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Application of hurdle model with random effects for evaluating the balance improvement in stroke patients

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Abstract

Background: Stroke is a prevalent cause of disability in adults. The fall is the most common balance and motor impairments, which affects the quality of life in stroke patients. This study aims to employ random effects hurdle model for evaluating the balance improvement in stroke patients under the occupational therapy.

Methods: In this longitudinal study with repeated measurement during one year between 2013 and 2014, the data was collected using non-random sampling method from three occupational therapy clinics. For a total of 38 stroke patients, the number of falls was recorded every two weeks. The random effects hurdle model and random effects zero inflated Poisson (ZIP) model were fitted to the data and were compared together. Data analysis was carried out using SAS Software version 9.2.

Results: The results of random effects ZIP model showed that the covariates of sex and age and affected side of stroke and follow up duration had statistically significant effect on balance improvement ($p< 0.05$). The occupational therapy has been effective on balance improvement more than 40% during one year.

Conclusion: The ZIP model with random effects can capture zero inflation and correlation structure in longitudinal count data simultaneously. Older patients, women and patients with left-side impairments were more at risk of fall and balance impairment, so they need more care and therapy.

Keywords: Stroke, Balance, Occupational therapy.

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Introduction

Stroke is a prevalent cause of disability and the most common disease in adults, older than 65years (1, 2). The incidence of stroke is between 100 to 300 per 100,000 people in Western countries. Based on studies in two past decades in Iran, the annual incidence of stroke in various ages ranged from 23 to 103 per 100,000 population (3, 4).

The stroke is interruption of blood flow due to bleeding or blockages. The stroke leads to symptoms such as deficiencies in consciousness, memory, cognition, sensation and movement, paralysis or weakness in one side of body, muscles atrophy and physical control problem(5). Two main types of stroke are ischemic stroke (embolic or thrombotic) or hemorrhagic stroke (bleeding)(6). Because of the balance and motor function impairments, the stroke patients would beat higher risk of fall. Thus, assessing the fall status is important among the elderly people. Also with study on peo-

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ple who are more at risk of fall, we can realize the amount of required care and treatment for patients (7-11). In general, the treatment of stroke patients includes three stages: acute phase, sub-acute phase and chronic phase (12). In the acute phase, the treatment usually includes of stabilizing the patient's clinical status and elimination of disease risk factors (13). In the sub-acute and chronic phases, the treatments are based on standard motor rehabilitation and intensive motor rehabilitation in order to increase physical abilities and reduce mortality rate (14). This study focused on occupational therapy which works with the intensive motor rehabilitation.

In many medical studies, the data is abundant (15-17). A problem that may lead to poor fit and unreliable results is accumulation of zero in count data. In this case, that suggests the use of two part models or the zero inflated models such as hurdle model or ZIP model. On the other hand, statistical modeling is the best approach for evaluation of risk factors in medical research. Also choosing the proper model is important. In this study, the hurdle model and ZIP model were employed. The Hurdle model has been introduced by CRAGG in 1971 (18) and reviewed by MULLAHY in 1986 (19). ZIP model has been introduced by Lambert in 1992 (20). One difference between these two types of model is that the hurdle model use to over dispersion and under dispersion situation, while ZIP model use to over dispersion only. Also, hurdle model has better fit in population with large sample size for longitudinal count data (21). Hurdle model and ZIP model have been developed, and different types of such models have been used in different studies (22- 24).

Many studies have been performed to investigate the status or prediction of fall in stroke patients such as: The Study that was determined the risk factors of falls (BAETENS and et al, 2011) (7), The longitudinal study on stroke patients with fear of falling (SCHEMID and et al, 2011) (9), a Longitudinal Study that investigated the prediction of Fall risk six weeks from onset of stroke (Nystrom and et al, 2013) (25), The study that predicted the chance of falling in patients with stroke using the Berg Balance index (Maedaandet al, 2009) (26), The study that evaluated effectiveness of occupational therapy on motor function in stroke patients (DAIVAand et al, 2008) (27). Another study was conducted to predict the risk factors of falls in patient after stroke (28-30). None of these studies used zero inflated model to analyze relation between balance and stroke characteristics, but they used a Poisson, negative binomial model, or other models (31,32).

The aim of this longitudinal study was to evaluate possible effective factors on balance improvement in stroke patients and also to predict the balance and motor improvement under the occupational therapy using hurdle model. We also compared this model with zero inflated Poisson model.

Methods

Participants and data

This longitudinal study was done for a period of one year between 2013 and 2014, using non-random sampling method at the Occupational Therapy Clinic at the School of Rehabilitation Sciences and the Rehabilitation department of SHOHADA Hospital and Ayatollah TALEGHANI Hospital in Tehran. Samples were taken from stroke patients (ischemic and hemorrhagic), complained of muscle spasticity and weakness or paralysis in one side of their body. Diagnosis of stroke was done according to medical records prepared by specialist doctors. In total, 38 patients were included. Demographic variables included in are as follows: sex, age, stroke duration (less than 6 months/ more than 6 months), side of stroke (right /left), stroke type (ischemic /hemorrhagic) and number of treatment sessions (between 20 to 50/more than 50). Number of falls was also recorded via interview every two weeks. Reevaluation was performed at the end of each visit. Follow-up times were different in patients due to inaccessibility to some of the patients dur-

ing study. Informed consent was taken from each patient. According to the zero inflated structure of the outcome variable with cluster on the subjects, the hurdle model and ZIP model with random effects were employed. These models are two part models that control both zero inflation and correlation structure in count data. These models were compared using goodness of fit indexes such as AIC, BIC and chi square. Data analysis was performed using SAS ver. 9.2 software. The significance was defined as $p< 0.05$.

Statistical models

The hurdle model (Cragg, 1971) and zero inflated Poisson model or ZIP (Lambert, 1992) deal with high occurrence of zeros in observed data and include two parts of parameters: one for zero inflation and the other for count data. These two models have one main distinction in how they analyze zero counts.

Let response y_{ij} denote the count for i^{th} subject at time j, i=1,.. , N, j=1,.., T. the probability of zero inflation and the mean of count data in Poisson model are denoted by π_{ij} , $0\leq \pi_{ij}\leq 1$ and μ respectively. The random effects parameter of θ is included in model because of longitudinal structure of data.

Hurdle model:

$$P\left(Y_{ij}=y_{ij}|\pi_{ij},\theta\right)=\begin{cases}\pi_{ij}, & y_{ij}=0;\\ \dfrac{\left(1-\pi_{ij}\right)e^{-\mu_{ij}}\mu_{ij}^{y_{ij}}}{\left(1-e^{-\mu_{ij}}\right)y_{ij}!}, & y_{ij}>0,\end{cases}$$

ZIP model:

$$P\left(Y_{ij}=y_{ij}|\pi_{ij},\theta\right)=\begin{cases}\pi_{ij}+\left(1-\pi_{ij}\right)e^{-\mu_{ij}}, & y_{ij}=0;\\ \dfrac{\left(1-\pi_{ij}\right)e^{-\mu_{ij}}\mu_{ij}^{y_{ij}}}{y_{ij}!}, & y_{ij}>0,\end{cases}$$

In both models, the Zero part analyzes as a logistic model with logit link function. In the other part which modeling with log link function, the count data in ZIP model followed of Poisson distribution whereas in the hurdle model followed of truncated Poisson distribution. In fact, the hurdle model considers all of zeros as zero inflation, while the ZIP model assumes zero inflation is mixture of zeros that occur with Poisson distribution and excess zeros [33]. In this paper, we used these models using SAS software to fit the model and estimate the parameters.

Results

In total, 38 patients (44.7% females and 55.3% males) were studied. The range of age was between 24 to 83 years, and the mean and standard deviation of age were 56.6 and 14.66 years, respectively. The characteristics of the stroke patients and also, the fall frequency percentage has been presented in Table 1. The fall percentage among stroke patients was as follows: 17.2% fell only once, 3 % twice and 1.3 % fell three times and 78.4 % of sample had no experience of fall that indicates zero inflation.

At first, the Poisson model as a general count model was fitted to data. This model can be used for small range of count data or

Table1. The frequency distribution of covariates and outcome variable (fall).

Parameters	Category	n	%	N (Fall)	% (Fall)
Sex	Male	21	55.3	9	14.8
	Female	17	44.7	20	27.3
Stroke type	Hemorrhagic	26	68.4	21	22.4
	Ischemic	12	31.6	8	20
Stroke duration	Less than 6 months(acute phase)	11	28.9	7	17.5
	More than 6 months(chronic phase)	27	71.1	22	23.3
Side of stroke	Left	11	28.9	10	25
	Right	27	71.1	19	20.2
Amount of treatment sessions	Between 20 and 50	27	71.1	19	22.9
	More than 50	11	28.9	10	20.4

Table 2. The results of zero inflation part of the random effects ZIP model to evaluate balance and motor improvement in stroke patients

Variable	Category	Estimate	Standard Error	OR	p
Age	-	-0.057	0.0002	0.94	0.023**
Sex	Male*	-0.037	0.0001	0.96	0.018**
	Female				
Stroke type	Hemorrhagic*	0.043	1.112	1.04	0.969
	Ischemic				
Stroke duration	Less than 6 months(acute phase)*	0.004	1.434	1	0.998
	More than 6 months(chronic phase)				
Side of stroke	Left*	0.053	0.002	1.05	0.039**
	Right				
Amount of treatment sessions	Between 20 to 50*	-0.019	2.254	0.98	0.993
	More than 50				
Fallow up duration	-	0.061	0.844	1.06	0.943

*Reference group. ** It is significant at level of 0.05.

Table 3. The results of non-zero part of the random effects ZIP model to evaluate balance and motor improvement in stroke patients

Variable	Category	Estimate	Standard Error	OR	p
Age	-	-0.008	0.011	0.99	0.493
Sex	Male*	0.29	0.406	1.35	0.469
	Female				
Stroke type	Hemorrhagic*	-0.08	0.454	0.92	0.854
	Ischemic				
Stroke duration	Less than 6 months(acute phase)*	0.02	0.382	1.02	0.954
	More than 6 months(chronic phase)				
Side of stroke	Left*	-0.25	0.131	0.78	0.063
	Right				
Amount of treatment sessions	Between 20 to 50*	0.09	0.55	1.10	0.858
	More than 50				
Fallow up duration	-	-0.54	0.203	0.58	0.011**

*Reference group. ** It is significant at level of 0.05.

small mean which only made more skewness in data. Before fit the zero inflated models, it is necessary to evaluate the amount of zero inflation in outcome variable using score test (34). The hurdle model resulted in 80% of zeros and ZIP model resulted in 70% zeros in count data (p-value< 0.001). So, both hurdle model and ZIP model was fitted to data. Also, zero inflated models had smaller chi-square statistics and better fit than Poisson model ($\chi^2_{Poisson}$=162, χ^2_{ZIP}=145, χ^2_{hurdle}=157).

Because of longitudinal structure of data, the random effects were added to models. Results of ZIP model with random effects, as presented in Tables 2 and 3, showed that

Table 4. The results of zero inflation part of the random effects hurdle model to evaluate balance and motor improvement in stroke patients.

Variable	Category	Estimate	Standard Error	OR	p
Age	-	0.001	0.019	1.01	0.949
Sex	Male*	-0.21	0.498	0.81	0.675
	Female				
Stroke type	Hemorrhagic*	0.16	0.568	1.18	0.772
	Ischemic				
Stroke duration	Less than 6 months(acute phase)*	0.05	0.822	1.06	0.048**
	More than 6 months(chronic phase)				
Side of stroke	Left*	0.23	0.558	1.25	0.686
	right				
Amount of treatment sessions	Between 20 to 50*	0.07	0.468	1.07	0.888
	More than 50				
Fallow up duration	-	0.51	0.199	1.67	0.014**

*Reference group. ** It is significant at level of 0.05.

Table 5. The results of non-zero part of the random effects hurdle model to evaluate balance and motor improvement in stroke patients.

Variable	Category	Estimate	Standard Error	OR	p
Age	-	-0.003	0.039	0.99	0.938
Sex	Male*	0.23	0.864	1.02	0.978
	Female				
Stroke type	Hemorrhagic*	-0.001	1.308	0.99	0.999
	Ischemic				
Stroke duration	Less than 6 months(acute phase)*	-0.03	0.430	0.97	0.026**
	More than 6 months(chronic phase)				
Side of stroke	Left*	-0.13	1.142	0.88	0.912
	Right				
Amount of treatment sessions	Between 20 to 50*	0.11	0.315	1.12	0.728
	More than 50				
Fallow up duration	-	-0.44	0.557	0.64	0.427

*Reference group. ** It is significant at level of 0.05.

Table 6. The results of goodness of fit test for comparison two models.

Model	ZIP model With random effects	Hurdle model with random effects
-2loglikelihood	165.8	170.6
AIC	199.8	204.6
BIC	227.6	232.4

the covariates of sex, age, side of stroke and follow up duration (time effect) were statistically significant ($p<0.05$).

The results of hurdle model with random effects, in Tables 4 and 5, showed that the covariates of stroke duration and follow up duration had significant effect on balance improvement in stroke patients ($p<0.05$).

The comparison of Hurdle model and ZIP model with random effects are presented in Table 6. The results showed that the ZIP model with random effects had less statistics than hurdle model.

Discussion

In this paper, the balance improvement was evaluated through fall assessment in stroke patients who were under treatment with the occupational therapy. Because of the longitudinal zero inflated structure of data, the random effects hurdle model was compared to a random effect ZIP model. According to goodness of fit indexes, the random effects ZIP model with less statistics had priority to random effects hurdle model; that was probably due to small range of count event in the present study. In review of studies with zero inflation, it is inferred that the amount of zero inflation and sample size and type of study (longitudinal or cross sectional) have important role on model selecting.

The results of ZIP model with random effects showed that the covariates of age, sex and side of stroke and follow up duration (time effect) had significant effect on balance improvement. With increasing age, odds ratio of balance improvement was decreased to 6 percent.

In fact, older patients were more at risk of fall. In a study, risk of repeated falls were higher in patients older than 65 years old (HR, 1.4); it was similar to our results. (29). Odds ratio of balance improvement among females was 4 percent less than male. Another study demonstrated a high prevalence of falls and fall-related injuries in female (OR, 1.5) (30).

Odds ratio of balance improvement in patients with right affected side was 5 percent more than patients with left affected side. In fact, the difference in balance improvement among left and right affected side was very small. In a study, logistic regression revealed a significant relation between left hemisphere lesion (right affected side) and fall (OR_{adj}, 0.28, $p=0.02$) (35), which confirmed the results of present study. In an-

other study it was indicated that the patients with right affected side (vs. left) have a 4 times greater risk of fall within 6 months (28). The present study was also showed that the fall rate decreased to 42% during a one-year follow up. In fact, the occupational therapy has been effective on balance improvement more than 40% during one year. In a study on the usefulness of occupational therapy for motor status via balance indexes in stroke patients, logistic regression model showed that the balance improvement under the occupational therapy was more in men and younger patients (27). That was according to the results of the present study.

In conclusion, older patients and women and patients with left affected side were at a higher risk of fall and balance impairment, so they need more care and therapy. Although the longitudinal count data with excess zeros almost occur, but few methods have been developed for analyzing related count data with excess zeros. The hurdle model with random effects had poor fit compared to the ZIP model with random effects. Thus choosing the correct model is important in order to reach reliable results.

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