ISSN- O: 2458 - 868X, ISSN–P: 2458 – 8687
National Library of Medicine - ID: 101731606

International Journal of Medical Science and Innovative Research (IJMSIR)
IJMSIR : A Medical Publication Hub
Available Online at: www.ijmsir.com
Volume – 7, Issue – 3, June – 2022 , Page No. : 139 - 142

Colocutaneous fistula with incidental finding of submucosal angiolipofibromatous polyps: A rare case report

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Citation this Article: Dr. Mangesh Londhe, Dr. Archana Buch, "Colocutaneous fistula with incidental finding of submucosal angiolipofibromatous polyps: A rare case report", IJMSIR- June - 2022, Vol – 7, Issue - 3, P. No. 139 – 142.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Enterocutaneous fistulas (ECF) are aberrant connections between the gastrointestinal tract and skin. They occur either spontaneously due to underlying malignancy, inflammatory bowel disease, radiation exposure, or as a complication of gastrointestinal surgery. The most common colonic polyps are epithelial in nature, arising in the mucosa followed by a small subset which are mesenchymal type arising in submucosa. The most common types of mesenchymal polyps are lipomas, leiomyomas, benign neural and vascular lesions, and gastrointestinal stromal tumors (GISTs). The vast majority of the mesenchymal polyps are composed of a single tissue type with mixed lesions being extremely rare. We herein present a case of colocutaneous fistula with incidental finding of unusual submucosal angiolipofibromatous polyps.

Keywords: Enterocutaneous Fistulas; Intestinal Polyps; Rare.

Introduction

An abnormal connection between the gastrointestinal tract and the skin is called enterocutaneous fistula (ECF). By some estimates ECF represents 88.2% of all fistulae.[1,2]. The most common cause of ECF is iatrogenic (75–85%) which includes trauma, operations for malignancy, associated with extensive adhesiolysis, or in the setting of inflammatory bowel disease (IBD). [3] Spontaneous ECF accounts for 15-25% of all ECF's which occur from IBD (most common), malignancy, appendicitis, diverticulitis, radiation, tuberculosis/actinomycosis, and ischemia. [4] Regarding intestinal polyps, the most common type are mucosal epithelial polyps. Submucosal mesenchymal polyps are rare. Among the mesenchymal polyps, the mixed mesenchymal polyps are even rarer which includes angiolipofibromatous poylps.[5] However simultaneous occurrence of ECF and angiolipofibromatous polyps has not been reported in literature available. Hence we report this case with unusual presentation.

Case Report

Sixty nine years, female presented with non-radiating pain in right iliac fossa (RIF) since 2 years. On examination the lump was tender with overlying skin showing discharging sinus which appeared 4 days back.

Since then the patient had fever and constipation. Patient was a known case of inflammatory bowel disease, on treatment since 20 years and had history of jaundice 2 years back. She had no past surgical history or any other medical history. Ultrasonography of abdomen revealed an ill-defined heterogeneously hypoechoic collection with fat stranding in RIF with thick internal echoes within of approximately 10-15cc. A fistulous tract was noted arising from the collection reaching upto external skin. Findings were suggestive of perforated appendicular abscess. Computerised Tomography (CT) scan: anterior wall of caecum appears adherent to right lower quadrant abdominal wall. Defect of approximately 10 mm was seen in the anterior caecal wall with collection of approximate 34x13mm in adjacent abdominal wall with small tract in subcutaneous plane reaching upto the skin. Appendix was visualized and appeared normal. Findings were suggestive of colocutaneous fistula. Biopsy from the tract was sent which revealed acute on chronic inflammation with absence of granulomas or malignancy. Patient underwent right hemicolectomy with fistula excision and specimen was sent for histopathological examination. The total specimen measured 23 cm in length (ileum – 6cm, caecum – 4 cm, ascending colon – 13 cm & appendix – 4cm) with overlying skin flap measuring 6x5x2cm. It showed a fistulous opening of 1 cm in diameter. On cut surface an enterocutaneous fistula was identified measuring 4 cm in length extending from the anterior caecal wall upto the skin surface. There were 2 submucosal polyps incidentally found at the junction of caecum and ascending colon measuring 2cm and 1 cm in the greatest dimension. Both these polyps were sessile with cut surface showing yellowish to whitish areas. (Figure 1)

Microscopy of fistulous tract revealed granulation tissue admixed with dense acute on chronic inflammatory cells with foreign body type giant cells. (Figures 2A,2B & 2C) The sections from polyps revealed submucosal proliferation of mature adipocytes in sheets with fibrous septa containing numerous vessels of varied nature (lymphatics, capillaries, small veins, venules, small arteries, and arterioles) lined by a benign endothelial lining. No vasculitis or thrombosis was noted. (Figure 3A & 3B)The adjacent colonic mucosa showed mild chronic inflammation. Thus the diagnosis offered was colocutaneous fistula with acute on chronic inflammation with angiofibrolipomatous polyps of caecum.

Discussion

ECF has been classified in several ways based on output, etiology, and organ of origin. Based on output, they are classified as high-output (>500 mL/24 hours) , low output (<200 mL/24 hours) and a moderate output (between 200 and 500 mL/24 hours) fistula while etiologically they are classified as iatrogenic and spontaneous. [3] Depending on organ of origin ECF are classified as type I (abdominal, esophageal, gastroduodenal), type II (small bowel), type III (large bowel), and type IV (enteroatmospheric, regardless of origin). [6]. In this case the ECF can be classified as low output, spontaneous and type III (large bowel) type. Submucosal mesenchymal polyps are rare types of intestinal polyps with angiolipofibromas being exceedingly rare. (5). Only few cases of colonic angiolipofibroma have been reported in the literature available [7]. These were polypoidal or pedunculated in nature, located most commonly in the left sided colon. Male predominance with incidence in middle aged men was noted. The microscopy reveals a disorganized overgrowth of mature cells and tissues normally present

in the colonic submucosa thus supporting a benign hamartomatous etiology rather than the possibility of a true neoplasm.[5] Colonoscopy is the gold standard to diagnose colonic polyps. However in this case two angiolipofibromas were diagnosed incidentally, the microscopy of which revealed submucosal proliferation of mature adipocytes in sheets with fibrous septa containing numerous vessels of varied nature (lymphatics, capillaries, small veins, venules, small arteries, and arterioles) lined by a benign endothelial lining. However there is no literature available regarding the synchronous finding of ECF and intestinal angiolipofibromas.The synchronous finding of these two entities raises the possibility of angiolipofibromatous polyp as a etiology behind ECF. However further studies on larger sample size is necessary to prove this association.

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Legend Figures

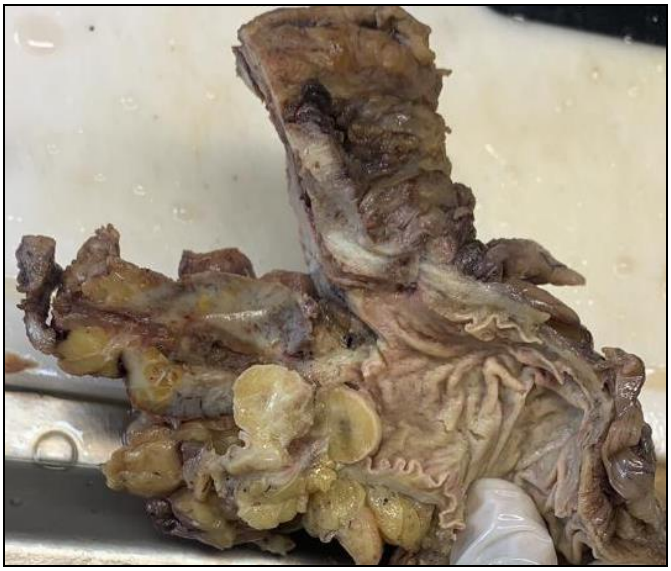

Fig. 1: ECF seen extending from anterior wall of caecum upto the skin surface (black arrow). Polyp with yellowish and whitish cut surface (red arrow)

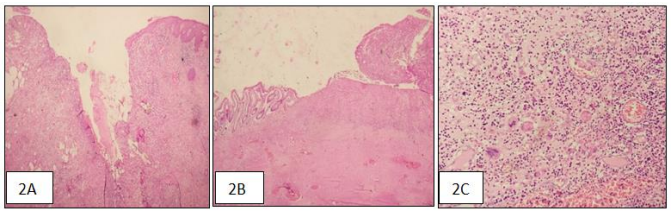

Fig. 2: 2 A: Skin with fistulous tract (H.& E. stain, 10X). 2B: Caecal wall with fistulous tract(H.& E. stain, 10X) 2C: Tract showing acute on chronic inflammation with giant cells (H.& E. stain, 100X)

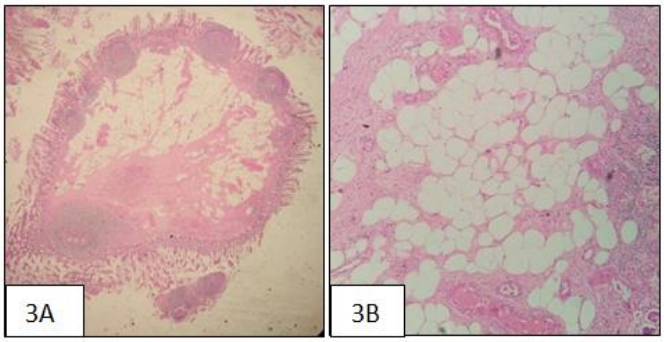

Fig. 3: 3A: Submucosal polyp (H.& E. stain, 10X) 3B: submucosal proliferation of mature adipocytes in sheets with fibrous septa containing numerous vessels of varied nature (lymphatics, capillaries, small veins, venules, small arteries, and arterioles) lined by a benign endothelial lining. (H.& E. stain, 100X)