# Bayesian Analysis of Doubly Inflated Poisson Regression for Correlated Count Data: Application to DMFT Data

# B.Gholami chaboki <sup>(1)</sup>, A. Akbarzadeh baghban <sup>(2)</sup>, T. Baghfalaki <sup>(3)</sup>, M. Khoshnevisan <sup>(4)</sup> and M. Heydarpour meymeh <sup>(5)</sup>

PhD Candidate, Department of Biostatistics, School of Allied Medical Sciences, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
 Professor, Proteomics Research center, Department of Biostatistics, School of Allied Medical Sciences, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
 Tehran, Iran.

(3) Assistant Professor, Department of Statistics, Faculty of Mathematical Sciences, Tarbiat Modares University, Tehran, Iran.

(4) Associate Professor, Preventive Dentistry Research Center, Research Institute of Dental Sciences, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

(5) PhD, English Language Department, School of Allied Medical Sciences, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

**CORRESPONDING AUTHOR**. Alireza Akbarzadeh baghban, Department of Biostatistics, School of Allied Medical Sciences, Shahid Beheshti University of Medical Sciences, Tehran, Iran.; E-mail address: akbarzad@gmail.com

**DOI: 10.2427/13224** Accepted on December 10, 2019

#### ABSTRACT

Outcome variables in clinical studies sometimes include count data with inflation in two points (usually zero and k (k>0)). Doubly inflated models can be adopted for modeling these types of data. In statistical modeling, the association among subjects due to longitudinal or cluster study designs is considered by random effects models. In this article, we proposed a doubly inflated random effects model using the Bayesian approach for correlated count data with inflation in two values, and compared this model with Bayesian zero-inflated Poisson and Bayesian Poisson models. The parameters' estimates by these models were obtained by Markov Chain Monte Carlo method using OpenBUGS software. Bayesian models were compared using the deviance information criterion. To this end, we utilized the total number of decayed, missed, and filled teeth of 12-year-old children and also conducted a simulation study. Results of real data and the simulation study revealed that the proposed model is fitted better than previous models.

Keywords: Bayesian analysis; Doubly inflated; DMFT data; Poisson regression; Random effects.

### INTRODUCTION

In clinical research, outcome variables include count data, such as the number of symptoms, number of seizures, post-surgical complications, number of hospitalizations, and number of decayed, missing, and filled teeth (DMFT). Generally, these kinds of data are not normally distributed but are mostly distributed in the Poisson, negative binomial, and binomial forms. In the Poisson distribution, unlike the normal distribution, the variance of the random variable depends on the mean, and thus the mean is equal to the variance. Count data depart from Poisson distribution due to the large frequency of farthest observation, whereas the variance is greater than the mean, and this is referred to as over-dispersion. In this case, negative binomial, generalized Poisson distribution, and also zero-inflated models are employed to analyze the data [1].

Zero-inflated Poisson (ZIP) regression is the most common model for analyzing count data with excess zeros. This model was introduced by Lambert in 1992. He assumed that, with probability P, only O is observed, and with probability 1-P, there is a Poisson ( $\lambda$ ) random variable. In this model, both probability P and mean  $\lambda$  depend on covariates with logit-link and log-link functions, respectively [2].

There may be samples in reality where the observation includes higher incidences of count zero as well as another count value, say k>0. We may call this case doubly inflated probability models. The doubly inflated Poisson (DIP) model simultaneously accommodates the count of zero, k, and other positive values. For instance, the DMFTindex which includes a count of DMFT is a major indicator for the dental health status of a person. In the Iranian National Oral Health Survey, it has been observed that the DMFT-index in children of age 12 includes a large amount of 0 and 1 [3].

Chandra (2011) introduced DIP and related regression models. She considered two DIP models,  $p_{IP}(p,\lambda)$  and  $DIP(p_1,p_2,\lambda)$ . In these models, beside the inflated zeros, there is another inflated value, k>0. The motivating examples in this study were the DMFT index in 1013 children and the patients' length of stay (LOS). Frequencies of 0 and 1 were larger in the observed DMFT count than other counts.  $DIP(p,\lambda)$ ,  $\underline{DIP}(p_1,p_2,\lambda)$ , and ZIP models have also been compared [4].

Bivariate DIP models were proposed by Pooja Sengupta et al. (2015). In this article, authors introduced a bivariate distribution for count data with the inflated count in both (0,0) and (k,k) cells for k>0. Data used in this article were taken from the Australian Health Survey. Bivariate responses included the joint count of  $y_{1}$ , the number of physician visits, and  $\mathcal{Y}_2$ , the number of medications prescribed for the patient. Cell frequencies were larger in (0,0) and (1,1) than all other cells [5]. Sumen sen et al. (2018) used Gaussian copula for modeling multivariate doubly inflated Poisson. A simulation study was used to investigate the proposed model [6]. Ishapathik Das et at. (2019) adjusted the multivariate distribution for the inflated frequencies. In the present study, the number of physician visits and also the number of prescribed medicine were used as the data and were analyzed using multivariate doubly inflated Poisson regression [7]. Joseph Mathews et al. (2019) in the book titled "Modern statistical methods for spatial and multivariate data", presented negative binomial distribution for multivariate doubly inflated count data and used Gaussian copula methods.

Assume that the count response  $\overline{y_1, y_2, ..., y_n}$  is independently distributed as  $\underline{DIP}(p_{1i}, p_{2i}, \lambda_i)$ . One approach to model a doubly inflated count response is to let W be a random variable with the following probability mass function:

$$P(W = w) = \begin{cases} p_1 \, if \, w = 0\\ p_2 \, if \, w = 1\\ p_3 \, if \, w = 2 \end{cases}$$

Where

 $(p_1 + p_2 + p_3 = 1), (p_1, p_2, p_3 \neq 0),$ 

 $(p_1 + p_2 + p_3 = 1), (p_1, p_2, p_3 \neq 0),$ 

and

(w = 0 is equal to y = 0); (w = 1 is equal to y = k);

and (w = 2 is equal to y = 0,1,2,3,...)

In this model, probabilities  $p_{1i}$  and  $p_{2i}$  can depend on covariates using the logit-link function and mean  $\overline{\lambda_i}$ can relate to covariates with the log-link function [4]. In clinical research, it is frequently observed that data are correlated due to hierarchical, multilevel, longitudinal, repeated measure, and cluster study designs. Random effects models are adopted in order to calculate the association between subjects in these studies. So far, numerous scientists have conducted studies with random effects in zero-inflated models, such as zero-inflated count model for longitudinal data by [8], multilevel zero-inflated generalized Poisson by [9], a Bayesian approach of joint models for clustered zero-inflated count data by [10], and hierarchical Bayesian analysis of correlated zero-inflated count data by [11]. Classical statistical methods such as the maximum likelihood estimate (MLE), the likelihood ratio (LR) test, and the method of moments techniques have been used for ZIP regression in multivariate, multilevel, hierarchical, longitudinal, cross-section models [9, 11, 12].

In the DIP study by Chandra (2011), parameters' estimates have been examined through maximum likelihood and the method of moments techniques. Moreover, in a study conducted by Sengupta et al (2015)., parameters' estimate has been obtained using the maximum likelihood method [4, 5].

The Bayesian approach is chosen as it overpasses the limitations of classic approaches when using asymptotic results. Also, in the Bayesian approach, unobserved heterogeneities in the data indicate simplicity [13].

The MLE method is attractive due to several desirable asymptotic properties. For example, the MLE gains an unbiased and efficient estimator (if it exists) when data are distributed normally and the sample size increases to infinity. However, these asymptotic assumptions do not necessarily hold in small samples. When the asymptotic normality of the MLE is not satisfied and/or data are uncommon such as zero-inflated or doubly inflated ones, confidence intervals by classic statistics are often misinterpreted. The



Bayesian analysis presents an alternative approach to handle these deficiencies [14].

Unlike the classic method, in the Bayesian approach, parameters are considered to be random, and a joint mass function is formed for both data and parameters. This joint function refers to the posterior distribution. The point and interval estimates of the parameters are obtained from the posterior distribution through Markov Chain Monte Carlo (MCMC) as a simulation-based method [15].

In this article, we implemented a Bayesian approach for doubly inflated correlated count data with random effects, and the parameters' estimate was achieved using the MCMC technique.

# **METHOD**

#### **Bayesian ZIP Model with a Random Effect**

Let  $y_{ij}$  be the response for the i<sup>th</sup> subject (i=1, 2,...,n) at the i<sup>th</sup> occasion (i=1, 2, ..., m), where n is the total number of subjects and m is the number of occasions. The mixture distribution of  $y_{ij}$  is:

$$y_{ij} \sim \begin{cases} 0 & \text{with probability } p_{ij} \\ poisson(\lambda_{ij}) & \text{with probability}(1-p_{ij}) \end{cases}$$
<sup>(1)</sup>

where  $\underline{p_{ij}}$  is the probability of observed zero and  $\lambda_{ij}$  is the mean of the Poisson distribution. The zero-inflated Poisson probability distribution with a random effect is equal to:

$$p(Y_{ij} = y_{ij}) = \begin{cases} p_{ij} + (1 - p_{ij}) e^{-\lambda_{ij}} y_{ij} = 0\\ (1 - p_{ij}) \frac{e^{-\lambda_{ij}} \lambda_{ij}^{y_{ij}}}{y_{ij}!}, y_{ij} = 1, 2, \dots \end{cases}$$
(2)

such that  $log(\lambda_{ij}) = X_{ij} \boldsymbol{\beta} + \boldsymbol{b}_i \otimes logit(p_{ij}) = Z_{ij} \boldsymbol{\alpha}$ 

$$\overline{\boldsymbol{\beta}} = (\beta_0, \beta_1, ..., \beta_l)^T$$
 and  $\underline{\boldsymbol{\alpha}} = (\alpha_0, \alpha_2, ..., \alpha_d)^T$  are

vectors of coefficients of  $X_{ij}$  and  $Z_{ij}$ , respectively,

and  $\overline{\boldsymbol{b}}_i$ ,  $\overline{(\boldsymbol{b}_i \sim N(0, \sigma^2))}$  is the random subject effect. Therefore, the likelihood function,

The first and most important step in the Bayesian approach is choosing appropriate prior distributions. Let  $\theta = (\beta, \alpha, \sigma^2)$  be the set of parameters for the abovementioned model. We assume independent priors for these parameters. If the joint prior distribution of parameters is  $f(\beta, \alpha, \sigma^2) = f(\beta) \cdot f(\alpha) \cdot f(\sigma^2)$  and we assume a normal distribution for  $\overline{\beta}$  and  $\alpha$  as well as an inverse gamma for  $\overline{\sigma^2}$ , then the joint prior distribution will be

$$f(\alpha,\beta,\sigma^{2}) = \prod_{k=0}^{d} \left[ \frac{1}{\sqrt{2\pi\theta_{\alpha k}^{2}}} e^{\left(\frac{-(\alpha-\mu_{\alpha k})^{2}}{2\theta_{\alpha k}^{2}}\right)} \right] \times \prod_{k=0}^{l} \left[ \frac{1}{\sqrt{2\pi\theta_{\beta k}^{2}}} e^{\left\{\frac{-(\beta-\mu_{\beta k})^{2}}{2\theta_{\beta k}^{2}}\right\}} \right] \times \left[ (\sigma^{2})^{(-a-1)} e^{\left(-b/\sigma^{2}\right)} \right] (4)$$

Where  $\beta$ ,  $\alpha \sim Normal(\mu, \theta^2)$  and  $\sigma^2 \sim IGamma(a,b)$ .

Upon multiplying the likelihood function (3) by the joint prior [4], the posterior distribution is given by:

# $f(\alpha, \beta, \sigma^2 | y, x, z) \propto L(\alpha, \beta, b_i | y, x, z). f(\alpha, \beta, \sigma^2)$

The Bayesian estimation of the parameters  $\alpha$ ,  $\beta$ , and  $\sigma^2$  requires that samples be taken from the full conditional distribution of each parameter. Unfortunately, it is not possible to directly sample from the full conditional distribution for  $\alpha$ ,  $\beta$ , and  $\sigma^2$ . As a result, a Metropolis-Hastings (M-H) algorithm in the MCMC method is utilized to generate samples within Gibbs iterations [14, 16, 17]. For the ZIP regression model, we obtained the full conditional posterior distributions for the parameters based on Equation [5] (See Appendix A).

#### 2.2 Bayesian DIP Model

#### 2.2.1 DIP Model with a Random Effect

There may come a time when the count response inflates in two points such as zero and another point (k>0). This has been introduced as the doubly inflated situation.

Suppose  $y_{ij}$  is the count response for the i<sup>th</sup> subject (i = 1, 2, ..., n) in the i<sup>th</sup> occasion  $\overline{(j = 1, 2, ..., m)}$  $y_{ij}$  with a mixture distribution is equal to:

$$y_{ij} \sim \begin{cases} 0 & \text{with probability } p_{1ij} \\ k & \text{with probability } p_{2ij} \\ \text{Poisson}(\lambda_{ij}) & \text{with probability } p_{3ij} \end{cases}$$
(5)

where

 $p_{3ij} = 1 - p_{1ij} - p_{2ij}$ 

Then, the DIP probability distribution,  $DIP(p_{1ij}, p_{2ij}, \lambda_{ij})$ , with a random effect, will be as follows:

$$P(Y_{ij} = y_{ij}) = \begin{cases} p_{1ij} + p_{3ij} \exp(-\lambda_{ij}) \text{ for } y_{ij} = 0\\ p_{2ij} + p_{3ij} \frac{\exp(-\lambda_{ij}) \lambda_{ij}^{k}}{k!} \text{ for } y_{ij} = k\\ p_{3ij} \frac{\exp(-\lambda_{ij}) \lambda_{ij}^{y_{ij}}}{y_{ij}!} \text{ for } y_{ij} = 1, 2, ..., \neq k \end{cases}$$
(6)

Probabilities  $p_{1ij}$  and  $p_{2ij}$  depend on covariates via the logit-link function and mean  $\lambda_{ij}$  formulates to covariates with the log-link function as demonstrated below:

$$\log\left(\frac{p_{1ij}}{p_{3ij}}\right) = A_{ij}\alpha; \ \log\left(\frac{p_{2ij}}{p_{3ij}}\right) = Z_{ij}\gamma; \ \log(\lambda_{ij}) = X_{ij}\beta + \mathbf{b}_i$$

in which  $\overline{A_{ij}, Z_{ij}, and X_{ij}}$  are the matrix of covariates in multinomial logistic and log-linear models, respectively,







 $\times \left[ p_{3ij} \frac{\exp(-\lambda_{ij}) \lambda_{ij}^{y_{ij}}}{y_{ij}!} \right]^{1-l(Y_{ij}=0)-l(Y_{ij}=k)}$ (8)

[4, 5, 18].

Where  $\lambda_{ij} = exp(\mathbf{X}_{ij}\beta + \mathbf{b}_i)$  and when  $fdp_{1ij} = A_{ij}\alpha$  &  $fdp_{2ij} = \mathbf{Z}_{ij}\gamma$ 

 $\lim_{t \to 0} |p_{1ij} = exp(f dp_{1ij}) / [1 + exp(f dp_{1ij}) + exp(f dp_{2ij})]$ 

$$\begin{split} p_{2ij} &= exp(fdp_{2ij}) / [1 + exp(fdp_{1ij}) + exp(fdp_{2ij})] \\ p_{3ij} &= 1 / [1 + exp(fdp_{1ij}) + exp(fdp_{2ij})] \end{split}$$

#### 2.2.2. Bayesian DIP Model with a Random Effect

According to the Bayesian approach, we need to determine the prior distribution for unknown parameters in the logit and log models. For fixed effects  $[\alpha, \gamma, \text{and } \beta]$ , we choose the normal distribution and assume an inverse gamma prior for the variance of random effect b. If  $\alpha$ ,  $\beta$ , and  $\gamma \sim Normal(\mu, \theta^2)$  and  $\sigma^2 \sim IGamma(a, b)$  and also if we assume the set of parameters for the DIP model  $\theta = (\alpha, \gamma, \beta, \sigma^2)$  to be distributed independently, then the joint prior distribution will be:  $f(\alpha, \gamma, \beta, \sigma^2)$ 

$$= \left[\frac{1}{\sqrt{2\pi\theta_{\alpha}^{2}}}e^{\left(\frac{-(\alpha-\mu_{\alpha})^{2}}{2\theta_{\alpha}^{2}}\right)}\right] \cdot \left[\frac{1}{\sqrt{2\pi\theta_{\gamma}^{2}}}e^{\left(\frac{-(\gamma-\mu_{\gamma})^{2}}{2\theta_{\gamma}^{2}}\right)}\right] \left[\frac{1}{\sqrt{2\pi\theta_{\beta}^{2}}}e^{\left(\frac{-(\beta-\mu_{\beta})^{2}}{2\theta_{\beta}^{2}}\right)}\right] \left[(\sigma^{2})^{(-a-1)}e^{\left(-b'/\sigma^{2}\right)}\right] (9)$$

Regarding the likelihood function [7] and joint prior distribution (9), the posterior distribution will be as follows:  $f(\alpha, \gamma, \beta, \sigma^2 | y, x, z, a) = L(\alpha, \gamma, \beta, \sigma^2 | y, a, z, x) \times f(\alpha, \gamma, \beta, \sigma^2)$  (10)

The mentioned posterior distribution is not computed directly. The MCMC algorithm is implemented to calculate parameters' estimates. For this purpose, we need to generate samples from the full conditional posterior distribution for the parameters  $\alpha, \beta, \gamma, \text{and } \sigma^2$  separately [14, 18, 19]. For the DIP regression model, we obtained the full conditional posterior distributions for the aforementioned parameters based on Equation [10] (See Appendix B).

#### 2.3 Bayesian Model Comparison

In this paper, we used the deviance information criterion (DIC) for determining the optimal Bayesian model. The DIC was proposed by Spiegelhalter et al. (2002) and is useful when parameters of Bayesian models have been obtained by the MCMC method. The general DIC formula is:

#### $DIC = 2E\{D(\theta)|y\} - D[\hat{\theta}] (12)$

where deviance is defined as -2 times the loglikelihood, that is,  $\underline{D(\theta)} = -2log[\underline{L(\theta)}] + c, \hat{\theta}$  is some Bayesian estimate of  $\overline{\theta}$  or posterior mean, median, or mode, and C is a fixed value [20]. For the Bayesian Poisson, ZIP, and DIP models, DIC is easily obtained by OpenBUGS where  $\underline{L(\theta)}$  is replaced with the likelihood in the mentioned models.

# SIMULATION STUDY

To establish the validity of the proposed Bayesian model, this section presents the results of a simulation study. Six series of correlated data of DIP were generated with inflatation in zero and one as the response variable. The DIP model used here had three parts: first, the probability of zero; second, the probability of one; and third, the Poisson model. Probabilities 0.6 and 0.3 and also 0.3 and 0.2 were adopted for zero and one, respectively. In the Poisson part of the DIP model, a binary, a discrete, and a time variable were considered in the log-linear model;

#### $\left|\log\left(\lambda\right)=\beta_{0}+\beta_{1}*x_{1}+\beta_{2}*x_{2}+\beta_{3}*x_{3}\right|.$

In the logistic part of DIP model, only the intercept was taken into account to keep the model simpler in order to compare with allocated probabilities. In the log-linear, the first random variable  $x_1$  was generated from a binary distribution with 0.4 probability of success; the second random variable  $\underline{x_2}$  was generated from integer values in the interval (0, 20), and the third variable was time with (1, 2, 3) values. The paired correlation for response in three times was considered to be 0.4.

Three simulation studies were performed with sample sizes of n=50, 100, and 200, and the procedure was repeated 1000 times. In each sample size the parameter values were fixed at  $\beta_0 = 1, \beta_1 = 0.4, \beta_2 = -0.1, \beta_3 = 0.6$  and probabilities of zero and one were used as mentioned above. A total of six data sets were generated; then, Poisson, ZIP, and DIP models in the Bayesian approach were fitted to these six generated data sets.

In the Bayesian DIP and Bayesian ZIP models, 60,000 updates were taken with one chain. After initial 20,000 burn-in iterations, every 15th sample was kept and finally 2666 samples were obtained. In the Bayesian Poisson model, one chain with 50,000 iterations and 10,000 burn-in samples were considered, and then every 10th sample was kept and ultimately 4000 samples were obtained. By checking the dynamic trace of Gibbs iterations and by calculating the Gelman-Rubin convergence statistic in every three models by the results of OpenBUGS software, those models were converged.

Three models were compared only based on estimation, relative bias, root mean square error (RMSE), and coverage probability of confidence interval (CP) measures of log-linear regression coefficients and

-0.15

0.63

0.5052 0.2085

0.0562 0.2338

**ES**<sup>α</sup>

-0.06

0.37

-0.08

0.54

-1.77

0.13

-0.03

0.38

0.0871

0.2465

-0.6561

-0.3719

100

49.1

real

value  $p_1 = 0.3$  $p_2 = 0.2$  $\beta_0 = 1$ 

 $\beta_1 = 0.4$ 

 $\beta_2 = -0.1$ 

 $\beta_3 = 0.6$ 

 $\substack{ \{ p_1 = 0.6 \\ p_2 = 0.3 }$ 

 $\beta_0 = 1$ 

 $\beta_1 = 0.4$ 

 $\beta_2 = -0.1$ 

 $\beta_3 = 0.6$ 

F	POISSON	1			4	ZIP		DIP						
Re.Bias <sup>ь</sup>	<b>RMSE</b> <sup>c</sup>	CP <sup>d</sup>	DICe	ES	Re.Bias	RMSE	СР	DIC	ES	Re.Bias	RMSE	СР	DIC	
-	-	-	586.5587	0.12	-0.5987 -	0.1797 -	18.2		0.24 0.16	-0.1917 -0.2103	0.0747 0.0668	-	570.9031	
1.0605	1.0606	18.5		0.2	-0.7960	0.7987	41.9	566.622	0.77	-0.2307	0.3141	89.8		
0.0825	0.5415	96.5		0.41	0.0288	0.5056	97.0		0.42	0.0504	0.3418	93.5		
0.1367	0.0470	97.5		-0.09	-0.0403	0.0424	97.3		-0.1	0.0541	0.0259	95.8		
0.0903	0.0800	76.1		0.56	-0.0590	0.0639	88.2		0.6	0.0092	0.0491	99.2		
-	-	-		0.06	-0.8075	0.2423	22.1		0.26 0.26	-0.1202 0.2952	0.1975 0.0944	-	289	
2.7726	2.7726	0	312	-1.58	-2.5838	2.5838	1.6	304.	-3.02	-4.0211	4.0526	51.5		
).6806	0.8918	94.7	.775	0.15	-0.6236	0.9031	94.8	550	-0.49	-2.2444	3.9032	94.6	388	
					1					1				

100

53.6

#### **TABLE 1. Paramete** distribution with 5

a. Estimate, b. Relative Bias, c. Root Mean Square Error, d. 95% posterior credible interval, e. Deviance Information Criteria

-0.04

0.35

-0.6089 0.0865

-0.4132 0.2606

#### TABLE 2. Parameters estimate of DIP, ZIP and Poisson Bayesian models for correlated data from generated data of DIP distribution with 100 sample size

		P	OISSON					ZIP		DIP					
real value	ES°	Re.Bias <sup>ь</sup>	RMSE	C₽d	DIC	ES	Re.Bias	RMSE	СР	DIC	ES	Re.Bias	RMSE	СР	DIC
$p_1 = 0.3$ $p_2 = 0.2$	-		-		_	0.13	-0.5546	0.1665	5.4	_	0.28 0.18	-0.0529 -0.0754	0.0374 0.0338	-	1145.2265
$\beta_0 = 1$	-0.1	-1.1031	1.1032	1.4	159	0.18	-0.8154	0.8154	13.6	123.7188	0.91	-0.0932	0.1645	92.9	
$\beta_1 = 0.4$	0.33	-0.1701	0.3698	96.7	.98	0.38	-0.0457	0.3396	96.5		0.41	0.0259	0.1949	93.4	
$\beta_2 = -0.1$	-0.08	-0.1871	0.0330	99.2	32	-0.09	-0.08	0.0287	98.6		-0.1	0.0341	0.0148	95.8	
$\beta_3 = 0.6$	0.56	-0.073	0.0636	73.1	]	0.57	-0.0474	0.0489	88.5		0.61	0.0176	0.0388	98.1	
	-	-	-	-	6	0.07	-0.8802	0.5281	-	6	0.33 0.26	-0.4477 -0.1183	0.2738 0.0523		<b>с</b> л
$\beta_0 = 1$	-1.68	-2.6774	2.6774	0	20	-1.49	-2.4936	2.4936	0	80	-1.71	-2.7145	2.7601	46.4	84
$\beta_1 = 0.4$	0.14	-0.6462	0.5943	94.8	.97	0.16	-0.6044	0.6017	95.2	02	0.15	-0.6354	1.2981	94.2	.8756
$\beta_2 = -0.1$	-0.03	-0.6826	0.0750	100	93	-0.04	-0.6389	0.0725	100	62	-0.09	-0.1221	0.089	98.0	
$\beta_3 = 0.6$	0.36	-0.4004	0.2439	26.3		0.34	-0.4338	0.2618	24.4		0.57	-0.0469	0.1188	96.3	

a. Estimate, b. Relative Bias, c. Root Mean Square Error, d. 95% posterior credible interval, e. Deviance Information Criteria

99.1

93.5

		Р	OISSON					ZIP			DIP					
real value	ES°	Re.Bias <sup>ь</sup>	RMSE	CPd	DIC	ES	Re.Bias	RMSE	СР	DIC	ES	Re.Bias	RMSE	СР	DIC	
$p_1 = 0.3$ $p_2 = 0.2$	-	-	-			0.13	-0.5601	0.1679 -	0.7		0.29 0.19	-0.0287 -0.0578	0.0258 0.0236	-	2271.031	
$\beta_0 = 1$	-0.08	-1.0782	1.0782	0.1	2270	0.19	-0.8052	0.8052	2.3	2210	0.94	-0.063	0.103	95.4		
$\beta_1 = 0.4$	0.32	-0.2066	0.2504	94.8	0.80	0.36	-0.1	0.2286	94.8	0.52	0.41	0.0128	0.1124	94.3		
$\beta_2 = -0.1$	-0.08	-0.1971	0.0269	99.7		-0.09	-0.095	0.0217	98.8	3	-0.1	0.0312	0.0092	96.5		
$\beta_3 = 0.6$	0.55	-0.0812	0.0552	62.2		0.57	-0.0519	0.0401	83.6		0.61	0.0193	0.0275	97.6		
$ \begin{cases} p_1 = 0.6 \\ p_2 = 0.3 \end{cases} $	-	-	-	-		0.08	-0.8542	0.5125	0	_	0.38 0.26	-0.3687 -0.1282	0.2296 0.0475	-	1174.4018	
$\beta_0 = 1$	-1.64	-2.642	2.642	0	122	-1.45	-2.4453	2.4453	0	204	-1.12	-2.1246	2.1765	44.2		
$\beta_1 = 0.4$	0.14	-0.6624	0.4111	93.7	4.	0.16	-0.6036	0.4059	94.3	.5]	0.32	-0.1949	0.5299	96.2		
$\beta_2 = -0.1$	-0.03	-0.6733	0.0692	100	] _	-0.04	-0.6231	0.0651	100	4	-0.08	-0.1625	0.0495	97.5		
$\beta_3 = 0.6$	0.37	-0.392	0.2354	8.3		0.34	-0.4279	0.2567	5.8		0.57	-0.0476	0.0871	94		

# TABLE 3. Parameters estimate of DIP, ZIP and Poisson Bayesian models for correlated data from generated data of DIP distribution with 200 sample size

a. Estimate, b. Relative Bias, c. Root Mean Square Error, d. 95% posterior credible interval, e. Deviance Information Criteria

probabilities. These measures equal to:

Relative Bias = 
$$\frac{1}{N} \sum_{k=1}^{N} \frac{\left(\hat{\theta}_k - \theta\right)}{\theta}$$

Where N is the number of repetitions  $\theta$  is the real measure of parameter and  $\underline{\hat{\theta}}_k$  is parameter estimation in the kth repetition.

$$RMSE = \sqrt{\frac{1}{N}\sum_{k=1}^{N} (\hat{\theta}_{K} - \theta)^{2}}$$

Among all the iterations, CP is the total number of times that the 2.5 percentile is smaller than the parameter estimate and the 97.5 percentile is larger than the parameter estimate; in other words, the number of times that Bayesian credible intervals include parameters' estimate. Different values of DIC were obtained for every model in order to choose the best-fitted model.

The results of estimation, relative bias, RMSE, CP, and DIC related to the parameters of DIP, ZIP, and Poisson Bayesian models for the generated data of DIP model with two probabilities of zero and one in three sample size are presented in Tables 1, 2, and 3. The results presented in Table 1 with sample size equal to 50 indicate that when zero and one contain half of the generated data,  $\overline{p_1 = 0.3}, p_2 = 0.2$ , parameters' estimates in the Poisson part of the Bayesian DIP model are closer to real values rather than in the Bayesian ZIP and Poisson models. Nevertheless, the DIC gained from all three models suggests that the Bayesian ZIP model is better fitted to these data. In the second part of Table 1, when zero and one contain 90% of the generated data,  $p_1 = 0.6$ ,  $p_2 = 0.3$ , parameters' estimate for some parameters in all three models are not near real values, but the value of DIC attained from the Bayesian DIP model is smaller than that of the other two models.

The results of DIP, ZIP, and Poisson Bayesian models for the data generated from the DIP model with 100 samples and two sets of probabilities for zero and one are given in Table 2. When  $p_1 = 0.3$ ,  $p_2 = 0.2$ , the DIC in three models shows that the Bayesian ZIP model is better fitted to these data, while the parameters' estimate in the DIP model are closer to real values than the other two models. When  $p_1 = 0.6$ ,  $p_2 = 0.3$ , parameters' estimate and DIC scale in three models show that the Bayesian DIP model is superior to the others; also, compared to its similar case in Table 1, the Bayesian DIP model is better fitted.

Table 3 includes the results of DIP, ZIP, and Poisson Bayesian models for data generated of the DIP model with 200 samples in two sets of probabilities for zero and one. Parameters' estimate in the DIP model are closer to real values when  $p_1 = 0.3$  and  $p_2 = 0.2$  and also  $p_1 = 0.6$  and  $p_2 = 0.3$ . Both relative bias and RMSE are smaller in the DIP model. In this model, the number of times that credible intervals include parameters' estimate is larger than that of other models. The DIC measure is smaller than the DICs of other models when  $p_1 = 0.6$  and  $p_2 = 0.3$  for the DIP model.





### **APPLICATION TO REAL DATA**

The basic oral health surveys supply an accurate basis for evaluating the present oral health status of any given population and its future oral health care needs. The WHO manual of oral health survey has encouraged countries to conduct standardized oral health surveys that are comparable internationally [21].

The last Iranian National Oral Health Survey was performed in 2012. In this survey, five age groups of 5-6, 12, 15, 34-54, and 65-74 years were considered. The examined indexes in children and adults were decayed teeth, gum disorders, dental trauma, and so on. A self-report questionnaire and oral examination were used for collecting information on demography, socioeconomic status, habits, and individual care level. Sample size was considered 300 people of each age group in each province of Iran based on WHO proposal. One of the pieces of information gathered in this survey was the total number of DMFT. Information about this survey conducted in 2012 showed that the DMFT index among 12-year-old children was inflated in zero and one. In other words, the DMFT index in 12-year-old children was doubly inflated. This feature is true in all populations investigated in Iran [3].

A part of the information from the National Oral Health Survey 2012 from Hamedan Province was utilized in this study. The purpose of this paper was to investigate the influence of different personal features such as the place of residence and eating habits, especially sugar, on the number of DMFT of 12-year-old children in Hamedan. After oral examination, the DMFT index was determined for all dentitions located in the right and left sides of the maxilla and mandible. Inflation was seen in zero and one in this data. The inflation of zero and one towards other counts can be observed in Figure 1. The frequency of zero is almost 60% and the frequency of one is nearly 30% in all dentitions.

In Table 4, the posterior summary of DIP, ZIP, and Poisson model on Hamedan DMFT data is provided.

		ZERO P	ART OF	MODEL			ONE P	ART OF	MODEL		POISSON PART OF MODEL					
	mean	SD	2.5%PC	median	97.5%PC	mean	SD	2.5%PC	median	97.5%PC	mean	SD	2.5%PC	median	97.5%PC	
Poisson																
Intercept	-	-	-	-	-	-	-	-	-	-	-1.34*	0.37	-2.11	-1.34	-0.63	
Sugar Score	-	-	-	-	-	-	-	-	-	-	0.08	0.07	-0.05	0.08	0.23	_
Sex	-	-	-	-	-	-	-	-	-	-	-0.21	0.15	-0.5	-0.21	0.07	1959
Resident	-	-	-	-	-	-	-	-	-	-	-0.09	0.15	-0.39	-0.09	0.21	
tau	-	-	-	-	-	-	-	-	-	-	1.09*	0.19	0.76	1.08	1.5	
ZIP																
Intercept	-9.79	30.32	-69.65	-9.76	50.12	-	-	-	-	-	-1.33*	0.37	-2.06	-1.32	-0.63	
Sugar Score	-29.0	19.39	-72.56	-26.33	0.93	-	-	-	-	-	0.08	0.07	-0.06	0.08	0.22	_
Sex	-5.7	30.01	-63.06	-5.42	53.06	-	-	-	-	-	-0.22	0.15	-0.5	-0.22	0.076	195
Resident	-6.42	28.22	-63.87	-6.27	47.58	-	-	-	-	-	-0.09*	0.15	-0.38	-0.09	-0.2	8
tau	-	-	-	-	-	-	-	-	-	-	1.08*	0.19	0.76	1.07	1.51	
DIP																
Intercept	-6.92	30.51	-65.44	-6.71	53.36	-2.39*	0.72	-3.89	-2.36	-1.08	-1.85*	0.58	-3.02	-1.83	-0.74	
Sugar Score	-29.8*	19.9	-74.13	-27.26	-0.31	0.16	0.14	-0.11	0.16	0.44	0.08	0.11	-0.13	0.08	0.3	
Sex	-7.48	29.45	-66.91	-6.08	49.79	-0.23	0.32	-0.87	-0.23	0.39	-0.22	0.24	-0.69	-0.22	0.24	681
Resident	-7.05	29.25	-66.27	-6.92	51.27	0.08	0.32	-0.58	0.09	0.69	-0.21	0.24	-0.71	-0.2	0.26	ω
tau	-	-	-	-	-	-	-	-	-	-	0.55*	0.12	0.35	0.54	0.82	

# TABLE 4. Parameter estimation of Bayesian DIP, ZIP and Poisson model for DMFT

These models include three independent variables of sex (male=1, female=0), place of residence (urban=1, rural=0) and sugar score. The number of subjects was 300 children of 12 years of age. These variables were considered in the Poisson model, Poisson part, and zero part of the ZIP model and in the Poisson part, zero part, and one part of the DIP model.

In the Bayesian DIP model, 160000 updates were taken with two parallel chains. A burn-in sample of 25000 was used and then every 100th sample was kept, until 2700 observations were obtained from two chains. In the Bayesian ZIP model we ran two chains of 60000 iterations with the first 20000 discarded as burn-in, and then every 20th sample was kept. Ultimately, a total of 4000 samples were obtained from the two chains. In the Poisson model we took 30000 updates with two parallel chains. After the initial 10000 burn-in iterations, 2000 samples with thinning 10 were obtained from each chain; in total, 4000 samples were obtained. Using the output of OpenBUGS software, convergence was evaluated visually through monitoring the dynamic traces of Gibbs iterations and computing the Gelman-Rubin convergence statistic.

A summary of the posterior parameters of three Bayesian models is presented in Table 4. This table includes the posterior mean, standard deviation (SD), 2.5 percentile, median, and 97.5 percentile. It should be noted that 2.5 and 97.5 percentiles provide equal tails for 95% posterior interval parameters' estimate. As demonstrated in Table 4, the DIC for the Bayesian DIP model is smaller than that for the Bayesian ZIP and Poisson models. Since the model with the smallest DIC is the best-fitted model, the Bayesian DIP model was the best fitted to the present data. In the Poisson part of the ZIP model, the place of residence was statistically significant, which means that children living in urban areas tend to have lower DMFT compared to their rural-living counterparts. In the zero part of the Bayesian DIP model, the sugar score was significant, meaning that the children with a high sugar score had a very low chance for taking zero in DMFT. Parameters of the random effect in all three models were significant.

# DISCUSSION

Doubly inflated models are useful for modeling the outcomes of count data with many zeros and many other counts such as k (k>0). Chandra (2011) introduced two DIP models for data analysis. She estimated parameters using two techniques, including maximum likelihood and the method of moments. Efficiency comparisons revealed that ML estimators can perform better than moment estimators for both models. [4].

Sengupta et al. (2015) developed two bivariate DIP models to develop paired count data with inflated frequencies. They demonstrated that maximum likelihood estimators were relatively more efficient than moment estimators [5].

Sumen Sen et al. (2018) obtained efficiency and favourable outcome in the modeling of the doubly inflated Poisson count data [6]. Ishapathik Das et al. (2019) found that Poisson regression model considering multivariate doubly inflated, provides better fit than the model ignoring inflated data [7].

In this study, a Bayesian doubly inflated random effects model was proposed for correlated count data with inflated frequencies at both zero and one. The motivated data were the total number of DMFT. Three models of Bayesian Poisson, ZIP, and DIP with random effects were compared on DMFT data using DIC. This criterion showed that the Bayesian DIP was better fitted than the other two models.

A simulation was also performed to compare the three models. The correlated count data of DIP model were generated with inflation in zero and one, and Bayesian Poisson, Bayesian ZIP, and Bayesian DIP models were fitted to this data. The results of the simulation study with  $p_1 = 0.3$  and  $p_2 = 0.2$  indicated that the parameters' estimates of the DIP model in each of three sample sizes, p = 50,100 and 200, were closer to real values than the other two models. On the other hand, values of DIC showed that the ZIP model fitted these data better than the other models. Given that the probability of zero was 0.3 and the probability of one was 0.2, it could be possible that these data were inflated on zero only, and that is why the zero-inflated model performed better.

The data generated on the DIP model with  $p_1 = 0.6$  and  $p_2 = 0.3$  were fitted in three models. Thus, it can be stated that, by increasing the sample size, parameters' estimates related to the zero and one probability in the doubly inflated model were closer to real values in comparison to that of the other two models. The value of DIC in the DIP model was smaller than the DICs of other models in each of the three sample sizes. As the probability of zero and one equaled 0.6 and 0.3, respectively, the generated data could be certainly doubly inflated. Since the proportion in other count data except for zero and one was 10%, the parameter estimate of the log-linear model (Poisson part) in Tables 1, 2, and 3 was far from the real value.

# CONCLUSION

It can be concluded that, in count data which are doubly inflated in zero and k (k>0), if zero and k contain more than half of the frequency of data, then the DIP model better fits the data.

# **APPENDIX A**

Full Conditional Posterior Distribution for the ZIP Regression Model



 $f(\boldsymbol{\beta}|\boldsymbol{\alpha}, \boldsymbol{\sigma}^2, \boldsymbol{x}, \boldsymbol{z}, \boldsymbol{y})$ 



$$f(\sigma^2|\alpha,\beta,x,z,y) \propto \prod_{i=1}^n \left\{ \int_{-\infty}^{+\infty} \prod_{j=1}^m \right\}$$

$$\left[\frac{e^{z_{ij}^{\alpha}}}{1+e^{z_{ij}^{\alpha}}}+\frac{1}{1+e^{z_{ij}^{\alpha}}}e^{-e^{x_{ij}^{\beta+b}i}}\right]^{I(y_{ij}=0)} \times$$

$$\left[\frac{e^{-e^{x_{ij}^{'}\beta+b_i}}\cdot e^{x_{ij}^{'}\beta y_{ij}+b_i y_{ij}}}{y_{ij!}}\right]^{1-I(y_{ij}=0)}\phi(b_i)db_i$$

$$\times \left[ (\sigma^2)^{(-a-1)} \; e^{\left( -^b / \! \sigma^2 \right)} \right]$$

#### **APPENDIX B**

Full Conditional Posterior Distribution for the DIP Regression Model

$$f(\boldsymbol{\alpha}|\boldsymbol{\gamma},\boldsymbol{\beta},\boldsymbol{\sigma}^{2},\boldsymbol{y},\boldsymbol{x},\boldsymbol{z},\boldsymbol{a}) = \prod_{i=1}^{n} \left\{ \int_{-\infty}^{+\infty} \prod_{j=1}^{m} [p_{1ij} + p_{3ij} \exp(-\lambda_{ij})]^{I(Y_{ij}=0)} \right\}$$

$$\times \left[ p_{2ij} + p_{3ij} \frac{\exp(-\lambda_{ij})\lambda_{ij}^{k}}{k!} \right]^{I(Y_{ij}=k)} \times \\ \left[ p_{3ij} \right]^{1-I(Y_{ij}=0)-I(Y_{ij}=k)} \phi(b_{i}) \ db_{i} \right\} \times \\ \left[ \frac{1}{\sqrt{2\pi\theta_{\alpha}^{2}}} e^{\left(\frac{-(\alpha-\mu_{\alpha})^{2}}{2\ \theta_{\alpha}^{2}}\right)} \right]$$

$$\begin{aligned} &f(\beta \mid \alpha, \gamma, \sigma^{2}, y, x, z, a) = \\ &\prod_{i=1}^{n} \left\{ \int_{-\infty}^{+\infty} \prod_{j=1}^{m} [p_{1ij} + p_{3ij} \exp(-\lambda_{ij})]^{I(Y_{ij}=0)} \\ &\times \left[ p_{2ij} + p_{3ij} \frac{\exp(-\lambda_{ij})\lambda_{ij}^{k}}{k!} \right]^{I(Y_{ij}=k)} \\ &\times \left[ \frac{\exp(-\lambda_{ij})\lambda_{ij}^{y_{ij}}}{y_{ij}!} \right]^{1-I(Y_{ij}=0)-I(Y_{ij}=k)} \phi(b_{i}) \ db_{i} \right\} \times \left[ \frac{1}{\sqrt{2\pi\theta_{\beta}^{2}}} e^{\left\{ \frac{-(\beta-\mu_{\beta})^{2}}{2\theta_{\beta}^{2}} \right\}} \end{aligned}$$

$$\begin{aligned} & \left[ f(\boldsymbol{\gamma} | \boldsymbol{\alpha}, \boldsymbol{\beta}, \sigma^{2} \boldsymbol{y}, \boldsymbol{x}, \boldsymbol{z}, \boldsymbol{a}) \right]^{I} \left[ \boldsymbol{\gamma}_{i=1}^{+\infty} \prod_{j=1}^{m} \left[ \boldsymbol{p}_{1ij} + \boldsymbol{p}_{3ij} \exp(-\lambda_{ij}) \right]^{I\left(Y_{ij}=0\right)} \\ & \times \left[ \boldsymbol{p}_{2ij} + \boldsymbol{p}_{3ij} \frac{\exp(-\lambda_{ij})\lambda_{ij}^{k}}{k!} \right]^{I\left(Y_{ij}=k\right)} \\ & \times \left[ \frac{\exp(-\lambda_{ij})\lambda_{ij}^{y_{ij}}}{y_{ij}!} \right]^{1-I\left(Y_{ij}=0\right)-I\left(Y_{ij}=k\right)} \phi(b_{i}) \ db_{i} \right] \times \\ & \left[ \frac{1}{\sqrt{2\pi\theta_{Y}^{2}}} e^{\left(\frac{-(\gamma-\mu\gamma)^{2}}{2 \ \theta_{Y}^{2}}\right)} \right] \end{aligned}$$

$$f(\sigma^{2}|\alpha,\gamma,\beta,y,x,z,a)$$

$$\prod_{i=1}^{n} \left\{ \int_{-\infty}^{+\infty} \prod_{j=1}^{m} \left[ p_{1ij} + p_{3ij} \exp(-\lambda_{ij}) \right]^{I(Y_{ij}=0)} \times \left[ p_{2ij} + p_{3ij} \frac{\exp(-\lambda_{ij})\lambda_{ij}^{k}}{k!} \right]^{I(Y_{ij}=k)} \times \right\}$$

$$\left[\frac{\exp(-\lambda_{ij})\lambda_{ij}^{y_{ij}}}{y_{ij}!}\right]^{1-l(Y_{ij}=0)-l(Y_{ij}=k)}\phi(b_i)\ db_i\right\}\times$$

 $\mathbf{k}!$ 

 $\left[ (\sigma^2)^{(-a-1)} e^{\left(-b/_{\sigma^2}\right)} \right]$ 

#### References

- 1. Hu M-C, Pavlicova M, Nunes EV. Zero-inflated and Hurdle Models of Count Data with Extra Zeros: Examples from an HIV-Risk Reduction Intervention Trial. NIH Public Access. 2011;37(5):367-75.
- Lambert D. Zero-Inflated Poisson Regression, With an Application to 2. Defects in Manufacturing. TECHNOMETRICS. 1992;34(1):1-14.
- Khoshnevisan MH. Oral Health Status in Iran-2012(INOHS-2012). 3. Oral Health Bureau, Ministry of Health and Medical Education.20132013.
- 4. Sheth-Chandra M. The doubly inflated Poisson and related regression models: Old Dominion University; 2011.
- 5. Pooja Sengupta N, Chaganty R, Sabo RT. Bivariate Doubly Inflated Poisson Models with Applications. Journal of Statistical Theory and Practice. 2015;Online:1-14.
- 6. Sen S, Sengupta P, Diawara N. Doubly inflated Poisson model using Gaussian copula. Communications in Statistics-Theory and Methods. 2018;47(12):2848-58.
- Das I, Sen S, Chaganty NR, Sengupta P. Regression for doubly 7. inflated multivariate Poisson distributions. Journal of Statistical Computation and Simulation. 2019:1-13.
- Zhu H, Luo S, DeSantis SM. Zero-inflated count models for 8. longitudinal measurements with heterogeneous random effects. Statistical methods in medical research. 2017;26(4):1774-86.
- 9. Almasi A, Eshraghian MR, Moghimbeigic A, Rahimib A, Mohammadb K, Fallahigilan S. Multilevel zero-inflated Generalized Poisson regression modeling for dispersed correlated count data. Statistical Methodology. 2015;30:1-14.
- 10. Fu Y-z, Chu P-x, Lu L-y. A Bayesian approach of joint models for clustered zero-inflated count data with skewness and measurement errors. Journal of Applied Statistics. 2015;42(4):745-61.
- 11. Dagne GA. Hierarchical Bayesian Analysis of Correlated Zeroinflated Count Data. Biometrical Journal. 2004;46( 6):653-63.
- 12. Wan W-Y, Chan JSK. A New Approach for Handling Longitudinal Count Data with Zero-Inflation and Overdispersion: Poisson Geometric Process Model. Biometrical. 2009;51(4):556-70.
- 13. Rodrigues J. Bayesian Analysis of Zero-Inflated Distributions. Communications in Statistics-Theory and Methods. 2007; 32(2):281-9
- 14. LIU H, POWERS DA. BAYESIAN INFERENCE FOR ZERO-INFLATED POISSON REGRESSION MODELS. Journal of Statistics: Advances in Theory and Applications. 2012;7(2):155-88.
- 15. Ghosh SK, Mukhopadhyay P, Lu J-CJ. Bayesian analysis of zeroinflated regression models. Journal of Statistical Planning and Inference. 2006;136:1360 - 75.
- 16. Buu A, Li R, Tan X, Zuckera RA. Statistical models for longitudinal zero-inflated count data with applications to the substance abuse field. statistics in medicine. 2012:1-13.
- 17. Ghosh P, Albert JM. A Bayesian analysis for longitudinal semicontinuous data with an application to an acupuncture clinical trial. Computational Statistics and Data Analysis. 2009;53:699 706.
- 18. Dagne GA. Bayesian semiparametric zero-inflated Poisson model for longitudinal count data. Mathematical Biosciences. 2010:224:126-13-.
- 19. Gelman A. Prior distributions for variance parameters in hierarchical models. Bayesian Analysis. 2006;1(3):515-33.

\*

- 20. Johnson W, Branscum A, Hanson TE, Christensen R. Bayesian ideas and data analysis: an introduction for scientists and statisticians. 2010.
- Organization WH. Oral health surveys: basic methods: World Health Organization; 2013.

ebph