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PREVALENCE OF INTELLECTUAL DISABILITIES AND EPILEPSY IN DIFFERENT FORMS OF SPASTIC CEREBRAL PALSY IN ADULTS

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SUMMARY

Background: *Spastic cerebral palsy may be interconnected with other neurodevelopmental disorders such as intellectual disabilities, and epilepsy. Brain synaptic plasticity and successful restorative rehabilitation may also contribute to diminish neurological deficit of patients having cerebral palsy. The aim of this study was to investigate the prevalence of intellectual disabilities and epilepsy in adult patients with different forms of spastic cerebral palsy and to find out correlation between the severity level of intellectual disabilities and epilepsy.*

Subjects and methods: *Adults diagnosed with different forms of spastic cerebral palsy were analyzed during a three-month period. The investigated features were: gender and age; form of cerebral palsy; the prevalence of intellectual disabilities and epilepsy. Intellectual disabilities were divided into 4 severity levels. The correlation between the severity level of intellectual disabilities and epilepsy was statistically analyzed.*

Results: *Intellectual disability was present in 55% of patients diagnosed with spastic cerebral palsy. Epilepsy was present in 36% of such patients. It was recorded in 51.1% of quadriplegic, 21.9% of diplegic, and 19.2% of hemiplegic patients. Intellectual disability was present in 73.8% of quadriplegic, 31.3% of diplegic, and 53.8% of hemiplegic patients. The statistically significant correlation existed between the severe intellectual disability and epilepsy.*

Conclusions: *Intellectual disabilities and epilepsy most frequently occurred in patients with most severe forms of spastic cerebral palsy. Epilepsy is strongly correlated to the severity level of intellectual disability. Such patients require additional special modes of treatment and restorative rehabilitation to improve the functional outcome.*

Key words: *spastic cerebral palsy - intellectual disability - epilepsy - prevalence*

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INTRODUCTION

It is recognized that spastic cerebral palsy (CP) may be interconnected with other neurodevelopmental disorders such as intellectual disabilities, and epilepsy (Bax et al. 2005). However, the prevalence of such disorders in CP patients may differ considering their age. It is also known that brain synaptic plasticity and successful restorative rehabilitation may contribute to diminish neurological deficit of CP patients (Winstein & Kay 2015). Therefore, we have speculated whether the prevalence of such disorders may be decreased in adult CP patients in comparison to children.

Cerebral palsy is a disorder of movement and positional control caused by non-progressive brain damage or deficiency in cerebral development. In order to confirm the diagnosis, a motor impairment due to brain non-progressive dysfunction expressed early in life needs to exist (Badawi et al. 1998).

Damage to the brain development may occur in the prenatal, perinatal and postnatal period. Prenatal events are responsible for the majority of damage (75-80%), while less than 10% of it occurs perinatally due to asphyxia or birth trauma (Sankar & Mundkur 2005).

Clinical signs may differ depending on the form of palsy and co-morbidity. Speech difficulties, hearing and vision disturbances, difficulty in learning, as well as intellectual disabilities, and epilepsy may also develop depending on the cause of brain damage (MacLennan 1999). Nonetheless, neurological deficit may gradually decrease as a result of alterations in synaptic connectivity, a process of brain plasticity that may occasionally initiate neurological recovery supported by restorative rehabilitation. Understanding the mechanism of brain plasticity is helpful to develop treatment programs to improve functional outcome (Bax 1987). The goal of restorative rehabilitation is to gain recovered function by repetitive exercise. Therefore, one is to expect neurological deficit of lesser form in adult CP patients undergoing rehabilitation programs in comparison to children.

Spastic form of palsy occurs most often, in about 75% of cases, while dystonic and ataxic forms are rare

(Bax 1987). Spastic tetraplegia/quadriplegia is a form of cerebral palsy usually accompanied by intellectual disabilities and epilepsy. Spastic diplegia is a form of cerebral palsy with more severe spasticity of the lower limbs, while the upper limbs spasticity may be discrete or nonexistent. In spastic hemiplegia there is a presence of unilateral spastic paralysis. In all forms of spastic cerebral palsy there is a resistance to passive movements and stretching, leading to contraction of the joints.

Intellectual disability is not an illness or a specific disability, but administratively designated name for a neurodevelopmental disorder of different genetic, social and health conditions having a common characteristic - significantly below average intellectual functioning. It is a disability characterized by significant limitations in both intellectual functioning and in adaptive behavior, which covers many everyday social and practical skills (Besag 2002). It may differ in its appearance depending on its severity level.

Epilepsy is a chronic brain disease of various causes marked by repeated seizures and usually accompanied by electroencephalographic (EEG) abnormalities. Epidemiological studies have shown that there is a high level of behavioral disorders, as well as intellectual disabilities in people having epilepsy regardless of their age (Wakamoto et al. 2000, Singh et al. 2003).

It is well known that cerebral palsy is often associated with intellectual disabilities and epilepsy (Bax et al. 2005). According to the results from Spain, intellectual disabilities were present in great majority of patients with CP (Brigas-Grande et al. 2002). Such disabilities were mostly present in quadriplegic patients and were correlated to epilepsy (McDermott et al. 2005).

The goal of this paper was to investigate the prevalence of intellectual disabilities and epilepsy in adult patients with different forms of spastic cerebral palsy and to find out possible correlation between the severity level of intellectual disabilities and epilepsy in such patients.

SUBJECTS AND METHODS

The study was conducted at the Center for Medical Expertise of the Federation of Bosnia and Herzegovina and the Center for Social Work of the Herzegovina-Neretva county during a 3-month period between January 15 and April 15, 2015.

A group of 100 adult patients diagnosed with different forms of spastic cerebral palsy from the area of Herzegovina-Neretva county were analyzed. Inclusion criteria were: spastic cerebral palsy of different forms; adult patients, more than 18 years old. Exclusion criterion was: patients younger than 18 years. The investigated features were: patients' gender and age; form of cerebral palsy; the prevalence of intellectual disabilities and epilepsy.

Intellectual disabilities were divided according to DSM-V into 4 severity levels: mild, moderate, severe and profound disabilities. The correlation between the severity level of intellectual disabilities and epilepsy in CP patients was also analyzed.

In the statistical data analysis of nominal and ordinal variables, χ^2 test was used. Fisher's exact test was used when the expected frequency of dichotomous variables were lacking. Categorical variables were expressed with more sub-module additional exact tests. Spearman's correlation (ρ) was used to determine if there was an association between the level of intellectual disabilities and prevalence of epilepsy. Accepted possibility of errors was when $\alpha<0.05$, and the differences between groups were accepted as statistically significant at $p<0.05$. For the statistical data analysis SPSS for Windows (version 17.0, SPSS Inc., Chicago, Illinois, USA) and Microsoft Excel (version 11th Microsoft Corporation, Redmond, WA, USA) were used.

RESULTS

The studied sample consisted of 62 males and 38 females aged between 18 and 58 years. The gender difference between the groups was statistically significant (χ^2 test=5.76; df.=1; p=0.016) (Figure 1). Gender distribution of patients did not differ significantly in their frequency according to the forms of spastic cerebral palsy (χ^2 test=0.78; df.=2; p=0.677) (Figure 2).

There was a statistically significant difference among the groups of patients regarding the age, with the predominance of 18-28 age group (χ^2 test=74.60; df=4; p<0.001) (Figure 3). There were 42 quadriplegic, 32 diplegic, and 26 hemiplegic patients in the studied sample (Table 1). Forms of spastic cerebral palsy did not differ significantly in their distribution (χ^2 test=0.78; df=2; p=0.677) (Figure 4).

Intellectual disability was recorded in 55 patients (Table 2), among them in 31 (73.8%) quadriplegic, 10 (31.3%) diplegic and 14 (53.8%) hemiplegic patients (Figure 5). The difference among the groups was statistically significant (χ^2 test=13.31; df=2; p=0.001). Mild level of intellectual disability was recorded in 6 (10.9%), moderate level in 16 (29.1%), severe level in 29 (52.7%), and profound level of disability in 4 (7.3%) patients.

Epilepsy was recorded in 36 patients (Table 2), among them in 24 (57.1%) quadriplegic, 7 (21.9%) diplegic and 5 (19.2%) hemiplegic patients (Figure 6). The difference among the groups was statistically significant (χ^2 test=14.09; df=2; p=0.001).

Intellectual disabilities were found in 27 out of 36 (75%) of patients with CP and epilepsy. Epilepsy was recorded in 13 (36.1%) patients with severe level of intellectual disability, 7 (19.4%) with moderate level, 4 (11.1%) with mild, and in 3 (8.3%) patients with profound level of disability (Figure 7). The difference

between the groups of patients with and without epilepsy was statistically significant (χ^2 test=7.84, df=1; p=0.005). Spearman's correlation coefficient of 0.30 demonstrated positive association between the level of intellectual disabilities and prevalence of epilepsy.

Table 1. Prevalence of the severity level of intellectual disabilities and spastic cerebral palsy

The severity level of intellectual disabilities	N (%) of patients with spastic cerebral palsy		
	Hemiplegia	Diplegia	Quadriplegia
Profound	1 (3.8)	0	3 (7.1)
Severe	3 (11.5)	3 (9.4)	23 (54.8)
Moderate	8 (30.8)	4 (12.5)	4 (9.5)
Mild	2 (7.7)	3 (9.4)	1 (2.4)
Without disability	12 (46.2)	22 (68.8)	11 (26.2)
Total	26 (100.0)	32 (100.0)	42 (100.0)

Table 2. Epilepsy to intellectual disabilities ratio in patients with spastic cerebral palsy

Variables	N (%) of patients with spastic cerebral palsy		χ^2 test	p
	Present	Absent		
Intellectual disabilities	55 (55.0)	45 (45.0)	1.00	0.317
Epilepsy	36 (36.0)	64 (64.0)	7.84	0.005

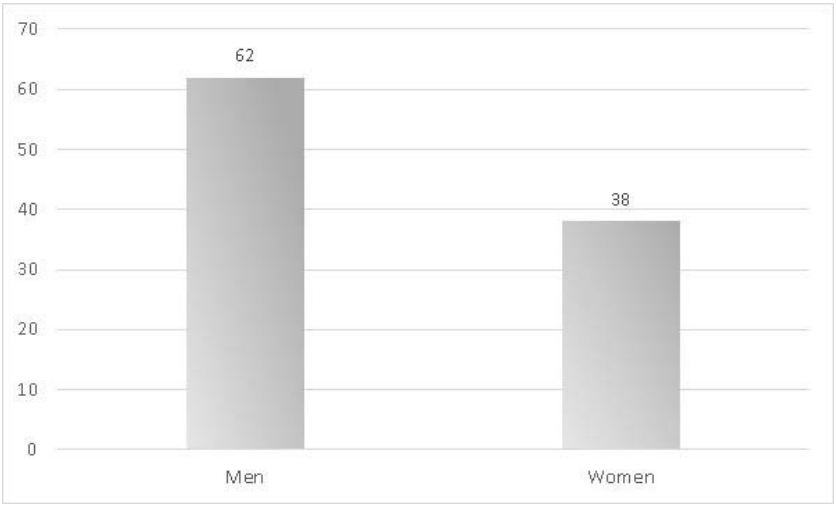

Figure 1. Gender distribution of patients with spastic cerebral palsy

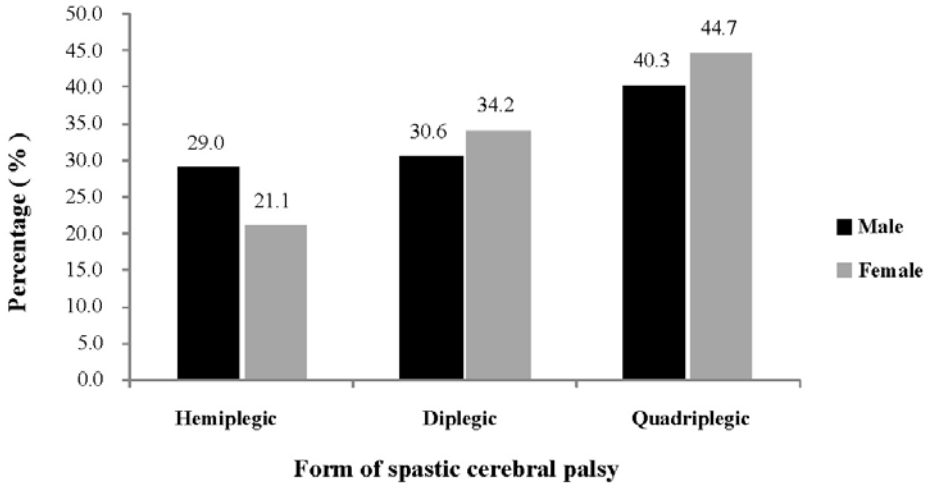

Figure 2. Gender distribution of patients according to the forms of spastic cerebral palsy

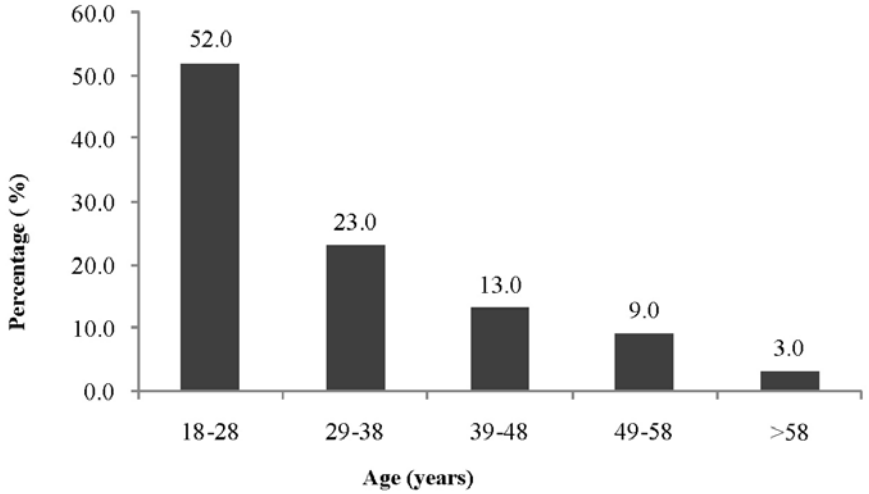

Figure 3. Age distribution of patients with spastic cerebral palsy

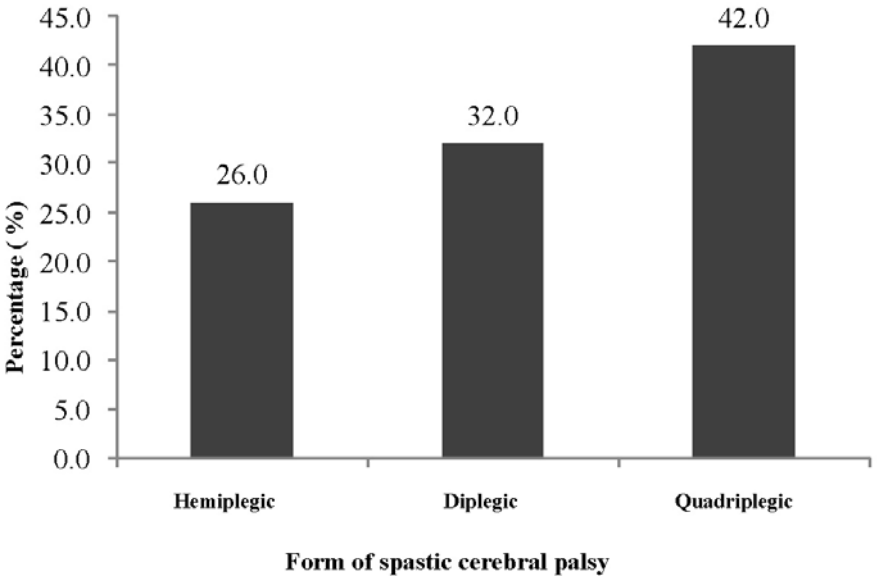

Figure 4. Distribution of patients according to the forms of spastic cerebral palsy

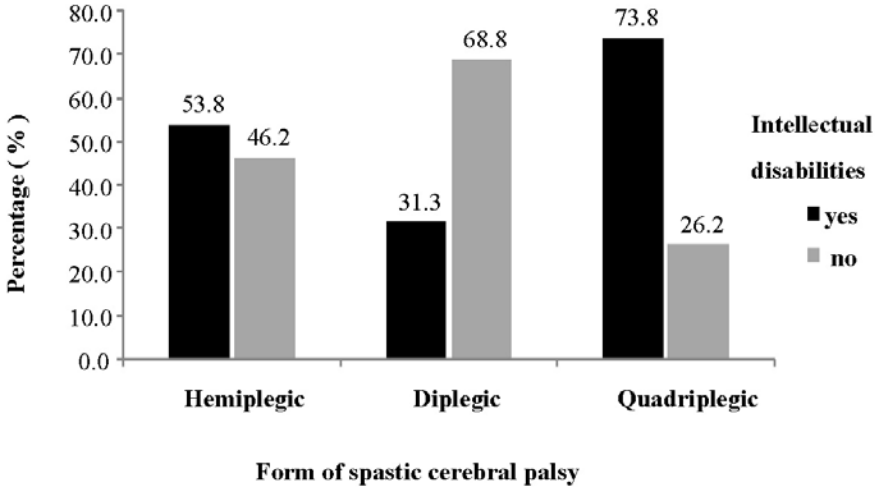

Figure 5. Prevalence of intellectual disabilities in different forms of spastic cerebral palsy

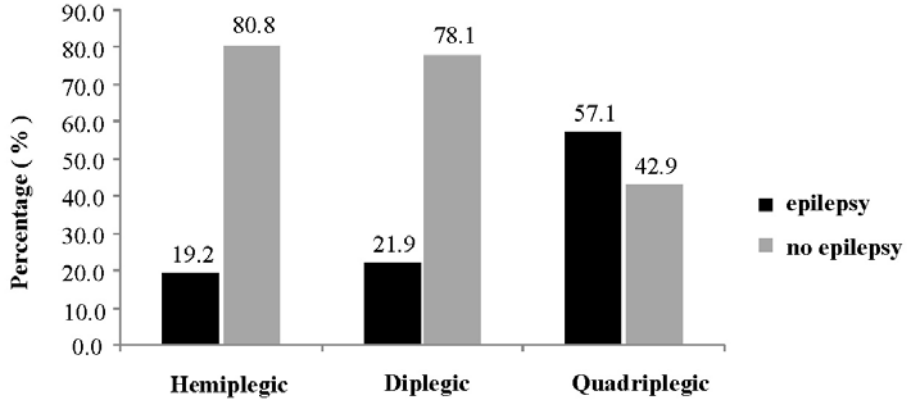

Figure 6. Prevalence of epilepsy in different forms of spastic cerebral palsy

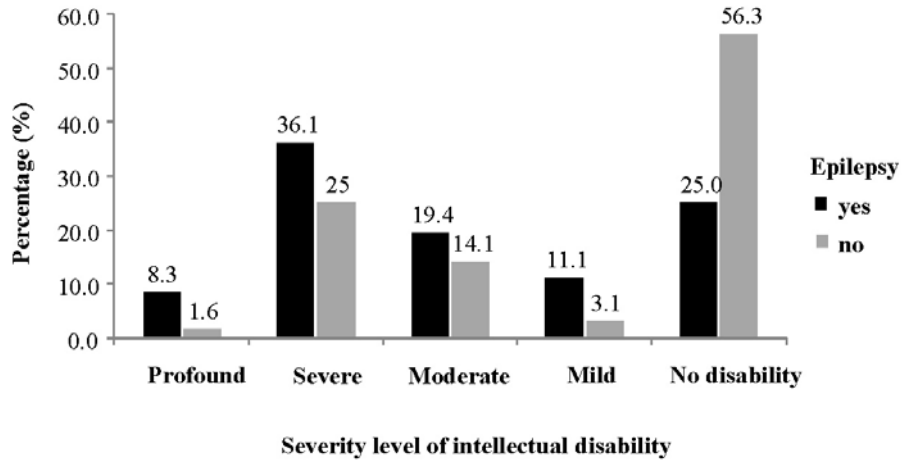

Figure 7. Prevalence of epilepsy in various forms of intellectual disabilties in adult patients with spastic cerebral palsy

DISCUSSION

Our research goal was to determine the prevalence of intellectual disabilities and epilepsy in adults with spastic cerebral palsy and to find out the frequency of epilepsy in various forms of intellectual disabilities in selected group of patients.

Regarding the age, the predominance of 18-28 age group was recorded, since more than a half (52%) of patients belonged to the youngest age group, what was statistically significant. Since CP is predominantly congenital disorder affecting mainly children, it is expected that the majority of adult patient were of younger age. The investigated group was consistent considering the distribution of CP different forms, since all forms were dispensed equally what made the analysis of patients more accurate. Intellectual disabilities were present in more than half (55%) of the patients with spastic CP, what coincides with the results obtained by the research from Spain (Bringas-Grande et al. 2002). Intellectual disabilities were most obvious in quadriplegic (73.8%), than in hemiplegic (53.8%) and diplegic (31.3%) patients. The difference among the groups was statistically significant, making quadriplegic patients more prone to intellectual disabilities. Therefore, the most serious form of spastic cerebral palsy (quadriplegia) was more frequent in patients having intellectual disabilities. These results were not much different from those obtained in other studies. Accordingly, Cioni G, et al, demonstrated that the great majority (82.5%) of subjects in their series with tetraplegia also had abnormal mental development (Cioni et al. 2008). However, our results indicate that diplegia, as less severe form of palsy appeared more frequently in patients with no intellectual disabilities (68.8%).

It was recorded that 36% of patients diagnosed with spastic CP also have epilepsy in our series, which is slightly more than obtained in a similar survey by Okumura, et al. (Okumura 1999). The study of children with CP showed that the 35% of them were also diagnosed with epilepsy (Singhi et al. 2003), what corresponds with our results on adults. Nevertheless, it is obvious that the prevalence of epilepsy in CP patients is significantly higher when compared to its prevalence in general population, which is 1% (Škrapa 2003). The majority of quadriplegic patients (57.1%) in our series were also diagnosed with epilepsy, which occurred less frequently in diplegic (21.9%) and hemiplegic (19.2%) patients, what was similar to other results from the literature (Andersson & Mattsson 2001). Hence,

quadriplegic patients were more prone to developing epilepsy when compared to those with other less serious forms of CP.

Intellectual disabilities were found in 75% of patients with CP and epilepsy in our series which was much higher than in a similar survey in which intellectual difficulties in adults were present in 40% of cases (Robertson 2015). Comparing this to the results from literature, it is obvious that in patients having epilepsy without CP, intellectual disabilities were less present (in 25.5% of patients) (Evenhuis et al. 2000). Among the patients with epilepsy in our series, the highest percentage belonged to those with a severe level of intellectual disabilities (36.1%).

Comparing these results with the prevalence of intellectual disabilities in general population that is less than 1% (Škrapa 2003), a connection between intellectual disability and CP is seen. Accordingly, one can conclude that if there is an increased prevalence of epilepsy recorded in patients with CP, intellectual disability will also increase substantially. The most likely explanation of this may be an overlap in the etiology of CP, epilepsy and intellectual disabilities, since certain similar risk factors contribute to the emergence of all these disorders. Additionally, the motor difficulties, and any intellectual disabilities, as well as seizures, are likely to stem from the same underlying pathology (Jekovec-Vrhovšek 2012). Hence, a severe level of intellectual disability was often manifested in quadriplegic patients having epilepsy in our series.

Since the prevalence of such disorders in CP patients may differ considering their age, we have speculated that it may be decreased in adult CP patients in comparison to children due to brain synaptic plasticity and as a result of successful restorative rehabilitation.

At the end of this paper we would like to underline the possible original scientific achievement of this study deriving from the fact that this was a research aimed at adult CP patients, while most of similar research is dealing with children and younger adult population. The fact that adult CP patients may benefit from restorative rehabilitation due to brain synaptic plasticity that is also happening in adults, may be one of the original contributions of this paper too.

We would also like to mention some limitations of the study stemming from its retrospective character, as well as relatively small number of available publications dealing with adult spastic CP, making our results difficult to compare with the results of others. Therefore, additional research is needed to confirm our findings.

CONCLUSIONS

Intellectual disabilities and epilepsy most frequently occurred in adult patients with most severe forms of spastic cerebral palsy such as quadriplegia. Epilepsy is strongly correlated to the particular severity level of intellectual disability in such patients. Therefore, adult patients suffering spastic cerebral palsy require additional special modes of treatment and restorative rehabilitation to improve the functional outcome, placing heavy burden on health care, educational, and social systems of the entire community.

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Contribution of individual authors:

Mladenka Vukojević: Conception and design, Analysis and interpretation of data;
Tomislav Cvitković: Acquisition of data
Bruno Splavski: Drafting the article, critical revision of the article;
Zdenko Ostojić: Analysis and interpretation of data;
Darinka Šumanović-Glamuzina: Critical revision of the article;
Josip Šimić: Participated in literature searches.

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