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Fitting Marginalized Two-Part Models to Semicontinuous Survey Data Arising from Complex Samples

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The data used to illustrate the proposed modeling approach is the publicly available Medical Expenditure Panel Survey Data. It is available at <https://www.meps.ahrq.gov>.

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Abstract

Objective. To accurately model semicontinuous data from complex surveys, we extend marginalized two-part models to a design-based inferential framework and provide guidance on incorporating complex sample designs.

Data Sources. 2014 Medical Expenditure Panel Survey (MEPS).

Study Design. We describe the use of pseudo-Maximum Likelihood Estimation and Jackknife Repeated Replication for estimating model parameters and sampling variance, respectively. We illustrate our approach using MEPS, modeling total healthcare expenditures in 2014 as a function of respondents' age and family income. We provide SAS and R code for implementing the extension, assessing model-fit indices and evaluating the need to incorporate complex sampling features.

Data Extraction Methods. Data obtained from www.meps.ahrq.gov.

Principle Findings. A 100 percentage-point increase in family income as a percent of the federal poverty level was associated with a 5-6% increase in healthcare spending. People over 65 had an increase of 4-5 times compared to those younger. Accounting for complex sampling in the models led to different parameter estimates and wider confidence intervals than the unweighted models. Ignoring complex sampling could lead to inaccurate finite population inference.

Conclusion. Researchers should account for complex sampling features when analyzing semicontinuous data from surveys.

Keywords: Marginalized two-part models; Complex sample survey data; Healthcare expenditures

What is known on this topic:

- Semicontinuous data are typically analyzed with marginalized or conditional two-part models

- Complex sample designs, often encountered in large national surveys, often require analytic approaches that account for these design features

What this study adds:

- Extends marginalized two-part modeling methodology to incorporate complex sampling features when analyzing semicontinuous data and demonstrates the importance of doing so
- Provides software for implementing the proposed approach

BACKGROUND

Semicontinuous data, typically characterized by a point mass of zero-valued observations paired with an often right-skewed continuous distribution of positive values, arise in many research contexts. Examples range from average daily alcohol use^{1,2} and health assessment or quality of life scores^{3,4} to annual medical expenditures,^{5,6} each of which contain a subsample with zero values combined with a positive distribution among those with a non-zero response.

Because of the two distinct data components, it is natural to analyze such data using so-called “two-part models”. Such models represent a class of modeling approaches that explicitly treat the stochastic process from which the zero values arise as distinct from the stochastic process governing the continuous positive values. Two-part models were originally proposed with a logistic or probit regression model for the probability of incurring a positive-valued response, while the second component utilized a log-link with a linear model for the continuous values conditional on them being greater than zero, referred to as the log-normal model.⁷⁻⁹ In this conditional formulation, the second component only provides inference on the subsample with positive responses. For this article, we term such models where the second component is conditional on incurring positive values as conditional two-part (CTP) models.

One limitation of CTP models is that covariate effects from the second part provide inference only on the conditionally positive subset. For inference on the entire population, the two parts must be combined through post-modeling computations. The exact form of the overall mean will depend on the model specification and the assumed distribution. The effect of a covariate on the overall mean will depend on values of the other covariates and sometimes also the reference value (e.g., the effect

of age for 40 vs. 50 years differs from the effect of age for 50 vs. 60 years). The complex relationships between marginal means and covariates inherently implies both non-linearity and heterogeneity.¹⁴

To simplify this relationship and allow for direct estimation of homogeneous covariate effects on the overall mean for the population, marginalized two-part (MTP) models have been more recently developed.^{15,16} These are similar to standard two-part models in that they explicitly model and account for the point mass at zero. They differ, however, in that the second component directly models the mean of the entire population, including both the zero and positive values as opposed to only the conditionally positive values. Therefore, the second component is an unconditional mean and covariates are directly interpreted as effects on the overall mean, without need for post-modeling computations.

Unfortunately, little work has incorporated complex sample design features into the two-part modeling framework. Semicontinuous data may be collected in complex sample surveys, which feature stratified sampling, cluster sampling, and unequal probabilities of selection for different population units. If the probabilities of selection are correlated with survey measures of interest (e.g., semicontinuous outcomes), then survey weights defined as the inverse of the probability of selection (and possibly adjusted for survey nonresponse) should be considered when estimating the CTP and MTP model parameters to ensure design-consistent estimation.²⁰ Furthermore, estimates of the sampling variance for each model parameter should also reflect the complex sampling features, as the use of cluster sampling and the incorporation of weights in estimation tends to inflate standard errors, and the use of stratified sampling tends to reduce them.²⁰ A failure to account for these complex sampling features in “design-based” analyses potentially leads to inaccurate finite population inferences about regression model parameters.²¹

To address this absence of prior work incorporating complex sampling features into the MTP modeling process, we outline the use of jackknife repeated replication for estimation of sampling variance and describe the use of pseudo-maximum likelihood estimation for estimating MTP model parameters from both the full sample and each replicate sample.²⁰ We develop software enabling a design-based approach to fitting MTP models and making inference about target populations, including estimation of sampling variability and design-based comparisons of model fit. To illustrate this extension, we model total health care expenditures as a function of respondent’s age and family income using the Medical Expenditure Panel Survey (MEPS) Data²² in 2014. Our

research is the first to provide theoretical explanations, applicable software, and empirical illustration for extending MTP modeling to a design-based inferential framework.

TWO-PART MODELS

We begin with a brief review of the two-part modeling framework. Both CTP and MTP models take the same generic likelihood form:⁷⁻⁹

$$L(\theta|y_i) = \prod_{i=1}^n [(1 - \pi_i)^{1_{(y_i=0)}} \times [\pi_i g(y_i|y_i > 0)]^{1_{(y_i>0)}}, \quad y_i \geq 0, i = 1, \dots, n$$

where θ refers to the vector of parameters defining the model, $\pi_i = \Pr(Y_i > 0)$, $1_{(\cdot)}$ is the indicator function, and $g(y_i|y_i > 0)$ is any density function applicable to the positive values of Y_i .

CTP and MTP models differ in the parameterization of the linear predictors in the second component. The CTP model is commonly parameterized as

$$\text{logit}(\pi_i) = \mathbf{z}_i' \boldsymbol{\alpha} \quad \text{and}$$

$$\mu_i = E(\ln Y_i | Y_i > 0) = \mathbf{x}_i' \boldsymbol{\lambda}.$$

In this specification, λ_k is interpreted as the effect of a one unit increase in the k th covariate on the mean of the log of the outcome conditional on the response being positive.

The MTP model specifies the linear predictors

$$\text{logit}(\pi_i) = \mathbf{z}_i' \boldsymbol{\alpha} \quad \text{and}$$

$$E(Y_i) = v_i = \exp(\mathbf{x}_i' \boldsymbol{\beta}).$$

This specification provides direct inference on the marginal combined mean of those with zero and positive values. Using linear combinations of the parameters in the second component of the model,

covariate effects on the overall marginal mean and standard errors are estimated directly from standard output. Specifically, $\exp(\beta_k)$ is interpreted as the multiplicative effect of a one-unit increase in the k th covariate on the overall mean. Under this parameterization, model-predicted marginal means and standard errors are easily obtained by estimating $\exp(\mathbf{x}_i' \boldsymbol{\beta})$ at the desired values of the covariates.

Within this class of models, various distributions can be applied to fit the data at hand. Specifically, the MTP model has been developed for a variety of flexible distributions and variance forms.¹⁵⁻¹⁸ The log-skew-normal (LSN)^{10,11} and generalized gamma (GG)^{12,13} are two flexible three-parameter distributions commonly used for highly skewed semicontinuous data; both take the simpler log-normal distribution as a special or limiting case. The details of these implementations have been reviewed elsewhere.^{14-16,18}

THE MTP MODEL: INCORPORATING COMPLEX SAMPLING FEATURES

To incorporate complex sampling features in two-part models, we follow the pseudo-maximum likelihood approach proposed by Binder.²³ A weighted estimator of the finite population likelihood for the generic MTP model is defined as follows:

$$L(\theta|y_i) = \prod_{i=1}^n \left[(1 - \pi_i)^{1(y_i=0)} \times [\pi_i g(y_i|y_i > 0)]^{1(y_i>0)} \right]^{w_i}, \quad y_i \geq 0, i = 1, \dots, n,$$

where w_i refers to the final survey weight for each responding case in the survey data set.

To estimate design-consistent sampling variance of the weighted maximum likelihood estimates, we employ Jackknife Repeated Replication (JRR). Briefly, JRR omits one sampling cluster at a time from a given sampling stratum, adjusts the survey weights for the remaining cases in that stratum, fits the MTP model of interest to that replicate sample (with adjusted weights), and then repeats this process for each of the remaining sampling clusters, saving the replicate estimates. A design-consistent sampling variance is estimated by leveraging the variance of the replicate estimates around the weighted full-sample estimate. As a general approximation, this procedure yields $a - H$

degrees of freedom, where a is the total number of sampling clusters and H is the total number of sampling strata. For additional details, see Heeringa et al. (2017).²⁰

We have developed a SAS macro (SAS Institute, Cary, NC) that incorporates complex sample features in MTP models by utilizing JRR for variance estimation and pseudo-maximum likelihood estimation for parameter estimation.²⁰ We have written R code²⁴ to adapt the Lumley and Scott²⁵ design-adjusted AIC (dAIC) and BIC (dBIC) calculations for comparison of competing models with all final survey weights and replicate weights incorporated. The SAS macro and R code are available for download in the online supplementary materials.

ILLUSTRATIVE EXAMPLE

Medical Expenditure Panel Survey (MEPS) data

We utilize the Medical Expenditure Panel Survey (MEPS) data collected in 2014 to illustrate estimation and inference for the MTP model accounting for complex sample features. MEPS data are collected from a complex, nationally representative sample of the civilian non-institutionalized population in the United States.²² The publicly available dataset provides final survey weights, along with sampling stratum (a total of 165 strata) and cluster (a total of 366 clusters) codes for each of the MEPS respondents.

We examine the association between family income as a percentage of the federal poverty line, respondent's age, and total health care charges reported in 2014, excluding prescription drugs. Approximately 22% of survey respondents indicated no health care charges in 2014, resulting in a significant portion of zero values in our outcome of interest and suggesting the need to consider two-part models.¹⁶ The unweighted mean (SD) overall expenditure per person was \$8,673 (\$40,611), while the weighted mean was \$9,880 (weighted SD=\$4,410,679). Expenditures reported ranged from \$0 to \$2,018,306. Family income as a percent of the poverty line ranged from -199% to 3,172%. Respondent's age was included as a binary variable with the value 1 indicating ages 65 and above (11% of the respondents) as of the year 2014 and the value 0 otherwise (88% of the respondents). There were 0.6% of respondents with missing age.

Analysis

To assess the association between health care expenditures and family income, we fit the following mean model:

$$\text{logit}(P(Y_i > 0)) = \alpha_0 + \alpha_1 x_{1i} + \alpha_2 x_{2i}$$

$$E(Y_i) = \exp(\beta_0 + \beta_1 x_{1i} + \beta_2 x_{2i})$$

where Y_i represents total healthcare expenditures, scaled to be measured in \$1000 increments, x_{1i} represents family income as a percent of the poverty line, scaled by 100, and x_{2i} indicates ages 65 and above. Thus, $\exp(\beta_1)$ represents the multiplicative effect of a 100 percentage point increase in poverty level on total health care expenditures, and $\exp(\beta_2)$ represents the ratio difference on total health care expenditures between the two age categories (age \geq 65 vs. age $<$ 65).

To account for the complex sample design of MEPS, estimation proceeded with a weighted likelihood, where the weight, w_i , was the person weight as provided in the MEPS dataset. For comparison, we conducted a purely model-based analysis by fixing $w_i = 1$ and ignoring the sampling strata/clusters to assess the impact of not incorporating the complex sampling features in analysis. We fit the MTP model using both approaches (fully design-based and fully model-based) and also considered three distributions (the log-normal, log-skew-normal, and generalized gamma) to assess whether the difference in model performance varied over distributional assumptions. Standard errors for the weighted models were computed with JRR. Given the 201 = (366 - 165) degrees of freedom for variance estimation, a critical t-value of 1.96 was used to form 95% confidence intervals. The importance of the weights for the parameter estimates was evaluated using the formal Wald tests outlined by Bollen and colleagues²⁶ and in Chapter 7 of Valliant and Dever,²⁷ and the fits of the three models estimated using the same approach were compared using design-adjusted AIC criteria.²⁵ All analyses were conducted in SAS 9.4 (SAS Institute, Cary, NC) and R 3.6.2.²⁴

RESULTS

Table 1 presents parameter estimates and corresponding 95% confidence intervals (CIs) from the standard model-based MTP models (“Unweighted”) next to weighted MTP models using JRR (“Weighted, JRR SEs”). The naive, unweighted models provided noticeably differing parameter estimates, particularly for the binary and overall mean intercept terms. In particular, for the log-normal model, the 95% confidence intervals for intercept terms do not even overlap. Fewer differences are noted for estimated effects of covariates (α_1 , α_2 , β_1 , and β_2). Distributional assumptions provided a noticeable change in estimates of β_0 , the overall mean intercept, as might be expected, but made very little difference in terms of the estimates of β_1 and β_2 . The JRR-estimated confidence intervals were significantly wider for the generalized gamma model than for other distributions, suggesting the GG MTP model may be somewhat more sensitive to incorporation of weights and complex sampling features.

The AIC of the naive unweighted models and the dAIC of the weighted models²⁵ are presented in Table 1. The dAIC values were larger than the unadjusted AIC values, but the AIC and dAIC ranked the three models based on the three distributional assumption in the same order (Log-skew-normal < generalized gamma < log-normal).

The need for weighted estimation was tested by performing Wald tests for the coefficients associated with three extra predictors in an unweighted MTP model – the final survey weights, the two-way interaction between the weights and the poverty predictor, and the two-way interaction between weights and the age predictor.²⁷ Under the three distributional assumptions, results of the Wald tests were $\chi^2(6) = 80.76$, $p < 0.001$ for the log-normal model, $\chi^2(6) = 84.97$, $p < 0.001$ for the log-skew-normal model, and $\chi^2(6) = 84.16$, $p < 0.001$ for the generalized gamma model. These significant results suggest that weighted estimates are needed for the current MTP models under all three distributional assumptions. In sum, we would likely employ weighted estimation of the log-skew-normal model for making inference about the marginal relationships of income and age with health care expenditures.

The models estimated that a 100 percentage point increase in family income as a percent of the federal poverty level was associated with 5-6% increase in total healthcare spending for the MEPS target population in 2014. At the weighted mean of the sample, \$9,880, a 6% increase represents a relatively small additional \$593 spent on healthcare expenditures in one year. Comparing to people who were less than 65 years old, those who were 65 and above had a 4-5 times increase in total health care spending. At the weighted mean of the sample, \$9,880, a 5-time increase represents a large additional \$49,400 on health care expenditures.

CONCLUSION

Two-part models have been growing in popularity with the ever increasing availability of electronic health record data. This expansion, combined with growing interest in interventions promising to bend the rising health care cost curve, has led to greater use of health care expenditure and utilization data that often follow semicontinuous distributions. Nonetheless, to our knowledge, design-based approaches to accounting for potentially informative complex sampling features have not previously been incorporated in the two-part modeling framework. Because ignorance of complex sampling features^{21, 28} or a failure to accommodate the point mass of zero-valued observations¹⁶ can both lead to erroneous inferences regarding MTP model parameters in finite populations, the ability to appropriately model zero-heavy outcomes collected via complex sample designs is an important advance in the use of these models.

For practice, we recommend fitting the models with and without the complex sampling features accounted for and assessing potential changes in inference. Notable changes in estimates without appreciable changes in design-adjusted SEs would suggest use of the weighted estimates for inference.²⁰ The formal Wald tests can and should be employed to compare weighted and unweighted models.^{26,27} The dAIC criteria can also be used to compare the fits with different distributional assumptions.

We provide a simple-to-use SAS macro allowing incorporation of survey weights with appropriate standard error estimation via JRR, as well as R code to evaluate appropriate fit of the weighted models. The macros incorporate any of three flexible distributional assumptions to allow for careful fitting of a variety of semicontinuous distributions. Coupled together, the illustrated extension and software provides analysts with a practical approach to appropriately conduct inference in the presence of data with many zeros arising from complex sampling.

Table 1: MTP model results for MEPS data example considering design-based and model-based approaches*

Parameter	<i>Log-normal MTP</i>		<i>Log-skew-normal MTP</i>		<i>Generalized gamma MTP</i>	
	Unweighted	Weighted, JRR SEs	Unweighted	Weighted, JRR SEs	Unweighted	Weighted, JRR SEs
α_0	0.87 (0.018) (0.84, 0.91)	1.10 (0.041) (1.02, 1.18)	0.88 (0.018) (0.84, 0.91)	1.10 (0.041) (1.02, 1.18)	0.88 (0.018) (0.84, 0.91)	1.10 (0.041) (1.02, 1.18)
α_1	0.10 (0.008) (0.09, 0.12)	0.12 (0.010) (0.10, 0.14)	0.10 (0.005) (0.09, 0.11)	0.12 (0.010) (0.10, 0.14)	0.10 (0.005) (0.09, 0.11)	0.12 (0.010) (0.10, 0.14)
α_2	1.47 (0.066) (1.34, 1.60)	1.55 (0.099) (1.35, 1.74)	1.47 (0.066) (1.34, 1.60)	1.55 (0.099) (1.35, 1.74)	1.47 (0.066) (1.34, 1.60)	1.55 (0.099) (1.35, 1.74)
β_0	1.51 (0.023) (1.46, 1.55)	1.56 (0.038) (1.48, 1.63)	1.95 (0.036) (1.88, 2.02)	1.87 (0.046) (1.78, 1.96)	2.08 (0.059) (1.96, 2.19)	1.89 (0.056) (1.78, 2.00)
β_1	0.04 (0.005) (0.03, 0.05)	0.04 (0.005) (0.03, 0.05)	0.05 (0.005) (0.04, 0.06)	0.05 (0.005) (0.04, 0.06)	0.05 (0.005) (0.04, 0.06)	0.05 (0.005) (0.04, 0.06)
β_2	1.6 (0.031) (1.54, 1.66)	1.54 (0.046) (1.45, 1.63)	1.56 (0.031) (1.50, 1.62)	1.51 (0.048) (1.42, 1.61)	1.56 (0.031) (1.50, 1.62)	1.52 (0.048) (1.42, 1.61)
AIC	171952		171270		171343	
dAIC	182729		182348		182388	

* Parameter estimate, with standard error (SE) underneath, followed by a 95% confidence interval for the parameter

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