Case Report

Adenocarcinoma in an Ileal Duplication

M Suresh Babu*, Mohammed Raza**

Abstract

Alimentary tract duplications are a rare congenital anomalies that usually present in childhood and occasionally in adults. To our knowledge, since 1884, there have been only 14 reported cases of ileal duplications in adults of which 3 patients had developed malignancy in the duplicated ileum. We report here such an extremely rare case of isolated ileal duplication in an elderly male in whom malignancy occurred and perforated.

INTRODUCTION

Alimentary tract duplications are rare congenital anomalies that usually present in childhood and rarely in adults. Clinical presentation of ileal duplication is most commonly related to complications such as intestinal obstruction, perforation or hemorrhage. Since 1884, there have been only 14 reported cases of ileal duplications in adults1 of which three had developed malignancy in the duplicated segment. We report here such an extremely rare case of isolated ileal duplication in which malignancy had developed and perforated.

CASE REPORT

A 59 years male patient who was not a known smoker or alcoholic presented with symptoms of fever with abdominal pain of one day duration. Abdominal pain was diffuse in distribution and continuous. Patient had history of vague abdominal discomfort from past three to four months with decreased appetite. Patient was a known diabetic on treatment for past 10 years.

Patient was moderately built and poorly nourished. There was mild pallor. There was no icterus, clubbing, lymphadenopathy or pedal edema. Patient was febrile with tachycardia and tachypnea. Cardiovascular and respiratory system examination were within normal limits. Abdominal examination showed abdominal distension with tenderness all over the abdomen. No peristaltic movements were seen. Bowel sounds were not heard and there was no free fluid. Per rectal examination was normal. Routine hematological and biochemical parameters were within normal limits except Hb-10gm%, total leucocyte count (TLC-12,100/cc) and ESR (50 mm/hr). X-ray abdomen showed features of distal ileal obstruction. Ultrasound scanning showed few dilated bowel loops in lower abdomen. A clinical diagnosis of intestinal obstruction/peritonitis was made and laparotomy performed.

Abdomen exploration showed purulent pus in the peritoneal cavity. There was a tubular structure resembling small bowel, communicating proximally with the ileum, 25 cms from the ileocaecal junction, arising from the mesenteric border of ileum. There was perforation with purulent discharge in the distal blind end of the tubular structure at antimesenteric border. The tubular structure with a part of adjacent parent ileum was resected and intestinal continuity was re-established with a two layered end to end anastomosis. The post operative period was uneventful.

The surgical specimen consisted of partially cut opened small intestine measuring 34 cms. There was a tubular structure measuring 16 cms which was attached to the small intestine. The cut section of small intestine was unremarkable. Routine hematological and biochemical parameters were within normal limits except Hb-10gm%, total leucocyte count (TLC-12,100/cc) and ESR (50 mm/hr). X-ray abdomen showed metastatic deposits of mucin secreting adenocarcinoma.

Sections from the parent small intestine showed no significant pathology. A diagnosis of mucin secreting adenocarcinoma in an ileal duplication was made. Patient was later subjected to chemotherapy. There was improvement in the appetite and general well being of the patient at one year of follow up.
DISCUSSION

Alimentary tract duplications are rare congenital anomalies. They may occur anywhere from the pharynx to the rectum with approximately 33% arising in the foregut, 56% in the midgut and 11% in the hindgut. The ileum is the single most common site (34%). Duplications in the alimentary tract are spherical or tubular congenital anomalies and were first described by Fitz. They are attached or adherent to the mesenteric side of alimentary tract, contain a wall of smooth muscle (usually two layers) and have a mucosal lining of some part of the alimentary tract. These features help to differentiate them from true and false diverticulae, as well as mesenteric cysts.

Duplications are presumed to result from disturbances in the embryonic development of gut. Intra cellular vacuoles are formed during the solid stage of alimentary tract development in the sixth embryonic week. These vacuoles could develop into cysts or coalesce into chains forming tubular duplications. The duplication wall contains all layers except where it is fused with the bowel, where a common muscular wall is shared. No plane of cleavage can be established between the duplication and adjacent bowel and blood supply to the normal alimentary tract may be disrupted by the local excision of duplicated segment.

Clinical presentation of ileal duplication is most commonly related to complications such as intestinal obstruction, perforation or hemorrhage. Common symptoms in adults include vague abdominal pain and dyspepsia. Majority of cases with duplication present in childhood and rarely in adults. To our knowledge, only 14 patients older than 15 years with isolated ileal duplication have been described in the literature of which malignancy had developed in 3 cases. In two cases it was adenocarcinoma while in the other it was squamous cell carcinoma. No diagnosis of enteric duplication was made preoperatively in any patient. Once duplication has been diagnosed it should be removed, even if it is not causing symptoms, because of the possibility of malignant transformation. Because duplicated segment shares a portion of its wall with the adjacent intestine and a common blood supply, a complete resection of the duplication and adjacent bowel is the treatment of choice.

To conclude, isolated ileal duplication is a rare congenital anomaly which rarely presents itself in adults. Adenocarcinoma in the duplicated ileal segment is an extremely rare complication which was observed in this case.

Acknowledgement

We acknowledge the help of Prof. C.P. Madhu, Prof. of Surgery, JSS Medical College Hospital, for his help during the surgical intervention of the patient. We thank Dr. G. V. Manjunath, Prof. of Pathology, JSS Medical College Hospital for his help in the histopathological examination of the surgical specimen. We thank Dr. Soumyasundaram, P.G. in Medicine department for her help in the preparation of the Manuscript.

REFERENCES


ERRATUM

The Correspondance entitled 'Pleural Effusion of Dual Etiology by Dr. RC Sahoo and Dr. PR Acharya was published in December 2007 as well as in January 2008 issue of JAPI. This has occurred due to printing error and the published correspondence in January 2008 should be treated as withdrawn. The error is regretted.

Hon. Editor