

## Original Article

**Application of spatial Besag, York and Mollie method in estimating interprovincial neck pain prevalence in the National Disease and Health Survey in Iran**Mina Ghorbanpour<sup>1\*</sup>, Kazem Mohammad<sup>1</sup>, Mirsaeed Yekaninejad<sup>1</sup>, Mohamad Ali Mansournia<sup>1</sup>, Hooshang Saberi<sup>2</sup>, Mahbubeh Parsaeian<sup>1</sup>, Arron Mungela Foma<sup>3</sup><sup>1</sup> Department of Epidemiology and Biostatistics, School of Public Health, Tehran University of Medical Sciences, Tehran, Iran<sup>2</sup> Department of Neurosurgery, Brain and Spinal Injury Research Center, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran<sup>3</sup> Department of Immunology and Biology, School of Medicine, Tehran University of Medical Sciences, Tehran, Iran

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## ABSTRACT

**Background & Aim:** Geographical analysis of the frequency of disease incidence can have an important role in the allocation of resources, facilities, and manpower in addition to the formulation and evolution of etiological assumptions. The main objective of this study was to estimate the prevalence of neck pain interprovincially, and set a disease mapping using spatial Besag, York and Mollie (BYM) with regard to surrounding neighborhoods. To reduce the incidence of neck pain in adulthood, identification of risk factors that predict the onset, and continuation of pain in the patients is important.

**Methods & Materials:** The population examined in this study was extrapolated from records of the “National Disease and Health Survey in Iran,” which had a data plan of a general population survey conducted during 1999-2000, in which adults responded on the incidence of neck pain. The participants were guided by a questionnaire that had an image on which they could identify the exact location of the pain.

**Results:** Explanatory variables in the model included sex, education level, area of residence, smoking, age and body mass index, and all of them showed a significant relationship with neck pain. To have a better model for a more reliable prediction, we grouped the provinces into divisions to have a more regular shape since the spatial BYM model cannot simultaneously account for population and spatial patterns. In neck pain, prevalence estimated by spatial BYM, Lorestan province with 7.85% had the lowest prevalence while Kurdistan province with 17.27% had the highest prevalence. Furthermore, in the male population, Ghazvin province with 5.53% had the lowest prevalence, whereas Kurdistan province with 10.33% had the highest prevalence of neck pain. Besides, in the female population, the Lorestan province with 10.33% had the lowest prevalence, while the province of Yazd with 22.45% has the highest prevalence of neck pain.

**Conclusion:** In this study, the model assumed included measurable and immeasurable factors to provide reliable estimates for each province. The application of spatial BYM method with the inclusion of the location of disease occurrence is a more efficient and reliable method for diseases mapping with a higher power of predictability.

\* Corresponding Author: Mina Ghorbanpour, Postal Address:  
 Department of Biostatistics, School of Public Health, Tehran

University of Medical Sciences, Tehran, Iran.  
 Email: ghorbanpour.minaa@yahoo.com

## **Introduction**

As a result of the increase availability of information on diseases and deaths, relevant ways to analyze this type of data are growing in credence so as to meet up with varying needs. One of these methods is the mapping of disease or mortality, which considers the geographical distribution of the disease or mortality alongside other risk factors. Disease mapping refers to a set of statistical methods aimed at obtaining accurate estimates of the incidence or prevalence of diseases or deaths which as a result, is set in the form of geographical maps (1).

Geographical analysis of rates of disease can have an important role in the allocation of resources and facilities and manpower in addition to the formulation and evolution of etiological assumptions and interventions in the areas that need special focus. Often these rates are calculated by classical means taking into account the assumption of independence between areas. However, there is no assumption of independence for environmental studies and especially health variables, and the neighboring areas have a spatial correlation which causes the distance between the points.

The observations that their dependency is induced by their position in space in the study, is demonstrated by the spatial data (2). This dependency is a function of the observation distance from each other. The assumption of independence greatly assists the comprehension of theoretical concepts. In many cases, observations are not independent and their spacial location increases their dependency on each other.

Advances in spatial statistical methods in epidemiology have been increasing steadily. One of the earliest examples of the important role of geographical analysis of disease was the analysis of cholera outbreaks in the east end of London by Snow (3) in 1854. The most essential aspect of spatial data is the propensity to take advantage of the correlation structure of the data to increase the accuracy of statistical analysis, and as a result increase its efficiency when compared to the classic methods.

In recent years, the full Bayes model has

been adopted by most applications for disease mapping using the information of adjacent units of the intended area. In Full Bayes model, the iterative method such as Markov Chain Monte Carlo method is used for estimating parameters. First used on the log relative risks in Bayesian mapping by Clayton and Kaldor, it assumes that the conditional variance is constant, and hence, it is inappropriate for asymmetrical maps with variably located neighbors. The model we have legislated was pioneered by Clayton and Kaldor (4) in 1987, and subsequently, developed by Besag et al. (5) in 1991.

One of the objectives of disease mapping involves using estimates obtained from space, to designate high-risk areas that need attention as well as the construction of the disease mapping.

Attempts have been made to evaluate the sensitivity of the spatial Besag, York and Mollie (BYM) model. These estimates have been restricted to a limited domain of Bayesian models (6-10).

So far numerous articles have reported in the field of disease mapping and the use of the spatial BYM method indicating the ability of this method in disease and mortality, to geographically analyze and express assumptions on the etiology. Some examples can be cited in studies of intestinal disorders in Manitoba, Canada (11), effects of air pollution on the prevalence of infertility in Barcelona Spain (12), 13 cancer deaths in Spain (13), and many others.

The main objective of this study was to estimate the prevalence of neck pain in Iran interprovincially, using spatial BYM in relation to surrounding neighborhoods, or in other words the development of spatial models to estimate the prevalence of neck pain interprovincially, in Iran.

Neck pain is the sensation of discomfort in the neck zone, and the neuromuscular system is one of the three commonly affected zones. Despite the fact that improved the quality of work and efficiency decreases neck pain, the repercussions of this pain include disability, reduced quality of life, as well as impairment of individual job performance. Therefore, to reduce the incidence of neck pain in adulthood, identification of risk factors that predict the onset and continuation of pain in these subjects is important (14). In

systematic studies on the general population, parameters - such as female sex, old age, previous history of back and neck pain - have been associated with the onset of neck pain.

The study of Makela et al. (15) in 1991 on 8000 people from the general population in Finland reported an unequal distribution of the prevalence of neck pain comprising 9.5% in men and 13.5% in women. In this study, we mention high weight as an important factor in the incidence of neck and shoulder pain. In a study that was done in 2000 by Vikat et al. (16), people with a body mass index (BMI) lower than 15, have a higher incidence of neck and shoulder pain. According to the data obtained from adults aged 18 years or older (n = 29,828) who participated in the 2002 National Health Interview Survey, which is a population-based survey of US adults, the prevalence of neck pain was reported in 13.7%, of which 11.6% were men, and 15.5% were women (17). Palmer et al. (18) in a study conducted in 2001 on 21000 people from the general population of Britain in the age group 16-24 years, reported that 12.8% of men and 19.1% women complained of neck pain within a week.

Akbari and Azari (19) in a study comprising 1377 adults, reported a prevalence of neck pain along with shoulder pain in 6.2% of people referred to physical therapy clinics in Tehran. They stated in their report that older aged people more than others had the propensity to experience neck pain.

## Methods

In this study, the population was extracted from archives of the “National Disease and Health Survey in Iran” recorded in a data plan that involved a general population survey conducted during 1999-2000, using cluster sampling technique. The statistical framework used household lists registered in the health departments of all provinces except Tehran, and a rate of a sample size to society which was projected to 1/1000 community. In this study, information about the population over 18 years (32,018 people) was examined. In spatial modeling for neck pain, explanatory variables

into the model included gender, education level, place of residence, smoking, age, and BMI.

**Besag, York and Mollie (BYM) model:** In this model used for estimating relative risks, area-specific random effects are decomposed into;

- i. A component that takes into account the effects that vary in a structured manner in space and also varies smoothly across areas (clustering or correlated heterogeneity) and
- ii. A component that models the effects that vary in an unstructured way between areas (heterogeneity or uncorrelated heterogeneity).

The model introduced by Clayton and Kaldor (4) and developed by Besag et al. (5), is formulated as follows:

$$y_i \sim \text{Poisson}(e_i \theta_i)$$

$$\log(\theta_i) = \mu + v_i + u_i$$

$$e_i = n_i \left( \frac{\sum_i y_i}{\sum_i n_i} \right)$$

Where,  $y_i$  is the number of cases of disease observed in area  $i$ ,  $e_i$  is the expected number and it turns out,  $n_i$  is the number of people at risk of disease,  $\theta_i$  is vector of relative risk and uncertainty,  $\mu$  is an overall level of the relative risk,  $u_i$  is the correlated heterogeneity, and  $v_i$  is the uncorrelated heterogeneity. Allocation of prior distribution in the full Bayes model is important, so as to determined prior distribution of random effects and overall average.

Bayesian modeling requires specification of prior distribution for random effects. The distribution model for the unstructured heterogeneity is,

$$V_i \sim N(0, \tau_v^2)$$

For the clustering component, a spatial correlation is used, where estimation of the risk in any area depends on neighboring areas. The conditional autoregressive model proposed by Besag et al. (5) is used,

$$[u_i | u_j, i \neq j, \tau_u^2] \sim N \left[ \frac{\sum_j W_{ij} u_j}{\sum_i W_{ij}}, \frac{\tau_u^2}{\sum_i W_{ij}} \right]$$

Where,

$$W_{ij} = \begin{cases} 1 & \text{if } i, j \text{ are adjacent} \\ 0 & \text{otherwise} \end{cases}$$

Joint distribution is,

$$\text{prior}(\tau_u, \tau_v) \propto e^{\frac{-\varepsilon}{2\tau_u}} e^{\frac{-\varepsilon}{2\tau_v}}, \tau_u, \tau_v > 0$$

The full posterior distribution is,

$$P(u, V, \tau_u, \tau_v | y_i) = \prod_{i=1}^n \left\{ \exp \frac{(-e_i \theta_i)(e_i \theta_i)^{y_i}}{y_i!} \right\} \times \tau^{-\frac{n}{\tau^2}} \exp \left\{ \frac{-1}{2\tau_u} \sum_i \sum_j (u_i - u_j)^2 \right\} \times \exp \left\{ \frac{-1}{2\tau_v} \sum_{i=1}^n V_i^2 \right\} \times \text{prior}(\tau_u, \tau_v)$$

This method is computationally convenient, using a modification of the well-known iterative reweighted least square algorithm. Here, we use Taylor expansion. In this study, R-INLA was used for data analysis. INLA uses Laplace approximations to obtain posterior marginals, numerical algorithms for sparse matrices and parallel computation via OpenMP. That is why the main advantage of INLA is a huge improvement of speed compared to MCMC alternatives.

Parameters  $\tau_u^2$  and  $\tau_v^2$  control the variability of  $u$  and  $v$ . In a full Bayes analysis, prior distributions must be specified for those parameters. We considered log-gamma distributions for both.

Modeling the heterogeneity and clustering variation represents a way of allowing for unmeasured explanatory variables. More specifically, modeling the heterogeneity variation allows for unmeasured variables which vary

between areas in an unstructured way. Modeling the clustering variation allows for those unmeasured risk factors that vary smoothing with location. Specifically, the clustering term may be thought of as a way of modeling the effect of location in a flexible way (20).

Where the pattern of variation of the covariate is similar to that of disease risk, location may act as a confounder. Now, since the clustering term stands for the effect of location, introducing it in the model causes the estimate of the regression coefficient  $\beta$ , to be controlled for the effect of location. Thus, when location acts as a confounder, we should not be surprised to see the regression coefficient change on introduction of the clustering term.

## Results

Spatial BYM statistical analysis was employed in our calculations, using R-INLA software. The gender distribution of population in this study constituted 14,571 (45.5%) men and 17,447 (54.5%) women, respectively. In educational level, 24,654 cases (77.2%) had no diploma while 7310 cases (22.8%) had either a diploma or a higher certificate. Among the 32,018 participants, 11,370 (35.5%) lived in rural areas and 20,648 (64.5%) in urban areas. Basic information of neck pain in different groups is reported in table 1.

**Table 1.** The prevalence of neck pain in the groups studied in adults

Variables	Number of study subjects	The number of cases of neck pain	Prevalence of neck pain (95% CI)
Country	32,018	3907	12.2 (11.84-12.56)
Age groups			
18-24	7593	281	3.70 (3.28-4.13)
25-34	8489	633	7.49 (6.90-8.02)
35-44	6438	836	12.99 (12.16-13.81)
45-54	3791	720	18.99 (17.74-20.24)
55-64	2550	604	23.69 (22.04-25.34)
≥ 65	3157	833	26.39 (24.85-27.92)
Sex			
Female	17,477	2907	16.66 (16.11-17.21)
Male	14,571	1000	6.86 (6.45-7.27)
Education level			
No diploma	24,654	3453	14.01 (13.57-14.44)
Diploma or higher certificate	7310	449	6.14 (5.59-6.69)
Area of residence			
Rural	11,370	1498	13.18 (12.55-13.80)
Urban	20,648	2409	11.67 (11.23-12.10)
Smoking			
Yes	4321	457	10.6 (9.66-11.49)
No	27,680	3449	12.5 (12.07-12.85)
BMI			
Normal	18,725	1999	10.68 (10.23-11.12)
Overweight	8667	1244	14.35 (13.62-15.09)
Obese	3581	609	17.01 (15.78-18.24)

BMI: Body mass index, CI: Confidence interval

**Table 2.** The true prevalence and estimated prevalence of neck pain by BYM method

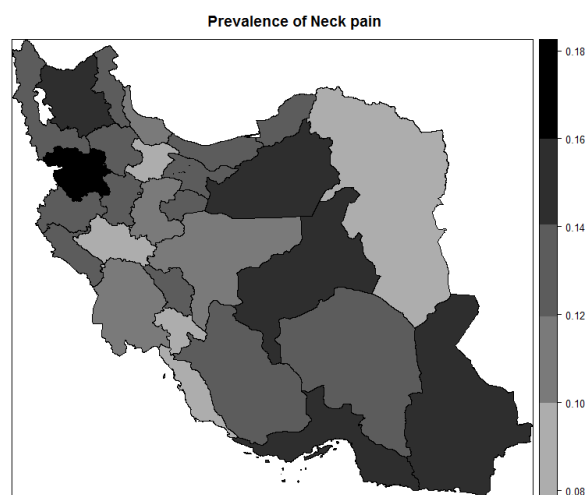
Province	Society		Spatial BYM method	
	True prevalence (95% CI)	Length of CI	Estimation of prevalence (95% CI)	Length of CI
Markazi	10.32 (7.84-12.80)	4.96	10.96 (8.76-13.34)	4.58
Gilan	10.73 (8.98-12.48)	3.50	10.86 (9.27-12.56)	3.29
Mazandaran	13.75 (11.98-15.52)	3.54	13.50 (11.88-15.21)	3.33
Azarbayjane Sharghi	14.43 (12.85-16.01)	3.16	14.32 (12.86-15.86)	3
Azarbayjane gharbi	13.21 (11.26-15.16)	3.90	13.26 (11.48-15.14)	3.66
Kermanshah	12.22 (10.01-14.42)	4.41	12.25 (10.33-14.31)	3.98
Khuzestan	9.90 (8.45-11.35)	2.90	10.06 (8.73-11.47)	2.74
Fars	12.02 (10.47-13.58)	3.11	12.09 (10.65-13.61)	2.96
Kerman	14.23 (12.05-16.41)	4.36	13.92 (11.94-16.03)	4.09
Khorasan	9.03 (8-10.05)	2.05	9.26 (8.28-10.28)	2
Esfahan	11.05 (9.72-12.38)	2.66	11.18 (9.94-12.48)	2.54
Sistan-Baluchestan	16.53 (13.46-19.59)	6.13	15.59 (12.87-18.54)	5.67
Kurdistan	17.62 (14.58-20.66)	6.08	17.27 (14.56-20.16)	5.60
Hamedan	12.12 (9.89-14.35)	4.46	12.55 (10.54-14.68)	4.14
Charmahal-Bakhtyari	14.47 (10.86-18.08)	7.22	13.97 (11.10-17.17)	6.07
Lorestan	6.64 (4.73-8.55)	3.82	7.85 (6.07-9.79)	3.72
Illam	13.20 (10.12-16.27)	6.15	12.83 (10.30-15.61)	5.31
Kohgiluyeh-Boyerahmad	9.28 (6.49-12.08)	5.59	9.78 (7.43-12.41)	4.98
Bushehr	8.65 (5.93-11.36)	5.43	9.40 (7.15-11.89)	4.74
Zanjan	13.60 (10.46-16.73)	6.27	13.74 (11.10-16.61)	5.51
Semnan	16.58 (12.83-20.34)	7.51	15.58 (12.55-18.95)	6.40
Yazd	15.73 (12.58-18.88)	6.30	15.49 (12.66-18.55)	5.89
Hormozgan	15.69 (12.47-18.91)	6.44	15.12 (12.40-18.10)	5.70
Tehran	12.91 (12.06-13.76)	1.70	12.85 (12.04-13.69)	1.65
Ardebil	12.96 (10.21-15.72)	5.51	12.66 (10.33-15.21)	4.88
Ghom	12.68 (9.45-15.90)	6.45	12.23 (9.55-15.22)	5.67
Ghazvin	6.41 (4.07-8.75)	4.68	7.96 (5.86-10.27)	4.41
Golestan	12.71 (10.21-15.22)	5.01	12.44 (10.26-14.80)	4.54

BYM: Besag, York and Mollie, CI: Confidence interval

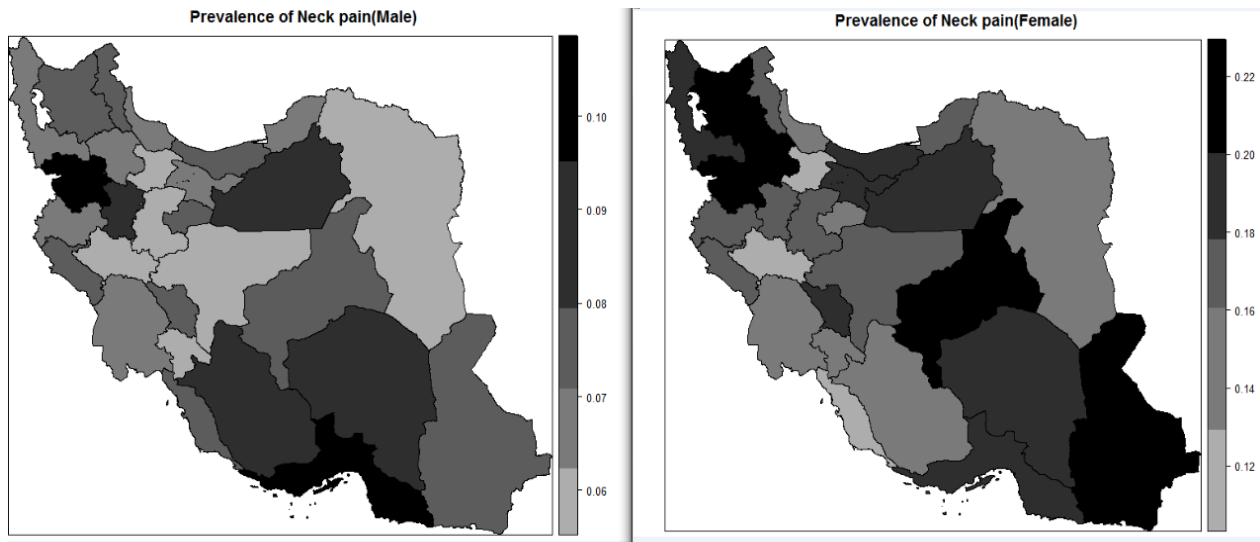
The prevalence and distribution of neck pain in the different provinces of Iran is presented in table 2. According to the table statistics, Ghazvin province with a 6.4% and Lorestan with 6.6% had the lowest neck pain prevalence, while Kurdistan province with 17.6% had the highest prevalence of neck pain. In the men population, Markazi and Ghazvin provinces with 3.1% and 3.3% had the lowest prevalence, whereas Semnan and Kurdistan with 12.5% and 12.3% had the highest prevalence of neck pain, respectively. In the women population, Lorestan with 8.4% had the lowest prevalence and Kurdistan with 22.7% had the highest prevalence of neck pain in Iran. In the rural areas, Bushehr with 5.6% had the lowest prevalence and Sistan-Baluchestan with 22.2% had the highest prevalence of neck pain. In the urban areas, Ghazvin with 5.2% had the lowest prevalence and Kurdistan with 18.5% had the highest prevalence of neck pain.

In spatial modeling for neck pain, explanatory variables in the model included sex, education level, area of residence, smoking, age and BMI.

In neck pain prevalence estimated by spatial BYM, Lorestan province with 7.85% and Ghazvin with 7.96% had the lowest prevalence and Kurdistan province with 17.27% had the highest prevalence of neck pain (Figure 1).



**Figure 1.** Geographic map of the spatial method to estimate the prevalence of neck pain (explanatory variables: sex, education level, area of residence, smoking, age and body mass index)



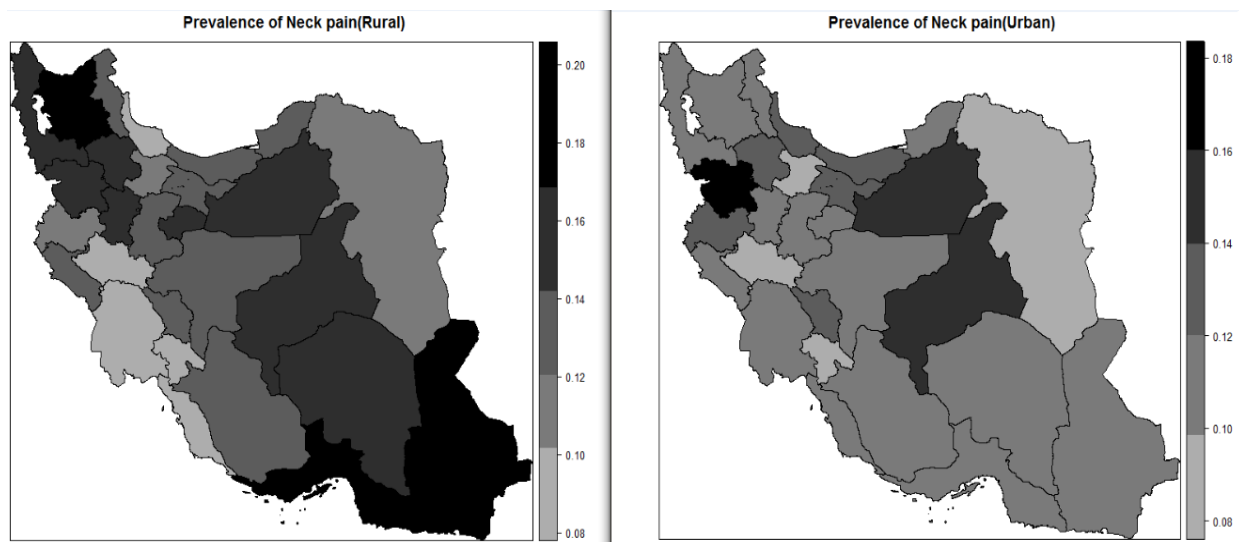
**Figure 2.** Geographic map of the spatial method to estimate the prevalence of neck pain in men and women population (explanatory variables: education level, area of residence, smoking, age and body mass index)

In the men population, Ghazvin province with a 5.53%, Khorasan 5.59%, Markazi 5.69% and Lorestan 5.88% had the lowest prevalence, while Kurdistan province with a 10.33% and Hormozgan with 10% had the highest prevalence of neck pain. In the female population, the Lorestan province with 10.33% and Bushehr with 10.58% had the lowest prevalence, whereas the provinces of Yazd with 22.45% and Kurdistan with 21.95% had the highest prevalence of neck pain (Figure 2).

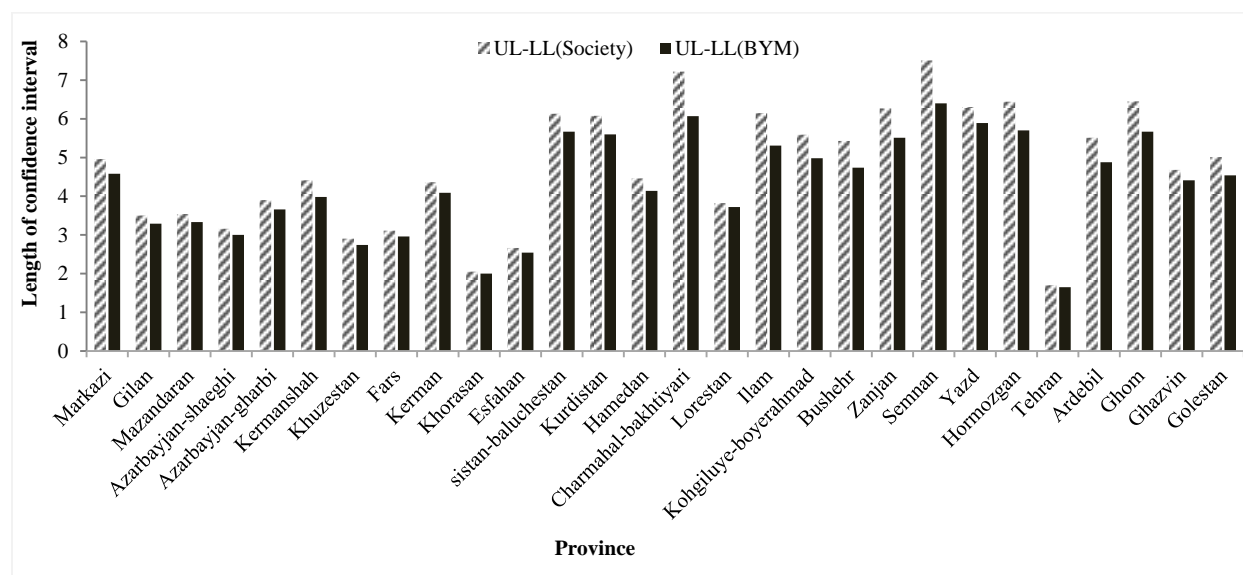
In the rural areas, Bushehr with 7.81% had

the lowest prevalence and Sistan-Baluchestan with 20.1% had the highest prevalence of neck pain. In the urban areas, Ghazvin with 7.61% had the lowest prevalence and Kurdistan with 17.35% had the highest prevalence of neck pain (Figure 3).

Estimates of the spatial BYM model is very close to the values of the true prevalence in each province. The length of the confidence interval for an estimate of the spatial BYM model was less than the length of true prevalence confidence interval in each province (Table 2).



**Figure 3.** Geographic map of the spatial method to estimate the prevalence of neck pain in rural and urban areas (explanatory variables: sex, education level, smoking, age and body mass index)



**Figure 4.** Comparison of the length of the confidence interval for neck pain prevalence between the true prevalence and its estimation by spatial Besag, York and Mollie method

Shorter length of the confidence interval in spatial BYM method means that this method has a good accuracy (Figure 4).

Table 2 shows the true prevalence and its estimation by spatial BYM method:

Coefficients and standard errors in the spatial BYM method is shown in table 3. All the variables entered into the model were significant. Chance of having neck pain in the rural subjects was 1.38 times more than in urban subjects. The possibility of having neck pain in the females was 2.13 times more than in males. The probability of having neck pain in people without an educational diploma was 3.92 times more than in people with a diploma or higher certificate. Furthermore, the chance of having neck pain in smokers was 2.73 times more than non-smokers. Furthermore, with each unit increase in age, the chance of neck pain increased by 0.06 and with each unit increase in

BMI, the chance of neck pain increased by 0.07.

### Discussion

According to our reports, the prevalence of neck pain in adults aged 18 years or older was 12.2% and the estimation of prevalence by the spatial method was 12.4%. The estimated prevalence of neck pain in men was 7.27%, and in women, it was 16.76%. Bovim et al. (21) in 1994 reported a 13.8% annual prevalence of neck pain in the general population of Norway. Strine and Hootman in 2007 (17), according to data obtained from adults aged 18 years or older (n = 29,828) comprising the participants of a 2002 National Health Survey interview, population-based survey of US adults, reported a 13.7% prevalence of neck pain. The gender distribution of prevalence comprised 11.6% men and 15.5% women.

**Table 3.** Coefficients and signficancy in spatial BYM method

Variables	Spatial BYM method		
	$\beta$ (95% CI)	OR (95% CI)	P-value
Area of residence (Reference: Urban)	0.323 (0.121-0.525)	1.38(1.129-1.690)	0.002
Sex (Reference: Male)	0.756 (0.031-1.481)	2.13(1.031-4.398)	0.040
Education level (Reference: Diploma or Higher certificate)	1.365 (0.0889-1.841)	3.92(2.432-6.305)	< 0.001
Smoking (Reference: No)	1.002 (0.224-1.780)	2.73 (1.251-5.931)	0.012
Age	0.054 (0.038-0.070)	1.06 (1.039-1.072)	< 0.001
BMI	0.067 (0.040-0.094)	1.07 (1.040-1.099)	< 0.001

BYM: Besag, York and Mollie, CI: Confidence interval, BMI: Body mass index, OR: Odds ratio

Study of Makela et al. in 1991 on 8000 people from the general population in Finland reported a 9.5% prevalence of neck pain in men and 13.5% in women (15). Palmer et al. conforming to a study conducted in 2001 on 21,000 people from the general population of Britain among the age group 16-24 years, 12.8% of men and 19.1% women recorded neck pain within a week (18).

In this study, the prevalence of neck pain in adults aged 18 years or older in Tehran was 12.85%. This rate had the following distribution per gender; in men, it was 6.39%, while in women it was 18.07%.

Akbari and Azari reported a 6.7% prevalence of neck pain in the general population of Tehran, of which 3.2% were men and 9.7% comprised of women (19).

In some other studies conducted among adults, neck pain was associated with metabolic syndromes, including obesity, and this relationship was more robust in men (16, 22, 23).

The data analysis of our studies demonstrated that the increased prevalence of neck pain had a direct relation with increasing age, and all of the explanatory variables in the model (including sex, education level, area of residence, smoking, age, and BMI), also showed a significant relationship with neck pain. On the other hand, Akbari and Azari in 1998 reported a prevalence of neck pain along with shoulder pain in 6.2% of people referred to physical therapy clinics in Tehran. They stated in their report that elderly people more than others had the likelihood of experiencing neck pain. In addition, Akbari and Azari pointed a direct relationship between age and prevalence of neck pain (19).

## Conclusion

The inclusion of the location of disease occurrence in this study introduces a new dimension of estimating the prevalence of neck pain, adding more credibility to estimation studies. The previous studies have either been univariate or multivariate, and to the best of our knowledge, this is the first study to consider the effect of locations of disease occurrence, thereby estimating the prevalence of disease with higher

fidelity. The application of BYM method is a more efficient and reliable method for diseases mapping with a higher power of predictability.

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