Digestive and Liver Disease 47 (2015) 927–932

Contents lists available at ScienceDirect

Digestive and Liver Disease

journal homepage: www.elsevier.com/locate/dld

Alimentary Tract

Quality of life in children with celiac disease: A paediatric cross-sectional study

Chiara Biagetti[a,*], Rosaria Gesuita[b], Simona Gatti[a], Carlo Catassi[a]

[a] Department of Pediatrics, Università Politecnica delle Marche, Ancona, Italy
[b] Centre of Epidemiology, Biostatistics and Medical Information Technology, Università Politecnica delle Marche, Ancona, Italy

ARTICLE INFO

Article history:
Received 21 January 2015
Accepted 16 July 2015
Available online 26 July 2015

Keywords:
Child
Gluten-free diet
Health-related quality of life

ABSTRACT

Background: Few studies investigated factors influencing the quality of life of children with celiac disease on a gluten-free diet.
Aims: To investigate the impact of the gluten-free diet on the psycho-physical well-being of celiac children.
Methods: In this cross-sectional study, we interviewed 76 celiac and 143 non-celiac children (2–18 years) by using a non-disease specific questionnaire (Pediatric Quality of Life Inventory Test) and we explored the impact of the diet on social life with an open-ended questionnaire. Scores were compared by Wilcoxon rank-sum test. A quantile regression analysis was used to evaluate the impact of celiac disease on score distribution.
Results: No significant differences in quality of life were found between the two groups (total score: 84.1 (81.1–87.2) vs 81.5 (79.7–83.4), median (95% CI), patients and controls respectively, $p = 0.4$). Treatment positively affected quality of life in children that showed "intermediate" scores in the Pediatric Quality of Life Inventory Test. Lowest scores were observed in children reporting a higher number of diet difficulties or co-morbidities.
Conclusions: Although celiac patients showed an overall good quality of life in comparison with a control group, by using appropriate analytical methods we elicited specific factors contributing to a lower quality of life in patients, such as co-morbidities and difficulties with the diet.

1. Introduction

Celiac disease (CD) is a permanent autoimmune disorder triggered by the ingestion of gluten in genetically predisposed individuals. Gluten is a protein complex found in cereals that are diffusely consumed in most countries, i.e., wheat, rye and barley. The clinical spectrum of CD is widely variable, ranging from severely ill patients complaining of both gastrointestinal and extra-intestinal symptoms, to completely symptom-free subjects [1]. Although CD can be successfully managed by strict adherence to a gluten-free diet (GFD), the dietary treatment limits the patients' food choice and influences the patient lifestyle and quality of life (QOL).

Recently the interest on how patients perceive the impact of chronic diseases and how their health state is modified by the therapeutic intervention is gaining increasing attention in medical care settings. Health-related QOL (HRQOL) is a multidimensional concept including physical, emotional, social and cognitive domains. What matters in HRQOL is the way patients feel about their functioning, not their functioning itself [2]. Considering the partially subjective nature of many symptoms experienced by subjects suffering from CD and the strong impact on social and personal life of adhering to a controlled dietary regimen, the perceived quality of life and overall well-being of each individual must be taken into account, when addressing CD patients' needs.

Several studies have been performed in adult CD patients aiming at investigating their HRQOL either at diagnosis and when dietary restrictions start, using unspecific QOL instruments [3–8]. In children, as well as in adults, a chronic illness, like CD, may interfere with daily life and impact QOL. So far only few studies have investigated HRQOL in celiac children, reporting conflicting results and

* Corresponding author at: Department of Pediatrics, Università Politecnica delle Marche, Via Corridoni, 11, 60123 Ancona, Italy. Tel.: +39 071 5962370; fax: +39 071 36281.
E-mail addresses: chiara.biagetti@ospedaliriuniti.marche.it, chiara.biagetti@libero.it (C. Biagetti), r.gesuita@univpm.it (R. Gesuita), simona.gatti@hotmail.it (S. Gatti), c.catassi@univpm.it (C. Catassi).

http://dx.doi.org/10.1016/j.dld.2015.07.009
1590-8658/

using different instruments [9–12]. By using a qualitative method of research, the Critical Incident Technique, we recently reported on the impact of the GFD on daily life of children with treated CD. We depicted several dilemmas experienced by children with CD in everyday life (specific emotions, relational difficulties and daily life managing issues), suggesting that children with CD experience strong emotions related to the need of adhere to the GFD, permeating several aspects of everyday life and deeply influencing their QOL [13]. The variability of results yielded by different questionnaires and the insufficient data about QOL in children with treated CD need further investigations. In order to systematically investigate the impact of CD on the HRQOL in children, we performed a cross-sectional study comparing a large group of CD children (enrolled in a tertiary level Pediatric Gastroenterology Center) to a group of non-celiac children. For this purpose, we chose a validated, easy and widely used questionnaire, tailored for paediatric patients and their parents.

2. Materials and methods

2.1. Patients

Between March 2007 and March 2011, eighty children (aged 2–18 years) with biopsy-proven CD, on a GFD from at least one year, were consecutively recruited in this study at the Gastroenterology Outpatient Clinic (Department of Pediatrics, Università Politecnica delle Marche, Ancona, Italy). Children under 2 years of age and/or affected by type 1 diabetes (T1D) were excluded. One hundred and fifty non-celiac healthy children were screened as controls, during their annual paediatric visit in three general paediatric clinics in the same area (Ancona, Italy). Both celiac and controls parents were informed about the study protocol and a written informed consent was obtained. The study protocol was approved by the Ethical Committee of the Università Politecnica delle Marche, Ancona, Italy.

2.2. Methods

The Pediatric Quality of Life Inventory (PedsQL™, Italian version) designed by J. Varni [14] was used to investigate QOL in both patients and controls. This is an unspecific 24-items self-reported questionnaire with proven validity and reliability in paediatric patients. As previously reported [14], the questionnaires were filled in by children older than 8 years (paediatric version) or by parents of children younger than 8 years (proxy version) and handed out at the time of the visit. Items were divided into 4 domains: physical functioning, emotional functioning, social functioning, school functioning. Raw scores in a 5-point frequency Likert scale (0 = never a problem; 1 = almost never a problem; 2 = sometimes a problem; 4 = almost always a problem) were reverse-scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0), with higher scores indicating better HRQOL. Then the Physical Health Summary Score (average of scores obtained by each subject in the physical functioning domain), the Psycho-Social Health Summary Score (average of scores obtained by each subject in the emotional functioning, social functioning and school functioning domains overall), and the total score (average of scores obtained by each subject considering all the sub-dimensions of the questionnaire) were calculated, with higher scores indicating better health and QOL. In order to explore more in details the impact of the diet on social life, three further aspects were investigated in both groups: (1) friends' parties attendance, (2) school canteen's attendance, (3) frequency of being in an angry mood or in a difficult situation in the 7 days preceding the enrolment (defined as number of disappointing events/week) and related reasons. A 5-point frequency Likert scale was used, as appropriate.

The result of serological markers (IgA anti-transglutaminase) performed in the 3 months before the enrolment was used as a measure of adherence to the GFD.

2.3. PedsQL™ data statistical analysis

Since QOL scores showed an asymmetric distribution confirmed by non-normality tests, a non-parametric approach was chosen for all the analysis. The Wilcoxon rank-sum test or Kruskal–Wallis test were used to compare quantitative variables between groups; medians and 95% confidence intervals (95% CI) and 25th–75th percentiles were used to summarize data. Chi-square test was applied to categorical variables and results were expressed as absolute and percentage frequency distribution.

Quantile regression was used to evaluate the impact of CD on HRQOL scores (adjusted for subject characteristics) and to assess factors affecting QOL in CD subjects [15]. Quantile regression is a non-parametric method, particularly suitable for variables that are not normally distributed because it does not impose modelling assumptions. Besides, it allows to analyze the effect of an exposure on different parts of the outcome distribution, providing a more complete view with the estimation of quantile-specific effect. In other words, by using this technique, it is possible to describe the impact of the exposure not only on the centre (the median) but also on every part of interest of the outcome distribution (i.e. the tails, the lowest or the highest quantiles). The nine deciles of the score distribution were considered in the analysis. Results for each HRQOL domain were graphically summarized, where the x-axis shows the values of the 9 deciles of the response distribution (that is the scores of HRQOL domains of PedsQL™) and the y-axis shows the effects of CD on QOL for each decile (continuous line) and 95% confidence bands (dotted lines). If the dotted lines do not cross the zero line, that is the 95% confidence bands do not include the zero value, the estimates significantly differ from zero.

All the analysis was performed using the R statistical package; a level of probability of 0.05 was used to assess the statistical significance.

3. Results

Eighty children with CD and 150 controls were initially invited to participate. Four CD patients were excluded, due to lack of parental consent in three cases and because of co-morbidity (T1D) in one case; seven controls were excluded because of lack of parental consent. Overall PedsQL™ was eventually administered to 76 CD subjects and 143 controls. In CD group median age at diagnosis was 3.5 years (25th–75th percentile: 1.9–7.0) and median duration of GFD was 3.9 years (25th–75th percentile: 1.7–5.4). Forty-three percent (95% CI: 32.1–55.3) were symptomatic at diagnosis (typical CD, i.e. abdominal distension, vomiting, diarrhoea, weight loss). Only two patients had slightly elevated serological markers detected in the three months preceding the enrolment.

Table 1 shows the demographic characteristics of enrolled subjects and the results of the psycho-social evaluation of CD patients and controls (PedsQL™ median scores). Overall no significant differences were found in Physical Health, Psycho-Social Health and Total PedsQL™ Scores between CD patients and controls. CD subjects experienced a significantly higher number of disappointing events in a week ($p = 0.009$) and more frequently did not attend school canteen ($p < 0.001$). Thirty-nine percent of CD patients reported at least one disappointing event related to the GFD.