World Journal of Gastroenterology

Online Submissions: http://www.wjgnet.com/1007-9327office
wjg@wjgnet.com
doi:10.3748/wjg.v17.i1.135

World J Gastroenterol 2011 January 7; 17(1): 135-136
ISSN 1007-9327 (print) ISSN 2219-2840 (online)

Duplicated appendix complicated by appendiceal cancer

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Received: October 15, 2010 Revised: December 1, 2010
Accepted: December 8, 2010
Published online: January 7, 2011

Abstract

A 37-year old male presented with an acute abdomen suggestive of an appendiceal perforation. Urgent laparotomy showed a duplicated appendix with one of the lumens involved with appendicitis and a focal periappendicular abscess while the other lumen had a localized appendiceal cancer. Recognition of congenital intestinal duplications in adults is important to avoid serious clinical consequences.

Key words: Duplicated appendix; Bifid appendix; Appendiceal cancer; Congenital duplication.

Peer reviewer: Dr. Benjamin Perakath, Professor, Department of Surgery Unit 5, Christian Medical College, Vellore 632004, Tamil Nadu, India

Freeman HJ. Duplicated appendix complicated by appendiceal cancer. *World J Gastroenterol* 2011; 17(1): 135-136 Available from: URL: http://www.wjgnet.com/1007-9327/full/v17/i1/135.htm DOI: http://dx.doi.org/10.3748/wjg.v17.i1.135

TO THE EDITOR

An article recently published in the *World Journal of Gastroenterology* concerned a duplicated vermiform appendix in an adult[1]. The authors emphasized the need for surgical intervention to avoid any future complication, in particular, the disconcerting possibility of malignant degeneration within the duplication[2,3]. Duplication of the appendix, fortunately, is distinctly rare, reportedly occurring in only 1/25 000 patients (0.04%) operated on for acute appendicitis[4]. Clinical features may depend on the type of appendiceal duplication as a number of variants have been described, specifically, type A with incomplete duplication and a common base (so-called bifid appendix), and types B or C, both forms having complete duplication with independent bases[5,6].

We had a similar, but unusual experience in a 37-year old adult male in August 1996. He presented with an acute abdomen thought to be a perforated appendix. An urgent laparotomy showed a type A duplicated or bifid appendix with an acute appendicitis involving only one lumen associated with a focal periappendiceal abscess and localized peritonitis. The other lumen showed a well-differentiated carcinoma extending into, but not through the muscularis propria. No vascular or lymphatic invasion was identified. Imaging studies and carcinoembryonic antigen testing were negative. Two weeks later, a further ileocecal resection was done. The resected intestine and 17 mesenteric lymph nodes showed no malignancy. Family history revealed that his father had synchronous mucinous colon cancers at the age of 45 years and his brother was diagnosed with colon cancer at the age of 26 years. A paternal uncle died of colon cancer at the age of 35 years. No other congenital abnormalities were recognized and there was no known contact with a toxic or noxious agent *in utero*. Colonoscopy screening at the ages of 20, 24 and 30 years was completed in other centers because of his family history of colon cancer and no abnormalities were detected. A colonoscopy in November 1996 in our hospital revealed no other abnormalities, similar to colonoscopies in 2000, 2005 and 2010. Because of his family history, genetic evaluation to exclude a HNPCC was negative including gene testing for MLH1 and MSH2.

Carcinoma in the intestinal tract developing with coexistent duplication has been rarely reported and appears

limited to only a small number of cases[3,7]. Appendiceal cancer has been associated with a duplication thought to represent a Meckel's diverticulum[8] as well as in a single prior report of a duplicated appendix[9]. While this presentation may also mimic colon cancer[10], missing a second duplicated appendix in this setting could lead to serious clinical consequences. In the present case, appendiceal duplication was fortunately recognized and an early stage carcinoma was successfully resected.

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S- Editor Tian L **L- Editor** Wang XL **E- Editor** Ma WH