

Clinical Case Report

Ulcerative colitis in the postpartum period

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ABSTRACT

We describe a scarcely reported case in which ulcerative colitis (UC) occurred in the postpartum period. The aims of this case report are to reinforce the recent assertion that a diet is a ubiquitous environmental factor in inflammatory bowel disease (IBD) and that a plant-based diet (PBD) is recommended for IBD. A 29-year-old woman normally delivered her first child. She first noticed bloody diarrhea 4.5 months after delivery. She was diagnosed with UC (left-sided colitis, moderate severity). Sulfasalazine induced remission. She then experienced and learned about PBD during an educational hospitalization. She resumed breast-feeding and stopped medication. An interview and questionnaire revealed a change in her diet 3 months after delivery, from a sound diet (plant-based diet score: 25) to an unhealthy diet (score: 9). It happened along with a change in residence, from her parent's home where her mother prepared traditional Japanese meals to her home where she prepared meals by herself. A feeling of release from childbirth prompted her to eat sweets and cheese despite being aware that the quality of the meals deteriorated. We described a scarcely reported case in which UC occurred in the postpartum period. It happened along with a change in her diet, from a sound diet to an unhealthy diet due to a feeling of release from childbirth. She replaced an omnivorous diet by PBD and stopped medication. The critical role of diet is largely ignored by healthcare professionals. We believe that greater appreciation of diet will change and improve management of IBD.

Keywords

Colitis, Ulcerative; Postpartum Period; Diet Therapy; Labor Onset; Environment.

INTRODUCTION

The incidence of inflammatory bowel disease (IBD), a collective term for ulcerative colitis (UC) and Crohn's disease (CD), has been increasing over time and expanding to different regions around the world, indicating that IBD is a global disease.¹ Rapid increase in IBD has been observed in Japan. The incidence and prevalence of UC in 1991 were 1.95/100,000 population and 18.12/100,000 population, respectively.² The prevalence increased to 63.6/100,000 in 2005.³

The incidence and prevalence of CD in 1991 were 0.51 and 5.85/100,000 population, respectively.² The prevalence increased to 21.2/100,000 in 2005.³ The number of patients receiving public medical aid for UC and CD in Japan were 2,546 and 258, respectively in 1977. These are 167,872 and 42,789, respectively in 2016.⁴ These are 66-fold and 166-fold increases in UC and CD, respectively, during the past 40 years.

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Like many other diseases, IBD is a polygenic disease thought to be triggered by environmental factors.¹ Among various environmental factors underlying IBD, a westernized diet in an affluent society can be identified as a ubiquitous environmental factor.⁵ We have provided plant-based diet (PBD) for IBD inpatients instead of a low residue diet or omnivorous diet since 2003.⁶ PBD incorporates many plant foods such as vegetables, fruits, beans, seeds, and nuts while minimizing animal foods (meat, fish), processed foods, and oils.⁷ There are various types of PBD depending on the degree of animal foods exclusion: vegan, lacto-ovo-vegetarian, semi-vegetarian, and pesco-vegetarian.⁷ Based on our far better outcomes by incorporating PBD for IBD than those in current practices, we for the first time recommended PBD for IBD.⁶ PBDs are listed as variations of USDA healthy eating patterns and are recommended to the public to prevent common chronic diseases.^{7,8}

Although increased relapse in UC during the postpartum period (6 months) has been reported,⁹ there is scarce knowledge on the onset of UC during the postpartum period. Here we report a case in which UC developed after dietary change from a prudent to an unhealthy diet during the postpartum period. The aims of this case report are to reinforce the recent assertion that a diet is a ubiquitous environmental factor in IBD and that PBD is recommended for IBD.

CASE REPORT

A 29-year-old homemaker spontaneously delivered her first child at full term in the end of September 2018 (Figure 1).

She breast-fed the baby. Three months later, she noticed a tendency toward constipation and distress in the epigastrium. She first noticed diarrhea mixed with blood at 20 weeks postpartum (Figure 1). The number of diarrhea episodes increased to 20 times/day in a few days and abdominal pain began to appear. A local doctor referred her to a tertiary care hospital, where she was hospitalized at 21 weeks postpartum (Figure 1).

Her past history was noncontributory except for palm exanthema at the age of 26. There was no family history of IBD. She reported not eating any foods that would likely cause diarrhea. Her family (husband and baby) did not have diarrhea. She did not take any supplements or medications.

Her height was 165 cm and body weight was 60 kg. She was afebrile. Physical examination was noncontributory except for mild tenderness in the left lower abdomen. Neither anal fistula nor anal skin tag was observed. Blood test disclosed a mild elevation of C-reactive protein (Table 1). Colonoscopy revealed diffuse inflammation from the rectum to the proximal descending colon with multiple yellow-white color spots¹⁰ (Figure 2). Ulcer was not observed. Biopsy specimens revealed crypt abscesses, goblet cell depletion, and mononuclear cell infiltration (Figure 3), which were consistent with findings of UC. Stool culture for pathogen, CD Chek (Techlab C. Diff Quik Chek Complete, Techlab Inc, VA, USA), and cytomegalovirus antigenemia¹¹ were negative. At this point, moderate UC (initial episode) with left-sided colitis in the extent¹² was diagnosed. Sulfasalazine 4.0 g/day was prescribed (Figure 1). Her symptoms gradually improved, then bloody stool disappeared. She was discharged after 2 weeks hospitalization. Soon after discharge,

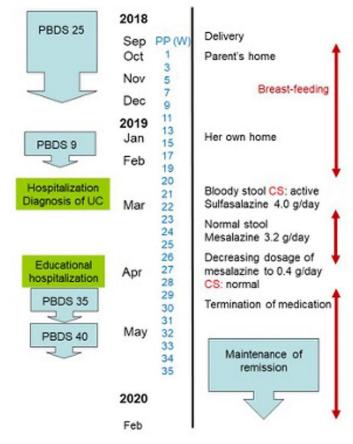


Figure 1. Timeline of case. PBDS = Plant-based diet score; UC = ulcerative colitis; PP = postpartum; W = week; CS = colonoscopy.

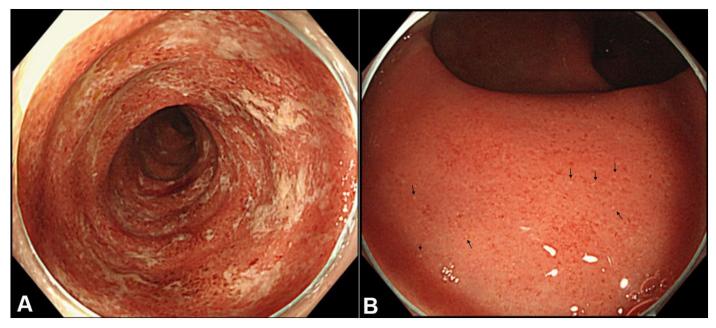


Figure 2. Photograph of colonoscopy at 21 weeks postpartum. A - Diffuse erythematous mucosa with patchy mucous adherences was observed in the sigmoid colon; <math>B - Diffuse inflammation without normal vascular pattern was observed in the rectum. Multiple yellow-white color spots (erosions) (arrows) were observed.

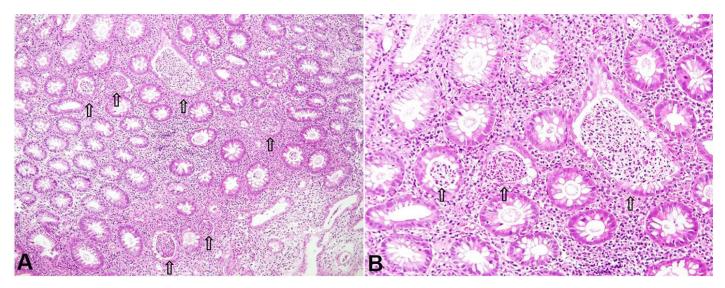


Figure 3. Photomicrographs of the biopsy specimen from the sigmoid colon. Crypt abscesses (arrows), goblet cell depletion, and mononuclear cell infiltration were observed. (**A**) (H&E, 100X); (**B**) (H&E, 200X).

sulfasalazine was replaced by mesalazine (3.2 g/day),¹³ and she resumed breast-feeding (Figure 1). Because she wanted to be free from medication, she asked her doctor to refer her to another tertiary hospital based on the information of PBD in IBD.

She underwent 12 days of educational hospitalization for UC¹⁴ at 27 weeks postpartum (Figure 1). At that time, there were no symptoms. C-reactive protein was normal and fecal occult blood was negative (Table 1). A lacto-ovo-semi-vegetarian diet (1700 kcal/day) with fish once a week and meat

every other week,¹⁵ a kind of PBD, was provided. She experienced PBD and had a dietary guidance of PBD. Colonoscopy at the end of hospitalization revealed restoration of vascular patterns which confirmed endoscopic remission of UC (Figure 1). She requested a lower dose of medication. Considering her excellent condition and our assertion that diet is generally more important than medication in the quiescent stage in IBD,¹⁵ the mesalazine dose was gradually decreased to 0.4 g/day on discharge (Figure 1). She was advised to continue PBD after discharge.¹⁴

Table 1. Laboratory data

ltem	(Normal range)	Unit	21 wks PPª	22 wks PP	23 wks PP	27 wks PP ^b	35 wks PP
Hemoglobin	11.6-14.8	g/dl	11.7	10.6	10.9	11.5	12.5
White blood cell	3300-8600	/mm³	7800	6300	5500	4500	4250
Neutrophil	38-72	%	61.3				
Lymphocyte	22-54	%	19.7				
Monocyte	2-7	%	11				
Eosinophil	0-5	%	7.4				
Basophil	0-2	%	0.6				
Platelet	15.8-34.8	x 10 ⁴ /mm ³	25.7	29.5	30.2	18.1	21.5
Total protein	6.6-8.1	g/dl	7.5			7.8	7.6
Albumin	4.1-5.1	g/dl				4.7	4.5
Sodium	138-145	mEq/l	141				
Potassium	3.6-4.8	mEq/l	3.9				
Chloride	101-108	mEq/l	103				
Choline esterase	201-421	IU/mL		192	244	309	319
C-reactive protein	0-0.14	mg/dl	0.44	0.23	0.02	<0.02	<0.02
Erythrocyte sedimentation rate	≤15	mm/hr			22	18	11
Fecal occult blood	<100	ng/mL				<50	

^aAdmission for active ulcerative colitis; ^bEducational hospitalization for ulcerative colitis. Blanks mean that the item was not tested. PP = postpartum.

An interview and questionnaire on lifestyle and dietary habits¹⁶ revealed a dietary change during the postpartum period. Midwives recommended Washoku¹⁷ for pregnancy and for lactation. Washoku is a traditional Japanese diet prevalent before dietary westernization and is similar to a presco-vegetarian diet. After delivery, she stayed at her parent's home for about 3 months. Her mother prepared Washoku at the midwife's recommendation. She subsequently returned to her own home at 14 weeks postpartum (Figure 1). She had a feeling of being released from childbirth. She prepared meals by herself. Although she was aware that the quality of the meals deteriorated, she had urges to eat sweets and cheese. She felt a tendency of constipation and distress in the epigastrium. This dietary change was assessed by plant-based diet score (PBDS).¹⁶ The score dropped from 25 to 9 (Table 2, Figure 1).

After the educational hospitalization, she was glad to be able to resume breast-feeding. She was well, then she completely stopped taking her medication 3 weeks after discharge. Her PBDS one month after the discharge (35 weeks postpartum) was 40, the highest score (Table 2). The values of hemoglobin and erythrocyte sedimentation rate became normal (Table 1). She has remained in remission without medication to the present (February 2020).

The patient has provided informed consent for publication of the case.

DISCUSSION

Although the etiology of IBD is generally stated as unknown, we first regarded IBD as lifestyle disease mediated mainly by westernized diet.¹⁵ PBD was designed to combat dietary westernization.^{6,15} It is apparent now that diets shape gut microbiota.¹⁸ Recently, basic research on the gut microbiome has provided a rationale for how PBD is superior to westernized diets.¹⁹⁻²¹ Westernized diet (high in fat, animal protein and sugar, low in dietary fiber) tend to decrease microbial diversity (dysbiosis), while PBD (low in fat, animal protein and sugar, high in dietary fiber) tend to increase microbial diversity.¹⁹⁻²¹ This difference in microbiota results in differences in microbial metabolites. Westernized diets result in

	Scoring by frequency of			-	Present case			
Food group	ser Daily	ving c 3-5		veek Rarely	Baseline (until 3 months postpartum)	After 3 months postpartum	Educational hospitaliza tion (12 days)	One month after educational hospitaliza tion
Positive score								
Vegetables	5	3	1	0	5	3	5	5
Fruits	5	3	1	0	5	3	5	5
Pulses	5	3	1	0	5	5	5	5
Potatoes/starches	5	3	1	0	1	1	5	5
Rice	5	3	1	0	5	5	5	5
Miso soup	5	3	1	0	5	1	5	5
Green tea	5	3	1	0	0	0	Oa	5
Yoghurt (plain)	5	3	1	0	5	5	5	5
Negative score								
Meat	-5	-3	-1	0	-1	-1	0	0
Minced or processed meat	-5	-3	-1	0	-1	-1	0	0
Cheese/butter/margarine	-5	-3	-1	0	-1	-5	0	0
Sweets/ice cream/milk shake	-5	-3	-1	0	-1	-5	0	0
Soft drinks (cola/carbonated beverages/juice)	-5	-3	-1	0	0	-1	0	0
Alcohol	-5	-3	-1	0	0	0	0	0
Bread	-5	-3	-1	0	0	-1	0	0
Fish	-2	-1	0	0	-2	0	0	0
Plant-based diet score (PBDS)					25	9	35	40

Table 2. Plant-based diet Score (PBDS) for Japanese patient with inflammatory bowel disease

^aGreen tea is recommended to drink at home but is not provided at a hospital.

increased production of ammonia, indols, phenols, and sulphide that may be detrimental to our health. In addition, they result in decreased production of short-chain fatty acids like butyrate. Short-chain fatty acids, particularly butyrate, have diverse beneficial effects in nutrition, immunity, and epithelial barrier function (enhanced mucous secretion and increased antimicrobial peptide). PBD result in increased production of short-chain fatty acids. Altogether, westernized diet are pro-inflammatory while PBD is anti-inflammatory.¹⁹⁻²¹ These observations indicate that westernized diets are susceptibility to not only IBD but also other chronic diseases.

Focusing on diets, we reported cases with new onset of IBD during a change in dietary habits: UC during a change of lifestyle,²² during low-carbohydrate weight-loss diet,²³ in the second trimester after

emesis gravidarum,²⁴ and CD after moving to Tokyo.²⁵ The present case of UC in the postpartum period is another example demonstrating that diet is a critical environmental factor in IBD.⁵ PBDS evaluate adherence to PBD: a higher PBDS indicates greater adherence to PBD.¹⁶ The present case's PBDS after 3 months postpartum was 9 (Table 2, Figure 1), which was comparable to the mean baseline PBDS (10.9) in 158 UC patients.¹⁶

When PBD was first provided to IBD patients, we assumed that diet was generally more important than medication to prevent relapse in the quiescent phase,¹⁵ and our findings have shown that to be true. Relapse rates in both UC and CD were far lower than those with current medication.^{6,14,15,26} PBDS in these patients were significantly higher than baseline PBDS not only in the short-terms of less than 2 years after discharge

but also even in the long-term: median 3.9 years¹⁴ and 6.4 years²⁶. This certifies that they consumed more of the recommended food and consumed less of the food that was discouraged compared with baseline. Although current practice recommends adherence to medication,²⁷ we emphasize that diet is more important than medication in the quiescent phase of IBD. Therefore, medication is withdrawn whenever patients have confidence in relapse prevention. The maximum PBDS score, 40 in the present case one month after educational hospitalization (Table 2, Figure 1), seems to be a reflection of her strong conviction that the disease is suppressed by sound lifestyle.

Unfortunately, lifestyle medicine including dietary habits is not fully appreciated in current medicine.^{28,29} Appreciation of lifestyle medicine is needed to prevent and treat current common chronic diseases in affluent society.

In conclusion, we described a scarcely reported case in which UC occurred in the postpartum period. It happened along with a change in her diet, from a sound diet to an unhealthy diet due to a feeling of release from childbirth. This case supports the recent assertion that a diet is a ubiquitous environmental factor in IBD⁵ and that PBD is recommended for IBD⁶. This observation should encourage researchers to explore the PBD properties in controlled studies.

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This study was carried out at Akita City Hospital, Akita City, Japan.

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