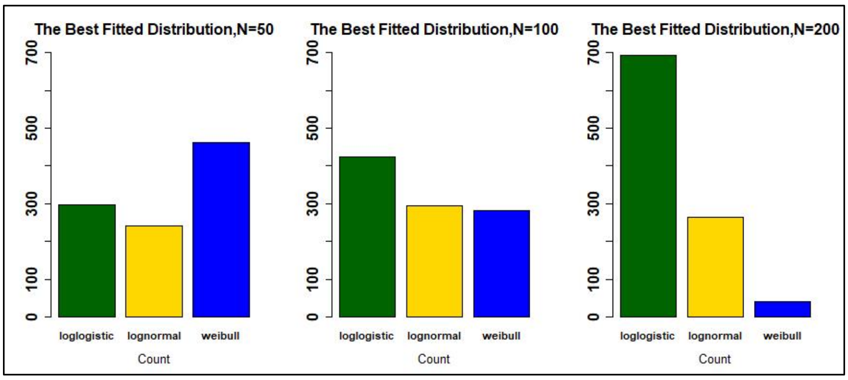

Figure 3: The best fitted distribution of the AFT model

Figure 4 depicts the summary of the significant covariates selected in the best fitted AFT model for 1000 samples for each different sample size. As the significance of the covariates depend on the selected data for the model, for some covariates the percentages tend to increase and decrease non-monotonically with sample size. However, it can be seen that with the increase in the sample size, ethnicity and gender variables are almost certainly included in the best fitted model for all three approaches.

As seen in Figure 4, a majority of the covariates are included in the best fitted model for all the sample sizes when the estimates are based on the model-based with weights approach compared to the other two approaches.

N=50, equal weights				N=100, equal weights				N=200, equal weights			
	Model Assited	Model with weights	Pure model	Covariate	Model Assited	Model with weights	Pure model		Model Assited	Model with weights	Pure model
Class	59%	93%	59%	Class	55%	89%	55%	Class	47%	77%	50%
ALENglish	73%	99%	61%	ALENglish	65%	97%	73%	ALENglish	76%	94%	77%
Gender	85%	85%	89%	Gender	92%	90%	91%	Gender	98%	98%	98%
CivilStatus	86%	81%	89%	CivilStatus	88%	78%	84%	CivilStatus	89%	87%	88%
Degree	86%	82%	89%	Degree	88%	80%	87%	Degree	91%	88%	90%
Ethnicity	91%	98%	94%	Ethnicity	99%	100%	98%	Ethnicity	100%	100%	100%

Figure 4: Percentages of the covariates present in 1,000 sample by sample sizes

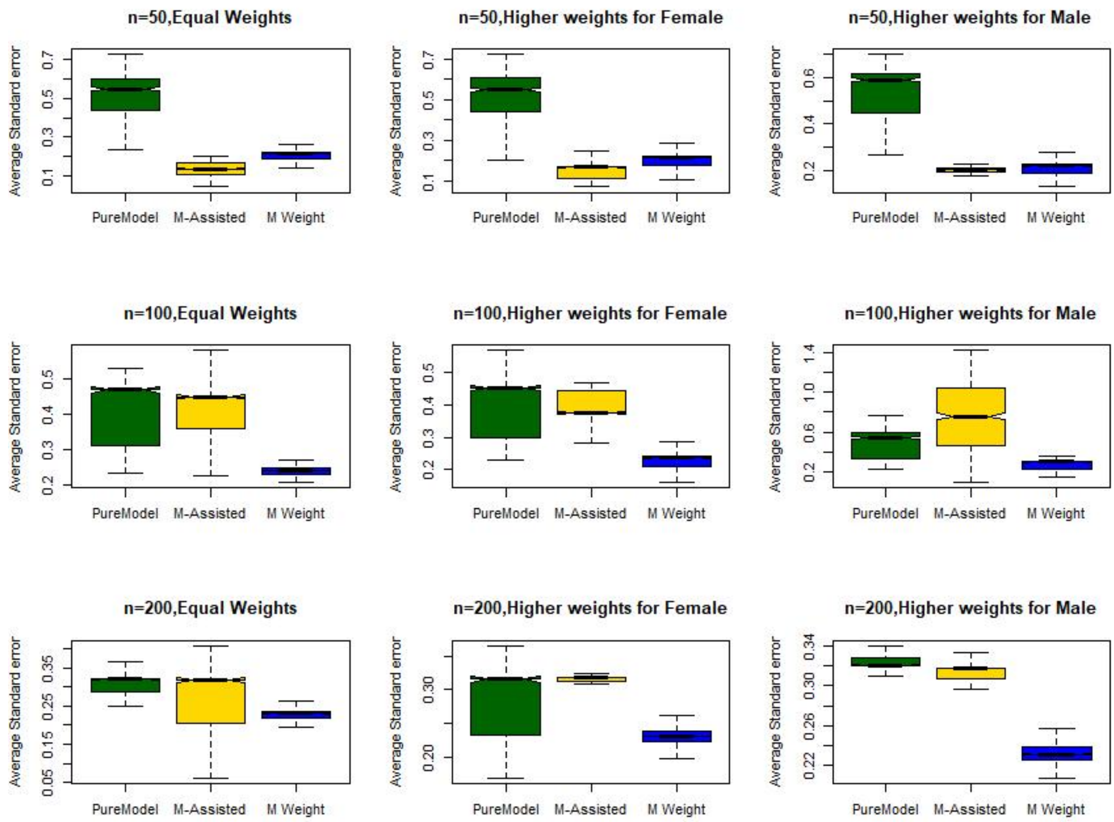

Figure 5: Average Squared Bias of the best fitted model with the best fitted distribution

It is essential to compare the estimations of the three methods for the best AFT model with the best fitted distribution. As stated in the methodology, the average squared bias, average standard error, and average MSE were considered instead of the total due to the absence of insignificant coefficients in some of the best fitted models using equations 12 to 14. The distributions of these statistics for 1000 observations are depicted in Figures 5 to 7. It is clear that the standard bias, standard error, and MSE are reduced with the increase of sample size.

As illustrated in Figure 5, when the sample sizes are proportionate to the population (sampling is ignorable), the three approaches end up having a similar average squared bias. However, when the sample sizes are disproportionate to the population sizes of the strata, the model-based approach with weights performed well when a fewer number of people are taken from small samples. Furthermore, it seems that the model-assisted and model-based with weight approaches perform well when sample sizes are small. For larger sample sizes, the model-assisted approach outperforms other approaches in terms of average squared bias.

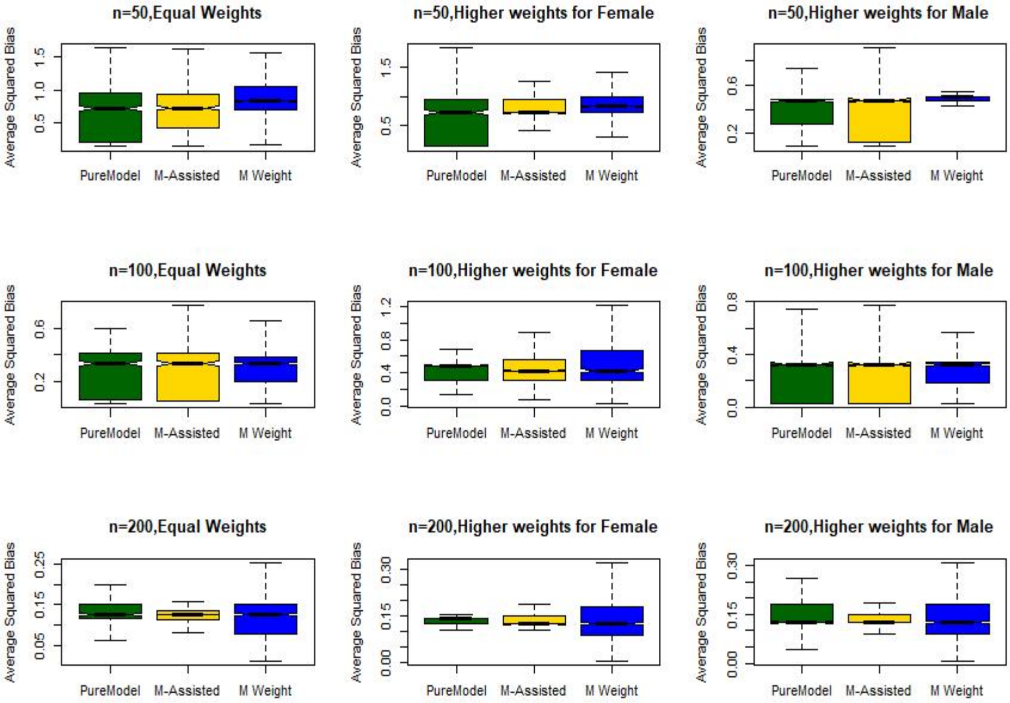

Figure 6: Average Standard Errors of the best fitted model with the best fitted distribution

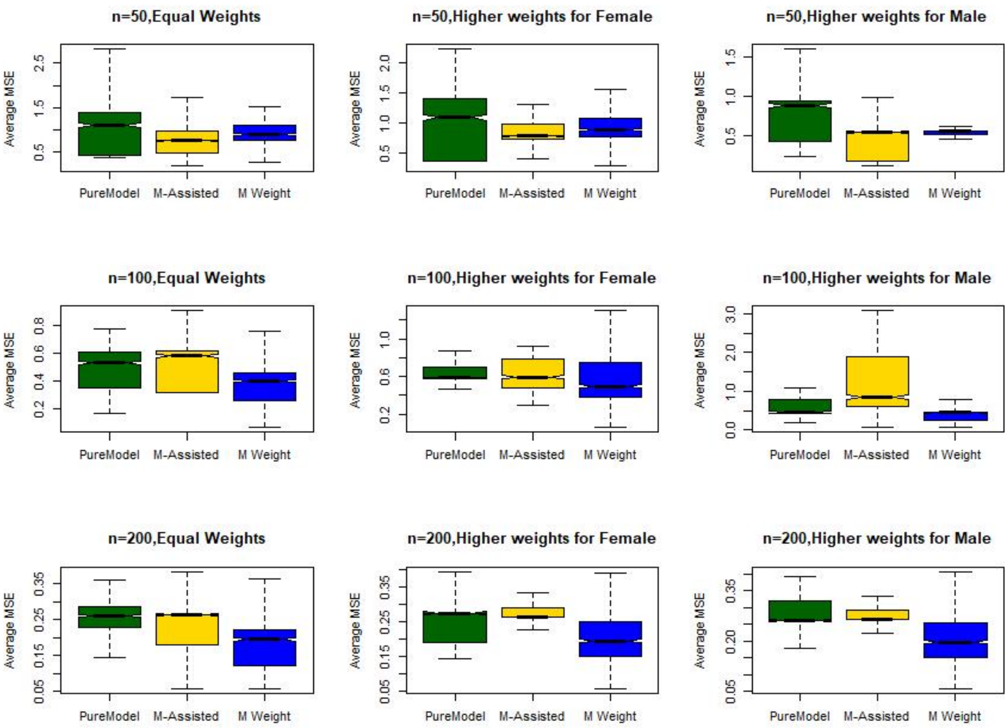

Figure 7: Average MSE of the best fitted model with the best fitted distribution

According to Figure 6, average standard errors are less for both model-assisted and model-based with weights approaches compared to the pure model-based approach for small sample size. Even though the model-based with weights approach outperforms the other two approaches for all cases, standard errors are not adjusted for the design.

The average MSE also depicted similar results, suggesting that the model-assisted or model-based with weights approaches are suitable for AFT model parameter estimation for small sample fractions (Figure7). Also the model-based with weights approach outperform the other approaches in all cases.

Effect of stratification and post-stratification

Even though all the above analysis considers gender as the stratification variable, the degree variable was also used to generate stratified random samples and most of the results are quite similar to those obtained when stratified by the gender variable.

Post-stratification is a method used for adjusting the sampling weights to account for underrepresented groups in the population. It is vital to investigate post-stratification on AFT model parameter estimations. Each stratified random sample is post-stratified by different variables such as ethnicity, class, and civil status to adjust weights to signify under representations. The best fitted AFT models with the best set of covariates were fitted for the post-stratified data using the three approaches. Even though there are only slight differences in the parameter estimation for the full model among the three strategies for post- stratified data, the differences are quite noteworthy for the best fitted model. Figures 8 and 9 show the average squared bias and standard error when n = 100, with disproportionate weights when gender was considered as the stratum variable. The top left most figure gives the estimates without a post-stratification and the rest are the estimates after post-stratifications.

As seen in Figure 8, model-based with weights and model-assisted estimations tend to be less biased after adjusting for post-stratification, except only when post-stratification is done by the class. Though it is difficult to identify the best post-stratification variable, ethnicity and civil status work better than other variables for both model-assisted and model-based with weights approaches.

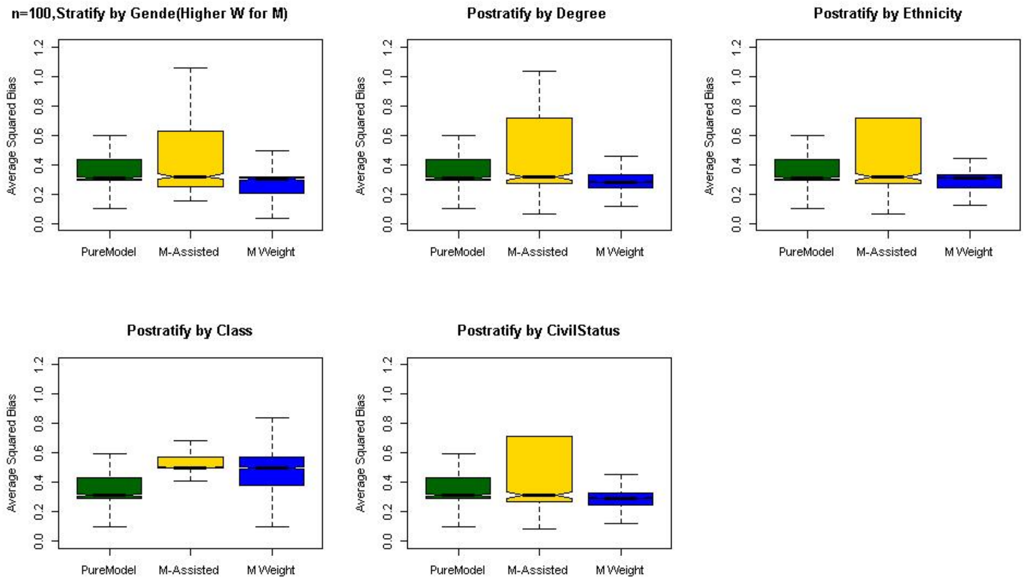

Figure 8: Average squared bias of the best fitted model for post-stratified samples

As depicted in Figure 9, the average standard error of the estimates of the model-based with weights and model-assisted approaches decreased after post-stratification comparatively, except when post-stratified by class for the model-assisted method. The average MSE follows a similar trend as in Figure 9, suggesting post-stratification by civil status, which produced a smaller MSE for both model-assisted and model-based with weights methods. Hence, both are comparatively better than pure model-based estimates.

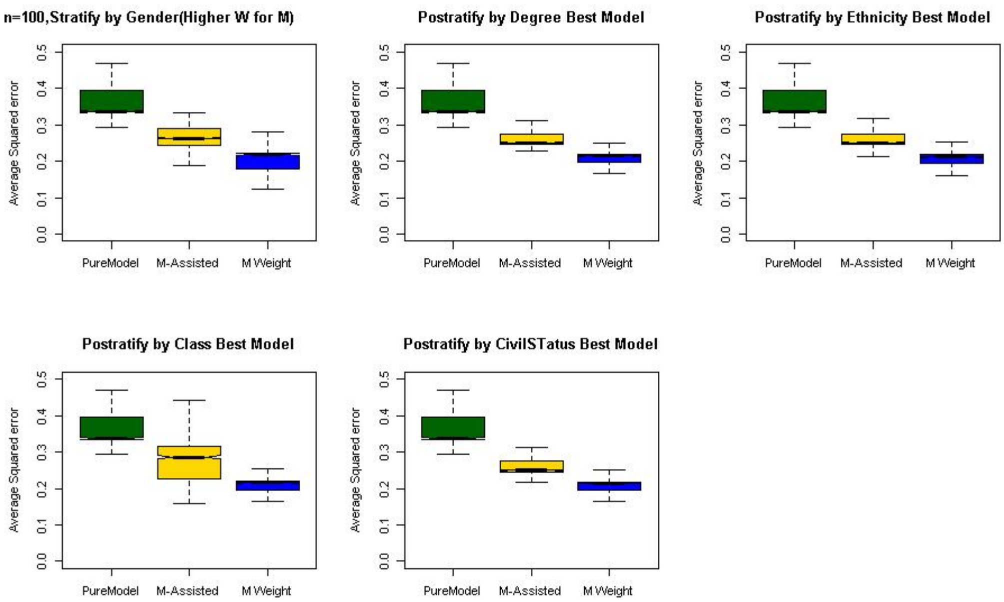

Figure 9: Average squared error of the best fitted model for post-stratified samples

As this study is based on a real dataset that contains data for waiting time to first employment of Arts graduates in Sri Lanka and explanatory variables, it is also important to highlight the findings of the AFT model. The AFT model revealed that gender, GCE Advanced level English grade, civil status, the class received, degree type, and ethnicity were the significant factors associated with the waiting time for the first employment of Arts graduates in Sri Lanka. Though the final model fitted to waiting time in this study was different from the past studies (Grilli, *et al*., 2001; Moore, 2006; Salas-Velasco, 2007), gender and degree type are the common factors associated with the waiting time to first employment in all studies.

CONCLUSION

This section highlights some of the substantive findings of the study by comparing the pure model-based, model-based with weights, and model-assisted methods of estimating model parameters of the Accelerated Failure Time model for complex survey data. It is commonly believed that the sampling weights and stratification will have a significant effect on these three ways of estimations. To explore these differences, a simulation study was conducted on a real dataset by resampling.

When all the covariates of the population model were considered for the sample data, the insignificant covariates deviate quite a lot from the true parameter values. Even though it was challenging to select the best approach for the full model based on squared bias, it was noticed that the model-based with weights and model-assisted methods outperform pure model-based methods in terms of standard error and MSE of estimates. However, as it is not appropriate to include insignificant variables in the model, the best set of covariates was identified for each sample based on the minimum AIC values for the AFT model with the best fitted distribution. It was discovered that most of the covariates of the model-based with weights method were significant compared to the other two. In general, model-assisted and model-based with weight approaches considerably improve with a decrease in sample sizes or an increase in the complexity of the sampling design. However, it is noted that the model-assisted approach was not the best across all measures used in this simulation study.

Often the sampling weights are adjusted to compensate for the coverage errors and non-responses using post-stratification, raking, and calibration. Therefore, the effects of the adjusted weights on model parameter estimations were also evaluated by post-stratified weights. It was noticed that the standard bias, variance, and the MSE of the model parameters were further reduced in model-based with weights and model-assisted methods depending on the used post-stratification variable. Hence, it is suggested to use post-stratified weights for the model-based approach and model-assisted methods to estimate AFT model parameters when the data are from a stratified random sample. However, one should be careful in selecting the post-stratification variable as it may sometimes worsen the bias of an estimate.

It is recommended to use the model-assisted approach or model-based with weights when the data is from a complex survey design when the software is available, since there is an impact for the conclusions of the analytical studies.

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