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SELF-ASSESSMENT AND PARENTAL ASSESSMENT OF QUALITY OF LIFE CHILDREN WITH COELIAC DISEASE

SAMOPROCENA I RODITELJSKA PROCENA KVALITETA ŽIVOTA DECE SA CELIJAKČNOM BOLEŠĆU

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Summary: Background: Celiac disease is a chronic autoimmune disease characterized by intolerance to gluten. Treatment is carried out by strict lifelong gluten-free diet. Study aims to determine the quality of life children and adolescents with celiac disease who are on controlled diet, based on self-assessment and proxy assessment by their parents. Methods: The study included 116 children and adolescents diagnosed with celiac disease who are on a gluten free diet for at least one year and the same number of their parents. The children were aged 5 to 18 years. The patients and their parents were recruited at the Institute for Maternal and Childs healthcare of Serbia and the University Children's Hospital in Belgrade. The used instruments were a child and parent version of the Pediatric Quality of Life Inventory and the Screen for Child Anxiety Related Disorder. Results: The results in both versions of the questionnaires Pediatric Quality of Life Inventory showed similar values. ($P > 0.05$, all analyzes). There was a greater value of three scores in instrument named Screen for Child Anxiety Related Disorder (questionnaire for children) compared to (questionnaire for parents). These are the total score ($p < 0.05$), separation anxiety ($p < 0.01$) and the avoidance of the school ($p < 0.05$). Conclusions: Disagreement between the children's selfassessment and parental assessment, justifies the importance of hearing subjective children's opinion on their quality of life as well as of their parents. This research has given us information about some of the critical points by which we can focus future studies.

Key Words: Celiac disease, children, gluten, diet, quality of life.

Sažetak: Celijakija je hronična autoimuna bolest koju karakteriše netolerancija na gluten. Lečenje se vrši striktnom doživotnom dijetom bez glutena. Cilj rada je da se utvrdi kvalitet života dece i adolescenata sa celijakijom koji su na kontrolisanoj ishrani, na osnovu samoprocene i procene od strane roditelja. Metode: Istraživanje je obuhvatilo 116 dece i adolescenata sa dijagnozom celijakije koji su na dijeti bez glutena najmanje godinu dana i isto toliko roditelja. Deca su uzrasta od 5 do 18 godina. Istraživanje je realizovano u Institutu za zdravstvenu zaštitu majke i deteta Srbije i Univerzitetskoj dečjoj klinici u Beogradu. Korišćeni instrumenti su dečija i roditeljska verzija Pedijatrijskog upitnika za procenu kvaliteta života (PedsQL) i Upitnika za pretragu anksioznih poremećaja kod dece (SCARED). Rezultati: Rezultati u obe verzije Pedijatrijskog upitnika za procenu kvaliteta života pokazali su slične vrednosti ($P > 0,05$, sve analize). Rezultati procene prisustva anksioznih poremećaja na dečjoj verziji SCARED skale, pokazuju statistički značajno veću vrednost tri skora u odnosu na procenu njihovih roditelja. To su, Ukupan skor ($p < 0,05$), Socijalna anksioznost ($p < 0,01$) i Izbegavanje škole ($p < 0,05$). Zaključak: Život sa celijakijom i bezglutenskom ishranom može biti težak za decu i adolescente jer se osećaju da su drugačiji od svojih vršnjaka zbog čega redukuju druženja i izoluju se. U ovoj studiji došli smo do saznanja da deca i adolescenti sa celijakijom imaju značajno niži ukupni QOL i to kako na osnovu samoprocene tako i na osnovu roditeljske procene..

Ključne riječi: celijakija, deca, gluten, dijeta, kvalitet života.

INTRODUCTION

Evaluation of the quality of life of the patients in general and therefore patients with celiac disease has become the practice of the past half century [1]. Celiac disease is an autoimmune disorder caused by the ingestion of gluten in persons who have a hereditary predisposition for this disease. It is a chronic, multi-organ disease in which is present a different degree of damage of the small bowel mucosa and the different spectrum of clinical symptoms [2]. Chronically ill children are more prone to physical, psychological, social problems than healthy children and that can affect the child's quality of life in relation to health in a negative way [3]. Early diagnosis of celiac disease reduces complications and contributes to a positive attitude towards the disease. Strict lifelong gluten-free diet with no peptides of wheat, barley, oats and rye is the only known treatment method [4]. Gluten-free diet can cause problems in the acceptance by the child and his family [5]. Children who are on a gluten free diet for a long time, often break the diet during their adolescence. An important factor of disrespect of restrictive diet in adolescence is a peer pressure, unclear labeling of food and bustling lifestyle of teenagers [6]. Research shows, that it is important for parents to accept the illness of their children because it has a positive impact on a child to accept its own disease easier [7]. Some studies confirm the high incidence of psychological problems such as anxiety, as problems of adaptation to the social life of children who are on a strict diet without gluten [8]. The problems are particularly pronounced in children of school age. This, because it is a period in which is important for the child not to be different from the other children [9]. Measuring quality of life in children, includes physical and psychosocial aspects, as well as individual health assessment [10]. There are studies that show differences in experience of the health quality of life between parents and their children [11]. Therefore, previous studies indicate that for the assessment of children 's quality of life is the most appropriate to use a double approach, and children's opinion to supplement by the opinion of parents [12]. Therefore, in this study we aim to determine the quality of life of Serbian children and adolescents with celiac disease who are on controlled diet, based on self-assessment and proxy assessment by their parents.

METHODS

Participants

Study subjects were pediatric patients with celiac disease which were treated at the Institute for Maternal and Childs healthcare of Serbia "Dr Vukan Čupić", in Belgrade and the University Children's Hospital in Belgrade and their parents. Participants were children of both sexes, age 5-18, with a diagnosis of celiac disease and gluten-free diet regime that lasted for at least a year. The exclusion criteria were the inability to read and write Serbian (for 8-year-olds and parents), severe neurological impairments, intellectual disability, and parental or children's unwillingness to cooperate. Subjects were recruited consecutively as they came to a gastroenterologist for a regular celiac disease follow-up visit and participation was on a voluntary basis. Parents and children older than eight years completed questionnaires by themselves, separately and under the supervision of one of the authors. Children age 5-8 completed questionnaires after their parents read and explained them questions.

Description of used instruments

Pediatric Quality of Life Inventory TM – PedsQL TM is a generic questionnaire for assessing the quality of life (QOL) of children and adolescents aged 5-18 years. There are versions for three age groups of children (age 5-7, 8-12 and 13-18) and parent version. Each version has 23 questions sorted into four scales, and answers are graduated from 1-5. The scaling of the questions took place through the Likert scale, which consists of 5 points: 0 - never, 1 - almost never, 2 - sometimes, 3 - often, 4 - always. Scales are Emotional functioning (8 questions), Social functioning (5 questions), School functioning (5 questions) and Physical functioning (5 questions). All answers were reverse scored and linearly transformed to a 0-100 scale. Higher scores indicate better HRQOL. If more than 50% of the answers were missing, the result was not counted, and if 50% or more questions were answered, the median score on the scale was calculated. PedsQL is one of the most comprehensive QOL assessment questionnaires, with a high level of reliability, validity and sensitivity measurement. Conceptually, PedsQL measures general well-being and functioning in everyday life, regardless of the child's health. Screening for anxiety disorders in children (Screen for Child Anxiety Related Disorder -

SCARED) version for a child and a version for parents, was used to assess a degree of anxiety in children and adolescents aged 5-18. SCARED contains 41 questions, with answers graduated 0-1-2 ("incorrect" - "partly true" - "true"). The questions are classified in the scales: Generalized anxiety (8 questions), Separation anxiety (8 questions), Social anxiety (7 questions), Avoiding school (4 questions) and Panic-somatic disorder (13 questions). The sum of certain questions represents the total scoring scale, and the sum of all questions is the total SCARED score (range 0-82). A greater score indicates the presence of a certain anxiety problem in a higher degree. If the total SCARED score ≥ 25 is considered to be an anxiety disorder at a clinically significant level requiring professional assistance. SCARED is a reliable, valid and sensitive questionnaire. SCARED is a comprehensive questionnaire for the self-assessment of anxiety disorders, which includes the symptoms of all anxiety disorders, which can occur in the child and adolescent periods [13,14].

Ethical considerations

The study was approved by the Ethics Committee of the Institute for Maternal and Childs healthcare of Serbia "Dr Vukan Čupić", in Belgrade and the University Children's Hospital in Belgrade. Respecting ethical requirements, all respondents were informed in writing form with the basic objectives of the research and information that the obtained data will be used exclusively for scientific purposes, as well as to

guarantee the anonymity of all received data and the identity of the respondents.

Statistical Analysis

The descriptive statistical measures, such as percentage, median, arithmetic mean, minimum-maximum format range, standard deviation and interval of 95% confidence with the upper and lower limits were used.

Student's t-test was used to compare the results between independent groups on an uninterrupted scale. Cohen's effect size corrected for sample size (d) as a measure of the magnitude of the impact, was computed and was interpreted as small (0.20-0.50), as moderate (0.51-0.80) or as high (over 0.81). All P-values < 0.05 was considered statistically significant. The χ^2 test was used to determine whether there is a significant difference between the expected frequencies and the observed frequencies in one or more categories. The analysis and data processing were performed using a software package for statistical analysis (Statistical Package for the Social Sciences - SPSS for Windows, version 23.0, 2015).

RESULTS

The study included a total of 116 children and adolescents aged 5 to 18 who have a diagnosis of celiac disease and 116 parents (one parent of each child). Demographic and clinical features of patients are given in Table 1.

Tabela 1. Opšte demografske karakteristike grupa

Table 1. General demographic characteristics of the groups
Tabela 1. Opšte demografske karakteristike grupa

Parameters	Coeliac Disease (n = 116)
Age (years), mean (SD)*	11.40 (3.91)
Age group, n (%)**	
young children, 5-7 years	22 (19.0)
children, 8-12 years	44 (37.9)
adolescents, 13-18 years	50 (43.1)
Gender, n (%)**	
male	39 (33.6)
female	77 (66.4)

* t-test, $p = 1.000$.

** χ^2 test, $p = 1.000$.

The parent who accompanied a child to the medical institution for a follow up examination, was filling a questionnaire. During this study, 88 (76%) of mothers has accompanied the child to the follow up examination and 28 (24%) of fathers Table 2.

Tabela 2. Struktura uzorka ispitivanih roditelja
Table 2. Structure of the sample by informants (Mothers or Fathers)

Informant	n	%
	Mother	88
Father	28	24,00

Note: Number of respondents (n = 116);

Results of PedsQL scores (questionnaire for children) and PedsQL (parent questionnaire) (p <0.05 All analyzes) were similar Table 3.

Table 3. Komparacija rezultata PedsQL (upitnik za dete) i PedsQL (upitnik za roditelja) u grupi ispitanika sa celijakijom

Table 3. Comparison of the assesment on Health related Quality of Life by children and their parents in PedsQL (version for child and parent).

PedsQL	Q.	M	SD	t (df)	p	D.AM	95% CI diff.		d
							LL	UL	
In whole	C	75,89	20,35	0,736 (115)	0,463	1,08	-1,83	3,98	0,10
	P	74,81	20,53						
Physical funct.	C	76,75	25,31	1,748 (115)	0,083	3,96	-0,53	8,45	0,23
	P	72,79	27,84						
Emot. funkc.	C	71,29	25,01	1,388 (115)	0,168	3,28	-1,40	7,95	0,18
	P	68,02	27,62						
Social funct.	C	81,55	24,02	-1,671 (115)	0,098	-3,13	-6,83	0,58	0,22
	P	84,68	22,07						
School funct.	C	73,81	24,04	-0,948 (112)	0,345	-1,86	-5,74	2,03	0,12
	P	74,91	28,97						
Psychosoc. funct.	C	74,91	21,31	-0,516 (115)	0,607	-0,80	-3,89	2,28	0,07
	P	75,72	20,93						

Note: Q. - Questionnaire; C - for a child; P - for parent

There is significantly higher value of three scores on SCARED (version for children) compared to SCARED (questionnaire for parents) was found. These are the total score (p <0.05), separation anxiety (p <0.01) and the avoidance of the school (p <0.05). The values of Cohen's d coefficient show that the biggest impact of differences in score Separation anxiety (d = 0.40), and lowest in score Avoidance school (d = 0.26) Table 4.

Table 4. Komparacija rezultata SCARED (upitnik za dete) i SCARED (upitnik za roditelja) u grupi ispitanika sa celijakijom

Table 4. Comparison of the emotional status assesment by children and their parents in SCARED (version for child and parent)

SCARED	Q.	M	SD	t (df)	p	D.AM	95% CI diff.		d
							LL	UL	
In whole	C	20,79	17,96	2,386 (114)	0,019	1,67	0,28	3,06	0,31
	P	16,79	14,22						
Panic-somatic dist.	C	5,77	6,41	1,432 (114)	0,155	0,71	-0,27	1,70	0,19
	P	4,07	4,83						
Generalized anxiety	C	5,00	4,39	0,267 (114)	0,790	-0,11	-0,72	0,95	0,04
	P	4,25	3,97						
Separation anxiety	C	3,62	3,57	3,030 (114)	0,003	1,09	0,38	1,80	0,40
	P	3,47	3,28						
Social anxiety	C	4,95	3,70	1,276 (114)	0,204	0,27	-0,15	0,69	0,17
	P	3,83	3,22						
School avoiding	C	1,45	1,94	1,993 (114)	0,049	3,85	0,02	7,68	0,26
	P	1,17	1,67						

Note: Q. - Questionnaire; C - for a child; P - for parents;

DISCUSSION

Each chronic disease and thus celiac disease, which if occurs in children during the period of their growth and development can affect the psychosocial development, self-perception, self-esteem, social functioning, and finally in a few studies have been found to significantly affect health related quality of life of children and adolescents [15]. In our study, children and adolescents with celiac disease and their parents gave similar answers in the pediatric questionnaire about quality of life (PedsQL), so all observed scores statistically were homogeneous. Our results are consistent with the results of European studies in which the evaluation of self-assessment of the quality of life of children and adolescents with celiac disease and assessment of their parents shows that the disease does not have significant negative impact on quality of life [16]. Unlike our results, other studies have found that parents have significantly poorer perception of health related quality of life of their children than the

children themselves [15,17]. In our study, comparing the assessment of children and adolescents with celiac disease with the assessment of their parents on screening emotional problems, we get a significantly higher value of three scores SCARED (questionnaire for the child in relation to the SCARED (questionnaire for parents). These are the total score ($p < 0.05$), separation anxiety ($p < 0.01$) and the school avoidance ($p < 0.05$). The results of several studies suggest that children and adolescents with celiac disease recorded high incidence of psychological problems such as anxiety, depression, fears, feelings of sadness and low tolerance for frustration, emotional hypersensitivity, shyness, irritability and showing problems of adaptation to social life. Children and adolescents avoid socializing with peers, they abstain to take meals with others outside the home and therefore can alienate [18,19]. Since the school environment and peers are having very important role in the normal development and functioning of children,

adaptation to school work and peer groups is a significant indicator of the functioning of children and adolescents who have a chronic disease and are on specific dietary treatment. Due to the specific disease and strict diets, children and adolescents absent from school, missing several weeks of classes and lose contact with their peers and peer activities and interactions. Results of some studies that have had to examine the impact of chronic disease on the behavior of children, show that the examined children present concern about the absence from school, separation from peers, the feeling of diversity, increased need for care and helping loved ones [20]. Schools and teachers should work with parents and to support children with these and other problems. It is necessary that teachers and peers improve their knowledge about celiac disease because children spend a large part of their time in schools under their control and interaction. The results of our study are consistent with previous studies that have dealt with this theme, showing that children and adolescents who suffer from celiac disease lack psychological support to overcome the problems they face [21, 22].

CONCLUSION

Incomplete agreement between self and proxy-assessment justifies the importance of

considering the opinion on quality of life from both, children and their parents. Respect of gluten-free dietary regime is an essential factor for obtaining optimal quality of life in relation to health. Children with celiac disease need the support of their families in respecting of gluten-free diet. It is necessary to provide psychosocial and educational support for children and adolescents who have difficulty to strictly adhere to the regime gluten-free diet and this is a task not only for parents but for teachers and health workers.

Conflict of Interest The authors declare that there is no conflict of interest

ZAKLJUČAK

Nepotpuna usaglašenost između samoprocene i roditeljske procene kvaliteta života dece sa celijakijom, opravdava značaj i potrebu, uzimanja u obzir mišljenja o ovome i od same dece i njihovih roditelja. Poštovanje dijetalnog, bezglutenskog režima ishrane je osnovni faktor za postizanje optimalnog kvaliteta života u vezi sa zdravljem. Deci i adolescentima sa celijačnom bolešću neophodno je obezbediti psihosocijalnu i obrazovnu podršku u striktnom pridržavanju bezglutenskog režima ishrane a to je zadatak ne samo za roditelje već i za nastavnike i zdravstvene radnike.

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