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Case Report

Colorectal Carcinoma at Diagnosis of Ulcerative Colitis in a 17 Years Old Female

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Abstract

Inflammatory bowel disease (IBD) is a known risk factor for colorectal cancer (CRC). The degree of risk is related to the duration and extent of colitis, as well as the severity of inflammation over time. We present a 17 years old female diagnosed with carcinoma in situ associated with ulcerative colitis (UC) at first colonoscopy for suspected IBD. She presented with non-bloody diarrhea of 3 years' duration. A clinical diagnosis of irritable bowel syndrome (IBS) was suggested elsewhere and her symptoms improved with loperamide. Hematochezia triggered an investigation for IBD. The colonoscopy revealed pancolitis and no tumor was seen. Histology confirmed characteristic signs of chronic colitis with focal ulcerations, crypt branching and abscesses and dense acute and chronic inflammatory infiltrates. Multiple biopsies showed signs of definite dysplasia, as well as the presence of P53 and k167 expression. Two independent pathologists confirmed high-grade dysplasia with carcinoma in situ in the descending colon. A proctocolectomy with ileo-anal J pouch anastomosis was performed. The pathology specimen confirmed a diagnosis of UC without skip lesions and a carcinoma in situ in the descending colon.

Rare cases of CRC at or shortly after diagnosis of IBD exist. Our case is exceptional in that CRC was found at the time of initial colonoscopy for suspected IBD. This emphasizes the fact that CRC can be present early after the onset of symptoms and may be delayed or missed when conducting surveillance strictly according to formal guidelines.

ABBREVIATIONS

IBD: Inflammatory Bowel Disease; CRC: Colorectal Cancer; UC: Ulcerative Colitis

INTRODUCTION

Inflammatory bowel disease (IBD) is a known risk factor for colorectal cancer (CRC). The degree of risk is related to the duration and extent of colitis, as well as the severity of inflammation over time. Other risk factors include primary sclerosing cholangitis and a family history of sporadic CRC. The American College of Gastroenterology (ACG) guidelines [1] and the American Gastroenterological (AGA) guidelines2 suggest surveillance based on duration of disease, not chronological age. Despite the lack of randomized controlled trials, surveillance

colonoscopy is recommended for patients with IBD at increased risk for developing CRC. Patients with extensive UC or CD of the colon are most likely to benefit from surveillance. Others known risk factor of CRC is colonic strictures in patients with UC, a shortened colon and multiple post inflammatory pseudopolyps [2]. Persistent inflammation is a risk factor for progression to colorectal neoplasia2. Surveillance colonoscopies are recommended starting 8 years after onset of symptoms with multiple biopsy specimens obtained throughout the entire colon [2]. Although there are inadequate data available to recommend optimal surveillance intervals, intervals of 1 to 3 years are suggested. The surveillance should be conducted as frequently in children as in adults as children diagnosed with IBD have at least an equivalent rate of CRC development compared to patients diagnosed at an older age. The AGA guideline evoked the controversy concerning the early age at onset of IBD as an independent risk factor of CRC as some reference report a highest cumulative risk of CRC if the disease began before 15 years of age while other data suggest an increased risk of CRC in patients with IBD diagnosed after the age of 40 years [2].

CASE PRESENTATION

We present the case of a 17 years old female diagnosed with carcinoma in situ associated with UC at the initial colonoscopy for suspected IBD. She presented with non-bloody diarrhea that lasted for three years. Physical exam and routine laboratory tests were unremarkable. A clinical diagnosis of IBS was suggested elsewhere and her symptoms improved with loperamide. Hematochezia triggered an investigation for IBD. The colonoscopy revealed pancolitis, with edematous and hyperemic granular mucosa, mucopurulent exudates and friability. No tumor was seen. Histology of colonic biopsies confirmed characteristic signs of chronic colitis throughout the colon with focal ulcerations, crypt branching and abscesses and dense acute and chronic inflammatory infiltrates (Figure 1). Biopsies from the ascending colon, transverse colon, descending colon and rectum showed signs of definite dysplasia, as well as the presence of P53 and k167 expression (Figures 2 and 3). Two independent pathologists confirmed high-grade dysplasia with carcinoma in situ in the descending colon. A proctocolectomy with ileo-anal

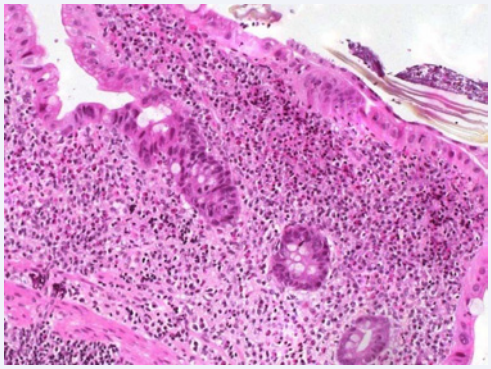

Figure 1 Transverse colon biopsy (200X) showing architectural distortion and a dense lymphoplasmocytotic infiltrate.

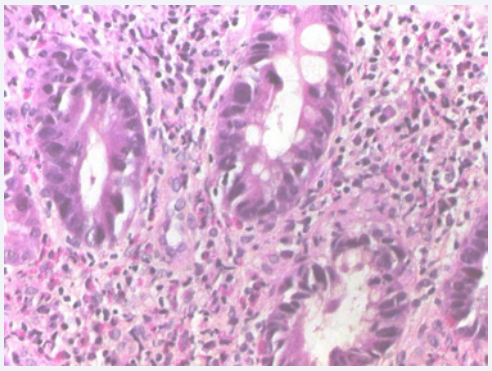

Figure 2 Descending colon biopsy (400X) showing basophilic atypical cells with mitosis and signs of stratification.

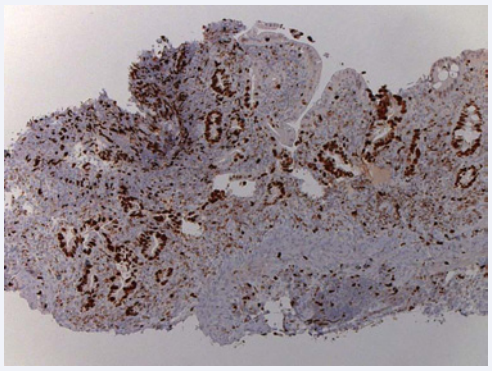

Figure 3 Staining for ki67 expression in the colonic mucosa.

J pouch anastomosis was performed. The pathology specimen of the resected colon confirmed a diagnosis of UC, without skip lesions. Histological assessment of the resected colon confirmed carcinoma in situ in the descending colon with areas of low-grade dysplasia and multifocal high-grade dysplasia with flat mucosa were identified in the cecum, ascending colon, transverse colon and rectum. In the three years following her ileo-anal J pouch procedure, there has been no recurrence of dysplasia in the rectal cuff. Recurrent episodes of pouchitis have occurred despite treatment with probiotics, and were successfully managed with antibiotics.

DISCUSSION

Although typically encountered in adults with longstanding UC, Lagercrantz first reported CRC in pediatric UC in 1949 [3]. Other cases were published over the ensuing 2 decades [4-7]. In 1961 the Mayo Clinic reported that 10% of UC patients below 21 years of age died of CRC [8]. Over the last 40 years improvements in the treatment and increased awareness of risk of CRC in IBD has impacted favorably on the incidence of CRC. Nevertheless, rare cases of CRC at or shortly after diagnosis of IBD exist. The first CRC associated with Crohn's disease in childhood was reported in 2001 in an adolescent with undiagnosed chronic abdominal pain for 5 years [9]. Recently, an adolescent male with CRC after only 3 years of UC was reported in a Danish population-

based study. No signs of dysplasia were found on initial biopsies [10]. The case presented here is exceptional in that CRC was found at the time of initial colonoscopy for suspected IBD. This emphasizes the fact that CRC can be present early after the onset of symptoms and may be missed, depending on the number of biopsy specimens taken during the colonoscopy. One recent adult study showed that the diagnosis of colorectal cancer is delayed or missed in a substantial number of patients (17–28%) when conducting surveillance, according to formal AGA and ACG guidelines [11]. It was recently suggested that surveillance with mucosal biopsies for dysplasia alone may be inadequate, as aneuploidy was found without dysplasia in patients as young as 16 years [12]. Molecular markers should not be applied to help stratify patients into low-risk and high-risk groups at this time. Further research to optimize detection of pre-cancerous changes in high-risk IBD patients is needed.

REFERENCES

1. Asher Kornbluth , David B Sachar. The Practice Parameters Committee of the American College of Gastroenterology Ulcerative Colitis Practice Guidelines in Adults: American College of Gastroenterology, Practice Parameters Committee. Am J Gastroenterol. 2010;105:500
2. Farraye FA, Odze RD, Eaden J, Itzkowitz SH, McCabe RP, Dassopoulos T. AGA medical position statement on the diagnosis and management of colorectal neoplasia in inflammatory bowel disease. Gastroenterology. 2010; 138: 738-745.
3. Lagercrantz R. Ulcertaive colitis in children. Acta paediatrica. 1949; 75: 89-151.
4. Mallory TB. Case records of the Massachusetts general hospital; case #36501. N Eng J Med 1950; 243; 967-970
5. Wheelock FC Jr, Warren R. Ulcerative colitis; follow-up studies. N Engl J Med. 1955; 252: 421-425.
6. Wilcox HR Jr, Beattie JL. Carcinoma complicating ulcerative colitis during childhood. Am J Clin Pathol. 1956; 26: 778-786.
7. Brown CH, Diaz RJ, Michener WM. Carcinoma of the colon and ileum in chronic ulcerative colitis with reflux ileitis. Report of a case of a sixteen-year-old boy. Gastroenterology. 1964; 47: 306-312.
8. Michener WM, Gage RP, Sauer WG, Stickeler GB. The prognosis of chronic ulcerative colitis in children. N Engl J Med. 1961; 265: 1075-1079.
9. Tiszlavicz L, Kapin M, Várkonyi A, Lõrincz A, Bartyik K, Várkonyi T. Adenocarcinoma of the colon developing on the basis of Crohn's disease in childhood. Eur J Pediatr. 2001; 160: 168-172.
10. Jakobsen C, Paerregaard A, Munkholm P, Wewer V. Paediatric inflammatory bowel disease during a 44-year period in Copenhagen Country: Occurrence, course and prognosis-a population-based study from the Danish Crohn Colitis Database, Euro J Gastroenterol Hepatol. 2009, 21: 1291-1301.
11. Lutgens MW, Vleggaar FP, Schipper ME, Stokkers PC, van der Woude CJ, Hommes DW. High frequency of early colorectal cancer in inflammatory bowel disease. Gut. 2008; 57: 1246-1251.
12. Markowitz J, McKinley M, Kahn E, Stiel L, Rosa J, Grancher K. Endoscopic screening for dysplasia and mucosal aneuploidy in adolescents and young adults with childhood onset colitis. Am J Gastroenterol. 1997; 92: 2001-2006.

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