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Food for Thought: Remission of Perianal Pediatric Crohn's Disease on Specific Carbohydrate Diet Monotherapy

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Abstract: There is growing interest among patients about the specific carbohydrate diet (SCD) as a treatment for Crohn's disease. In the meantime, deep remission in patients using SCD as their sole treatment has not been documented. We report a case with perianal and ileocolonic Crohn's disease in whom SCD monotherapy successfully induced and maintained not only clinical, but also endoscopic, radiographic and histologic (ie, deep mucosal remission) remission as well.

Key Words: inflammatory bowel disease, Crohn's disease, specific carbohydrate diet, dietary therapy, nutrition

INTRODUCTION

Diet is well established to play a role in both the pathogenesis and treatment of inflammatory bowel diseases (IBD) (1). As such, dietary therapies (eg, specific carbohydrate diet [SCD]) are of high interest to many patients and families battling the disease. Herein, we describe a unique case of sustained clinical, biochemical, radiographic, endoscopic, and histological remission in a pediatric patient with new onset penetrating Crohn's disease (CD) on exclusive SCD therapy. This observation lends further impetus towards developing optimized clinical trials to better understand the efficacy and mechanism of action of SCD in the treatment of IBD and stresses the importance of a shared decision-making approach when dealing with an uncurable, chronic illness in a pediatric patient.

CASE REPORT

An 8-year-old, previously well, female presented to the emergency room for evaluation and management of a large, left labial abscess. She ultimately underwent incision and drainage (I & D) of the abscess with placement of a vessel loop drain (removed at time of diagnostic endoscopy). Her perineal examination under anesthesia, performed at the time of her I & D, revealed an anterior anal skin tag and evidence of a small, healed, external fistulous opening in the right lateral perianal position. Our patient was discharged from the hospital on post-operative day 1. She was to complete a 14-day course of amoxicillin-clavulanate and follow-up with gastroenterology for evaluation of underlying inflammatory bowel disease.

At the time of gastroenterology follow-up, our patient had completed 2 weeks of antibiotics. She endorsed occasional abdominal pain but was otherwise asymptomatic. She denied pain or drainage at the site of her, now drained, labial abscess. Growth and weight gain were noted to be normal for age. Laboratory evaluation at the time of the visit (see Table 1) was pertinent for elevated fecal calprotectin, elevated C-reactive protein, normocytic anemia, and hypoalbuminemia. Subsequent endoscopic and histologic evaluation supported the diagnosis of gastric and ileocolonic CD (diagnostic histology and endoscopic photographs shown in Fig. 1Aa, Ab, Ca, Cb, Ea, Eb; Simple Endoscopic Score for CD [SES-CD] > 15). Further diagnostic imaging (eg, MRI pelvis and MRE) showed a simple, right-sided, low-lying, dry perianal fistula (2.1 × 0.3 cm) without abscess formation (see Fig. 2A) and no radiographic evidence of small bowel inflammation outside the terminal ileum. Her Pediatric CD Activity Index (PCDAI) score at time of diagnosis was 15.

Following diagnosis, our patient and family were extensively counseled regarding potential treatment options, and it was recommended for her to begin infliximab induction and maintenance therapy given the penetrating phenotype of her CD. However, given a desire to avoid potential side effects associated with life-long anti-TNF therapy, our patient's family decided to seek a second opinion at our facility.

After a thorough discussion with our patient's family regarding treatment options and long-term care goals, we mutually agreed to begin SCD monotherapy. Notably, our patient never received any additional IBD specific therapies, including steroids, following her diagnosis and prior to starting her SCD. Following initiation of the SCD diet, our patient demonstrated steady clinical and biochemical improvement (see Table 1). Approximately 1 year into her diagnosis and with reported rigorous and diligent adherence to the SCD, our patient showed good weight gain for age (~2 kg increase since diagnosis), clinical and radiographic resolution of her perianal disease (Fig.2B), and normalization of her laboratories (Table 1). Follow-up

TABLE 1. Pre- and post-SCD treatment laboratories

	At diagnosis/SCD initiation	2 mo on SCD	3 mo on SCD	6 mo on SCD	1 y on SCD
Calprotectin (<50 µg/g)	948	380	31	21	7
Hemoglobin (11.5–15.5 g/dL)	10.8	11.8	12.0	12.4	12.2
Hematocrit (35%–45%)	34.2	36.7	37.1	36.5	35.6
Albumin (3.6–5.1 g/dL)	3.4	4.2	4.0	4.4	4.1
C-reactive protein (<0.8 mg/dL)	10.7	0.5	0.5	0.5	0.7
Erythrocyte sedimentation rate (<20 mm/h)	9	7	8	11	7

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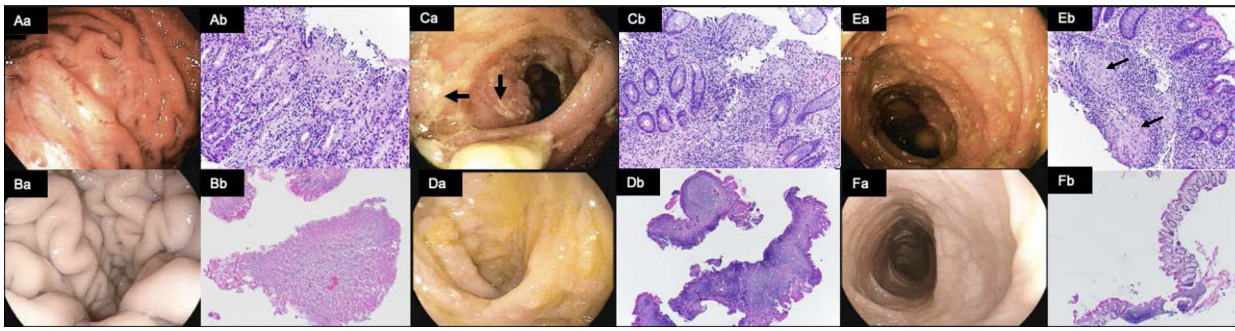

FIGURE 1. Aa) Endoscopic image of the stomach before SCD; (Ab) corresponding gastric histology prior to SCD showing significant chronic, active inflammation (H&E 200×). Ba) Endoscopic image of the stomach after treatment with SCD; (Bb) corresponding gastric histology after treatment with SCD showing complete remission (H&E 40×). Ca) Endoscopic image of the terminal ileum before SCD (black arrows depicting example areas of ulceration). Cb) Corresponding histology of the terminal ileum prior to SCD showing significant chronic, active inflammation (H&E 200×). Da) Endoscopic image of the terminal ileum after treatment with SCD. Db) Corresponding histology of the terminal ileum after treatment with SCD showing complete remission (H&E 40×). Ea) Endoscopic image of the descending colon before SCD. Eb) Corresponding histology of the descending colon prior to SCD showing chronic, active colitis with non-necrotizing granulomas (arrows) (H&E 200×). Fa) Endoscopic image of the descending colon after treatment with SCD. Fb) Corresponding histology of the descending colon after treatment with SCD with complete remission (H&E 40×).

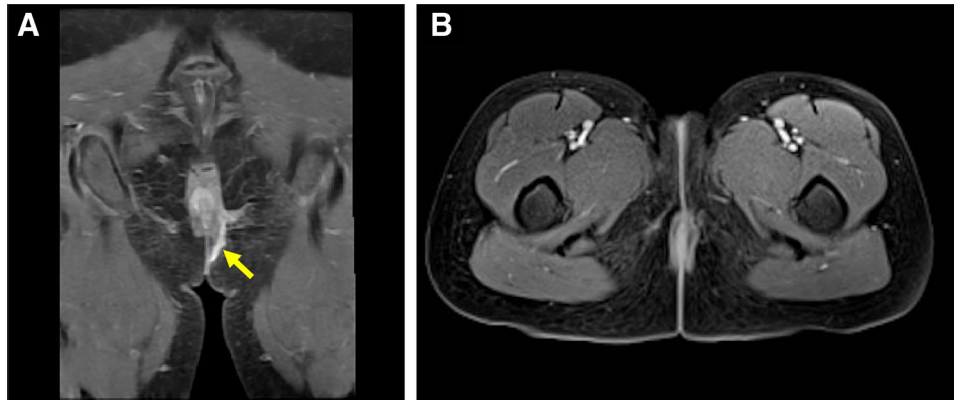

FIGURE 2. A) Pelvic magnetic resonance imaging. Coronal post contrast T1 weighted sequence showing a perianal fistulous tract in the left lower quadrant (yellow arrow). B) Magnetic resonance enterography. Axial post contrast T1 weighted sequence showing resolution of the perianal fistulous tract ~1 year on SCD monotherapy.

surveillance EGD and ileocolonoscopy demonstrated endoscopic and histological remission (follow-up endoscopic and histopathological photographs shown in Figure 1Ba, Bb, Da, Db, Fa, Fb). To date, our patient remains in clinical remission of her CD on strict SCD monotherapy. She continues to follow in our clinic on a biannual basis for clinical evaluation and biochemical monitoring.

DISCUSSION

The role of diet in the pathogenesis of IBD is strongly implicated (1,2). Diet is a key determinant of the gut microbiome and dietary changes are known to result in altered microbial composition (3). An improper imbalance (eg, dysbiosis) characterized by decreased microbial diversity is a hallmark of IBD (3). When combined with impaired mucosal barrier function, dysbiosis can become a potent driver of mucosal inflammation (3). Diet has also been shown to alter immune function through interactions with gut mucosal defenses and inflammatory cells (3). It follows that diet is not only immensely consequential in the development and propagation of IBD but potentially therapeutic as well (2).

The SCD is based on an exclusion of complex carbohydrates, sugar, and most dairy and processed foods that are believed to be poorly absorbed and proinflammatory (4). The theory assumes that carbohydrates have an outsized influence on the gastrointestinal microbiome balance (4). Studies examining the SCD in pediatric patients with inflammatory bowel disease have been promising (2,4–8). Clinical, biochemical, and mucosal improvements have been shown in small pediatric cohorts with both nonpenetrating CD and ulcerative colitis (5–7). There are inherent difficulties, however, for objectively studying dietary treatments (8) and there has yet to be conclusive evidence that SCD monotherapy is capable of inducing mucosal healing in patients with CD (9).

Despite the potential benefits of the SCD and favorable risk benefit profile when compared to standard immunosuppressive IBD therapies, the SCD remains difficult to maintain for many patients (6). Any attempt at liberalizing the diet poses the potential risk of a disease flare. In addition, the intensive and restrictive nature of the diet can prove isolating for many children and lead to a decreased quality of life. Nonetheless, SCD therapy remains of high interest to many patients and families (see https://specificcarbohydratedietassociation.org/)

seeking to avoid the risks associated with life-long immunosuppressive medications.

To our knowledge, this is the first documented case of sustained clinical, biochemical, radiographic, endoscopic, and histologic remission in a pediatric patient with penetrating CD on SCD monotherapy. Our case emphasizes the importance of shared decision-making between physician, child, and family when dealing with chronic, uncurable conditions (see https://www.aap.org/en/practice-management/providing-patient--and-family-centered-care/shared-decision-making/). Through the information exchange of treatment evidence, options, benefits, and risks in combination with the family's values and preferences, the physician, parent, and patient can better deliberate to determine the best short- and long-term treatment plans. As seen in our case, such an approach serves to improve knowledge and lessen decisional conflict whilst also leading to a successful patient outcome.

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