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Recent Progress in Nutrition

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Case Report

The Low FODMAP Diet for Children with Inflammatory Bowel Disease and Overlapping Functional Gastrointestinal Symptoms – a Case Series

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Academic Editor: Arrigo F. G. Cicero

Special Issue: Nutrition and Nutritional Management of Inflammatory Bowel Disease

Recent Progress in Nutrition
2023, volume 3, issue 2
doi:10.21926/rpn.2302009

Received: February 11, 2023
Accepted: June 26, 2023
Published: June 29, 2023

Abstract

The low FODMAP diet (LFD) is a dietary intervention developed for and used particularly for the management of functional GI symptoms (FGIS) in adults with irritable bowel syndrome (IBS). It has also been proposed to improve coincident functional gut symptoms in adults with inflammatory bowel disease (IBD). This report describes the outcomes of the LFD in a series of children with IBD with concurrent FGIS. This study included children aged between 7-14 years with quiescent IBD and overlapping FGIS. All participants received three dietetic consultations in line with best practice recommendations. The IBS Global Improvement Scale was used to assess symptom outcomes. Mean (standard deviation [SD]) and number (percentage) were calculated for continuous and categorical data. Seven (77.7%) of the nine children experienced GI symptom improvement after completing the LFD for four weeks. All seven children had recurrence of symptoms in response to challenge with fructans and three also experienced symptoms of lactose intolerance. The LFD may be considered as a dietary option to help alleviate overlapping FGIS in children with quiescent IBD when provided by a

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qualified paediatric gastroenterology dietitian. Further studies are now required to support these findings.

Keywords
Inflammatory bowel disease; Low FODMAP diet; paediatrics; functional gastrointestinal symptoms; dietetics

1. Introduction

Fermentable oligosaccharides, disaccharides, monosaccharides, and polyols (FODMAPs) are short-chain carbohydrates that when incompletely absorbed in the proximal gastrointestinal (GI) tract can be fermented by colonic bacteria, leading to increased osmotic activity and excess gas production [1, 2]. Consequently, this causes gut symptoms including alternating stool patterns (diarrhoea and constipation), abdominal pain, bloat/distention and less frequently, upper GI symptoms including reflux and nausea. The low FODMAP diet (LFD) is an extensively described dietary intervention used particularly for the management of functional GI symptoms (FGIS) in adults with irritable bowel syndrome (IBS) [1]. The use of the LFD has also been proposed as a therapeutic option to improve coincident FGIS in adults with inflammatory bowel disease (IBD) [3-7].

IBD is an idiopathic disease that features active and chronic inflammation in the GI tract [8]. The two main phenotypes of IBD are Crohn's disease (CD) and Ulcerative colitis (UC). Symptoms of IBD may include abdominal pain, weight loss, malnutrition, joint pain and diarrhoea [8]. In children, ongoing symptoms and uncontrolled inflammation can directly impact upon growth, development and quality of life. Given that both CD and UC are considered incurable, management options focus upon establishing control of inflammation and maintenance of remission over time [9]. Dietary interventions are also important in the management of IBD. These approaches are especially relevant in children, during times of rapid growth and developmental change.

A recent meta-analysis, including two randomised controlled trials and four prospective clinical studies, showed that the use of the LFD resulted in a reduction in the severity of GI symptoms in adults with IBD who also experienced diarrhoea predominant IBS, abdominal pain, bloating or nausea, but not in those with constipation [10]. There are currently no data on the use of the LFD in children with quiescent IBD. The aim of this report was to describe symptom outcomes in a series of children with IBD and concurrent FGIS treated with a LFD.

2. Case Report

2.1 Methods

2.1.1 Patient Group

This case series included children aged 7 to 14 years with quiescent CD or UC, determined by the child's most recent blood inflammatory markers, faecal calprotectin, disease activity scores [11], and overlapping FGIS consistent with the NICE guidelines for IBS [12]. Children were referred to a

paediatric dietitian by a paediatric gastroenterologist between 2017-2022 for LFD education and monitoring. Background details, including patient characteristics, medications, disease location, current disease activity and medical history, were obtained from online medical records. The use of partial enteral nutrition was permitted (low FODMAP formula excluding lactose, fructans and fibre); however, children must have been consuming a 'normal/habitual' diet to be included in this case series.

2.1.2 Protocol

Children and/or their families received a total of three dietetic consultations in line with adult best practice recommendations (Figure 1) [13]. Children had an initial dietetic consultation within 3 weeks of the referral being made, a second consultation 3-4 weeks after commencing the restriction phase, and a third consultation after completion of the reintroduction phase. At each consult, a consistent series of questions were asked to determine the frequency and severity of symptoms, assess the child's dietary intake, and the efficacy of the LFD during the reintroduction phase to determine specific carbohydrate intolerance(s). The dietitian monitored adherence to the four-week restriction phase during a weekly phone call.

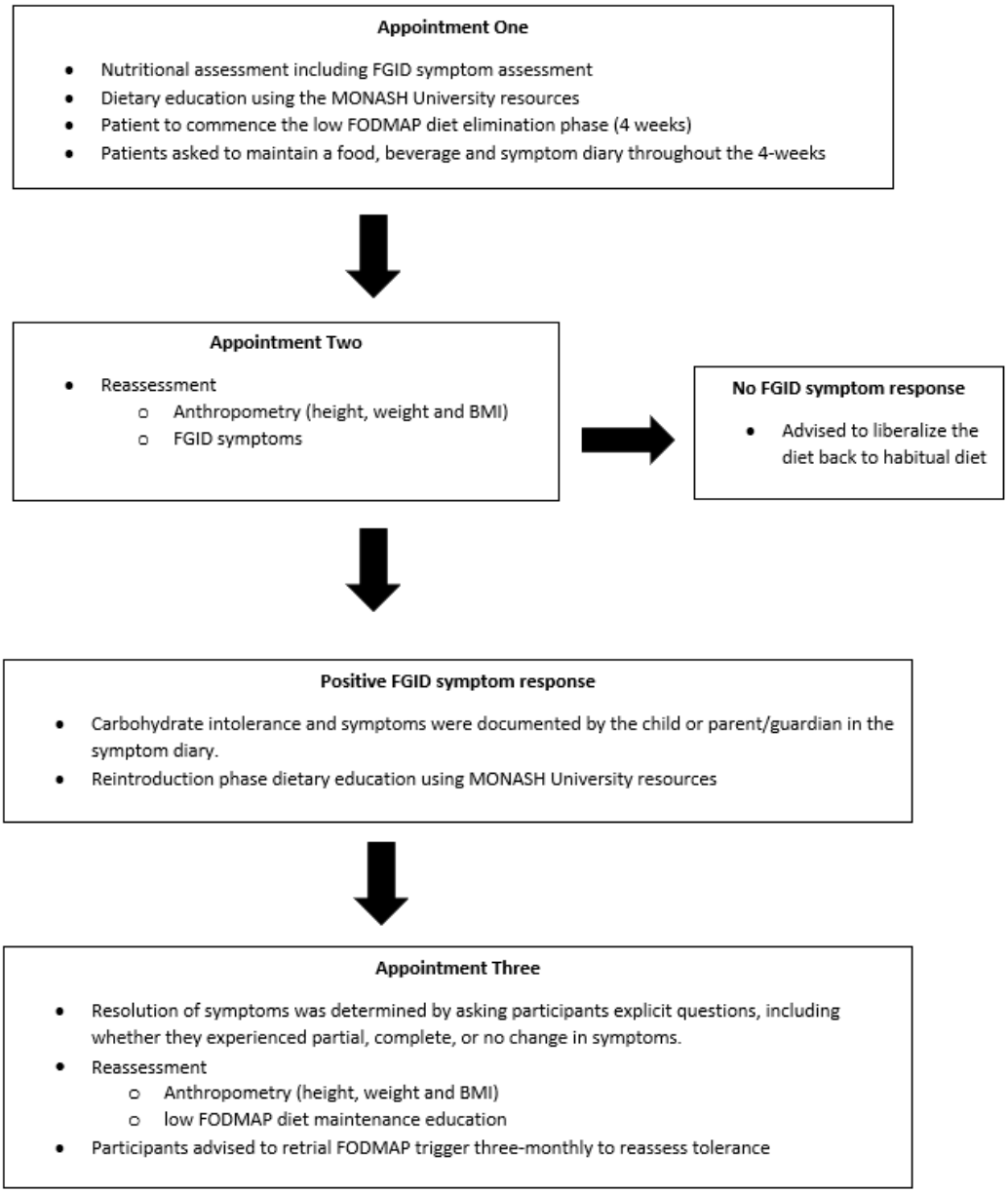

Figure 1 The low FODMAP dietary protocol.

There are currently no published protocols for the use of the LFD in children. The MONASH University, Australia, adult FODMAP resources were adapted for use in children. Sample meal plans were created to replicate the average child's lunch box in alignment with the New Zealand (NZ) Ministry of Health's Healthy Eating Guidelines for Children and Young People [14].

2.1.3 Anthropometry

Height and weight were measured at the initial and second consultations using the same clinic scales and stadiometer. Weight was measured using a bariatric scale (SCALE 954, SECA, New Zealand), and height was measured using a wall-mounted stadiometer (SECA 264, SECA, New Zealand), in light-weighted clothes. Body mass index (BMI) was calculated from weight and height measurements and reported as z-scores [15].

2.1.4 Gastrointestinal Symptom Response

GI symptom response was assessed using subjective measures, including physician documentation in patient notes based on the NICE guidelines [12], standardized questions asked by the dietitian during consultations, and patient symptom diaries. Carbohydrate intolerances were determined subjectively during the LFD reintroduction phase based on any symptom exacerbation following the carbohydrate challenges. Any GI symptoms that occurred after consuming a predetermined quantity of a specific FODMAP were recorded by the participant/parent in his or her symptom diary as the presence or absence of symptoms. Patients were asked to provide a detailed description of any symptoms they experienced in their food diary provided. Ethical approval was granted by the University of Otago Ethics Committee (Health) HD17/168.

2.1.5 Outcome Measures

Effectiveness of the intervention was measured using a symptom response survey via telephone one week after the third and final dietetic consultation [16]. A modified version of the validated eight-item IBS Global Improvement Scale questionnaire was utilised to assess symptom outcomes [16]. Data from the retrospective telephone survey were collapsed to provide clinically meaningful data. Symptom and magnitude of improvement data were collapsed into dichotomous responses of "improved" (moderately improved, substantially improved) or "not improved" and "worsened/no change" (no change, moderately worse, substantially worse), "moderately improved," and "substantially improved" respectively. Satisfaction responses were collapsed into agree and disagree responses, and a composite score was calculated to determine overall subjective diet efficacy. The patients and/or their parent were asked to rate symptom changes for bloating, abdominal pain/discomfort, diarrhoea and nausea using a 7-point Likert scale (substantially worse, moderately worse, no change, moderately improved, substantially improved, or never had the symptom) [16].

Patients were also asked to rate their satisfaction with symptom response and the LFD. These statements were scored using a 5-point Likert scale (strongly disagree, disagree, neutral, agree, and strongly agree). All questionnaires were completed anonymously and confidential. To reduce any response bias, patients were encouraged to answer questions honestly.

2.1.6 Statistical Analysis

All data were analysed using Microsoft Excel 2018. Mean (standard deviation [SD]) and number (percentage) were calculated using for continuous and categorical data, including baseline characteristics, anthropometry, GI symptoms, and outcomes. Using the post-intervention telephone survey and the subjective outcomes pre- and post-LFD completion, the differences for symptom response and satisfaction were calculated.

3. Results

3.1 Participant and Baseline Characteristics

Nine children (7 female) with quiescent IBD (7 CD) completed the LFD (Table 1). The mean number of years since diagnosis was 3.1 (1.3-3.5). Inflammatory markers including CRP and faecal

calprotectin (FC) were normal except for one child having mildly elevated FC at baseline (67 ug/g).

Table 1 Participant characteristics and demographics.

	IBD (n = 9) Mean [SD] or n [%]
Phenotype	
Crohn disease (CD)	7 (77.7)
Ulcerative colitis (UC)	2 (22.2)
Age, years	11.6 (2.12)
Gender,	
Male	2 (22.2)
Weight z-score	0.44 (1.24)
Height z-score	0.56 (1.09)
BMI, kg/m^2 z-score (range)	0.58 (-0.66 – +0.99)
Disease location (Montreal Classification)	
L1 (Terminal ileum)	0 (0)
L2 (Colon)	3 (33.3)
L3 (Ileocolonic)	4 (44.4)
L4 (Upper GI)	3 (50)
P (perianal involvement)	1 (0.17)
E2 (left-sided UC-distal to splenic flexure	2 (22.2)
Inflammatory markers (range)	
CRP (<5 mg/L)	<3 (1-5)
Faecal calprotectin (0-50 ug/g)	27.67 (10-67)
Years since diagnoses [range, years]	3.1 (1.3-3.5)
PCDAI/PUCAI score	
<10 Remission	9 (100)
Concurrent Therapies	
Infliximab	2 (22.2)
Corticosteroids	0 (0)
Thiopurines	5 (55.6)
Partial enteral nutrition only	2 (33.3)
Prior resection	0 (0)
>1 disease flare in the last 4 years	4 (44.4)

BMI, body mass index using the WHO standard indices; PGA, physician global assessment; PCDAI/PUCAI, paediatric Crohn's disease/Ulcerative colitis activity index; SD, standard deviation; WHO, World Health Organisation.

3.2 FODMAP Restriction

Seven (77.7%) of the nine children experienced GI symptom improvement while two (22.2%) reported no change in symptoms (Table 2). No children experienced worsening of GI symptoms.

Table 2 Gastrointestinal symptom response from baseline to completion of the reintroduction phase.

	Baseline		Follow up		
Symptom	**Symptom Present n (%)‡**	**No change**	**Moderately improved**	**Substantially improved**	**Improved†**
Abdominal pain	7 (77.7)	2 (33.3)	0 (0)	5 (55.6)	5 (71.4)
Bloat/distention	7 (77.7)	0 (0)	0 (0)	7 (77.7)	7 (100)
Nausea	2 (22.2)	1 (11.1)	0 (0)	1 (11.1)	1 (50)
Diarrhea	6 (66.7)	1 (11.1)	0 (0)	5 (71.4)	5 (83.3)
Composite score§	9 (100)	4 (44.4)	0 (0)	17 (188)	**7 (77.7)**

†77.7% of children experienced some improvement of symptoms, dichotomised as improved (moderately or substantially) or not improved.

‡Total number of children experiencing symptoms in each category.

§Totalled outcome scores for symptoms experienced.

3.3 FODMAP Reintroduction

The LFD reintroduction phase was completed by the seven children who experienced symptom relief during the restriction phase. All seven children had recurrence of symptoms in response to the fructan challenge and three also experienced symptoms after ingestion of lactose. There was no reported increase in symptoms in response to fructose, galacto-oligosaccharides, mannitol or sorbitol challenges.

3.4 Anthropometry

While following the LFD restriction phase, the seven children who responded to the LFD gained a mean of 110 g, ranging from -1.1 kg to +285 g. One child who did not experience GI symptom improvement with the LFD lost 1.1 kg likely related to ongoing abdominal pain and poor appetite. The other non-responsive child experienced no change in weight.

3.5 Patient Satisfaction

Eight (88.8%) children reported the written information/resources provided were easy to understand, and six (66.7%) also found the LFD easy to follow. Three (33.3%) children including the two LFD non-responders and one LFD responder were not satisfied with their overall symptom improvement, and three were not interested in changing their diet further to improve symptoms.

4. Discussion

The LFD is commonly used in the management of adults with lower FGIS with few studies assessing its use in children with IBS [17]. A small number of studies have shown the LFD to be a useful treatment for FGIS in adults with IBD [10], but none have evaluated its use in children with IBD. This case series presents initial data on the efficacy of the LFD in reducing overlapping IBS-like

symptoms in children with quiescent IBD. Overall, the LFD was efficacious in seven of the nine children, with observed carbohydrate intolerance to fructans particularly. There was no adverse short-term impact on weight. These findings are similar to those previously reported in adults with IBD [10]. The LFD appears to be more efficacious for those with symptoms of flatulence, diarrhoea, abdominal pain and distention [10]. Moreover, the carbohydrate intolerances identified during the reintroduction process are similar for both adults and children, with fructan and lactose intolerance being the most common [18].

It is essential that dietary restrictions in children (including the LFD) are overseen by specialist dietitians to ensure nutritional adequacy. To mitigate impacts on nutritional status and gut microbiome diversity, the FODMAP restriction phase should not be prolonged and FODMAPs should be reintroduced into the diet to test for tolerance. The average weight gain experienced by children in this case report was 100 g. This may partly be explained by the observed symptom improvements and subsequently, improved appetite secondary to resolution of GI symptoms. It has previously been reported that GI symptoms may lead to altered appetite and skipping meals to manage symptoms [19]. On the other hand, one child lost weight during the restriction phase, likely a result of persistent abdominal pain (as reported by this patient). In this case, the LFD elimination phase was ceased after day 10 to prevent further weight loss.

In a randomised controlled trial study by Cox et al., [20], a 4-week LFD was demonstrated to significantly improve the gut-symptom scores of adult patients with quiescent IBD compared with the control diet group ($P = 0.007$). Furthermore, Health-related quality of life scores were also significantly improved among those who consumed the LFD ($p = 0.042$). This is worth noting given that a recent study by Brown et al. [21] showed that children with CD have significantly reduced Food-related quality of life (FR-QoL) compared with a control group of healthy children. Similarly, a recent study by Cox et al., [22] illustrated that although total FODMAP and fructan intakes were lower in adults with active IBD compared with controls, the FR-QoL was significantly lower in all IBD study groups (active disease, inactive disease with non-inflammatory gut symptoms and inactive IBD without gut symptoms (all $p = 0.001$)) irrespective of disease activity status. Consequently, it would be important to also evaluate the FR-QoL of children with quiescent IBD and non-inflammatory gut symptoms who undertake the LFD, to determine how this compares with adult IBD patients.

The limitations of this case series are its small sample size and non-randomised, observational methodology. In addition, the longer-term impact of the LFD including its effect on the microbiome and inflammatory status were not assessed. Moreover, the symptom outcomes described were reliant on patient self-report. The strengths are that this is the first real-world, clinical-setting case series to report on the efficacy of the LFD in children with IBD. Children and families were directed by a qualified and experienced paediatric dietitian who documented the impact of the LFD in standard fashion over the different dietary phases and recorded growth concurrently. Furthermore, this case series provides preliminary data that will enable an accurate assessment of effect size to ensure that future studies investigating the use of the LFD to treat paediatric functional GI symptoms are powered appropriately.

This case series demonstrates the LFD may be considered as a safe dietary option, under the direction of a suitably qualified paediatric gastroenterology dietitian, to help alleviate overlapping FGIS in children with quiescent IBD. The real-world data presented enables hypotheses to be

generated and then tested with confidence. Given these promising findings, further studies are now required to more definitively ascertain the efficacy of the LFD in children with quiescent IBD.

Acknowledgement

Written consent was obtained from the patient and/or their parent/guardian to participate in this case series.

Author Contributions

Conceptualization, methodology, investigation, and writing—original draft preparation: S.B., C.L.W., C.F., R.B.G. and A.S.D. All authors have read and agreed to the published version of the manuscript.

Funding

Professor Andrew Day is supported in his research by funding from Cure Kids New Zealand.

Competing Interests

The authors have declared that no competing interests exist.

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