

Correction of anal prolapse associated with resolution of cloacogenic polyp lesions. Implications to anorectal cancer

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Abstract

Background: We report two rare cases of inflammatory cloacogenic polyps of the anorectum. The first case involves a 50 year old white female who presented with chronic diarrhea, prolapse, and rectal bleeding. The second case presents a 67 year old white male who presented with hemorrhoids and rectal bleeding. It is hypothesized that correction of prolapse will resolve the pathologic changes. In these two patients, at one year follow-up, there has been gradual resolution of pathologic changes following resection of lesions and correction of prolapse with stapled technique.

Investigation: Physical examination, colonoscopy, and hemorrhoidectomy with correction of prolapse

Diagnosis: The pathologic findings of these lesions are presented. A review of the literature in relation to this colonic lesion is presented.

Management: Inflammatory cloacogenic polyps (ICPs) are benign lesions arising from the transitional zone of the anorectal junction and may macroscopically resemble anorectal malignancies. ICPs are being recognized lately with increasing frequency and treatment options are similar as to those for other submucosal lesions of the colon and may include conservative therapy, and endoscopic resection for small lesions. Surgical resection for larger lesions can be used as treatment if there is a threat of obstruction, a question of underlying malignancy, or if they are prolapsed induced. Surgery is

the most common path for treatment. Surveillance is necessary with patients that show severely dysplastic ICPs since they have been associated with anal neoplasias as well as squamous cell carcinomas. It is hypothesized that correction of the prolapse will resolve the pathologic changes.

Keywords: Inflammatory Cloacogenic Polyps, lesions of the colon, mucosal prolapse, cancer.

The cases

Case 1 involves a 50-year-old white female with a history of emphysema, chronic diarrhea, and diverticular disease. She reported having a black tarry stool and intermittent episodes of bright red blood per rectum. Her medication included Excedrin[®], Advair 250[®], Spiriva[®], and Sbuterol[®]. Her past surgical history included a hysterectomy and a Frykman-Goldberg procedure. The patient reported alcohol use to be 6 beers per week and having quit tobacco use in 2002. Her family history was negative for colon cancer in any first degree relative. On physical exam there was no evidence of adenopathy. Chest and abdominal examinations were normal. Rectal examination revealed significant anal prolapse and a degree of failure of the anal sphincter. Colonoscopy was performed and revealed diffuse diverticular disease with narrowing of the sigmoid colon, erythema around the splenic flexure, and large anterior anal polyp. Biopsies of the polyp were found to be non-malignant. The patient was then scheduled for a stapled hemorrhoidectomy with prolapse correction, which was performed 2 days later. During hemorrhoidectomy the prolapse was treated and multiple anterior anorectal tumors were resected. Pathologic investigation post procedure showed that upon microscopic examination, three inflammatory cloacogenic polyps of the anal transition zone were found. Polyp 1 was found in the anterior rectum, was tan-pink in color, and was 5 cubic centimeters in size. Polyp 2 was found in the right superficial rectum, was also tan-pink in appearance, and was 0.96 cubic centimeters in size while Polyp 3 was found in the left part of the rectum, was also tan-pink in appearance, and was roughly 4.9 cubic centimeters in size. The polyps had moderate to severe dysplasia, with the larger polyps being the more dysplastic. The dysplasia was focused in the squamotransitional epithelial components. This dysplasia was monitored closely as it became chronic on biopsies on follow up colonoscopies. A Human

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papillomavirus (HPV) test was suggested to the patient as literature suggests some viral involvement in the inflammatory cloacogenic polyp and anal intraepithelial neoplasia. A year after the correction of the prolapse her problems with severe dysplastic polyps have shown a gradual resolution.

The second case 2 involves the presentation of a 61 year old white male with a history of heart disease, hypertension, and diabetes. He reported having intermittent episodes of bright red blood per rectum. His medication included Plavix[®], simvastatin[®], Glipizide[®], HCTZ[®], among other medicines that were needed for his health problems regarding diabetes and heart disease. His past surgical history included heart stents and a left eye injury. The patient denied alcohol use and had quit smoking in 2007. On physical exam there was no evidence of adenopathy. Chest and abdominal examinations were normal. The patient was discovered to have had class IV prolapse upon rectal exam. The patient's last colonoscopy was 10 years ago. The patient had the hemorrhoidectomy procedure done, where the prolapse and an associated mass of hemorrhoids were removed. The hemorrhoids were of an unknown severity due to a large anterior mass that was associated with them. Pathology later showed upon microscopic examination using Hematoxylin and Eosin (*H&E*) staining that the rectal mass was an inflammatory cloacogenic polyp that was negative for adenomatous change and malignancy. Upon resolution of the prolapse and resection of the neoplasm the patient's symptoms of rectal bleeding and pain were resolved, however given the presence of the polyp a colonoscopy will follow to monitor any changes.

Discussion of diagnosis

Inflammatory Cloacogenic Polyps (ICPs) is a rare type of anorectal polyp that was first described in literature by Lobert and Appelman in 1981 (Lobert & Appelman 1981). ICPs arise from the epithelial transition zone at the anorectal junction and have a distinctive histological appearance (Mathialagan et al. 2000). As defined by Walls the anal canal commences at the level of the levator ani muscle and extends distally 3-5 cm to the anal margin (Walls 1958). The anal margin is a subjective point positioned at the inferior margin of the internal anal sphincter. Squamous mucosa lines the distal canal while more colorectal type mucosa ranks the superior aspect. Between these two regions lies the anal transition zone. The lining of this zone includes anal ducts and glands with stratified columnar or transitional epithelium (Lobert & Appelman 1981) and the zone is only 0.5-1.2 cm long (Mathialagan et al. 2000). ICPs have become a term that describes a polypoid mucosal prolapse that occurs in the anorectal area and is distinguished by ulceration and erosion on the surface. Other features may include villiform hyperplasia, crypt architectural disarray, fibromuscular proliferation within the lamina propria, and variable degrees of chronic inflammation (Parfitt & Shepherd 2008). They are characterized by a tubulovillous pattern of growth. There is no set age of occurrence for these lesions, and cases have appeared in adults as well as children (Washington et al. 1993; Bass et al. 1995; Poon et al. 1997). The most common symptoms of ICPs are bleeding per the rectum, straining to defecate, swelling of the anus, and pruritis (Lobert & Appelman 1981; Mathialagan et al. 2000). These symptoms are probably not caused by the polyp themselves, but rather lead to endoscopic investigation which inadvertently leads to the discovery of the polyp (Mathialagan et al. 2000). They are classically associated with hemorrhoids and local prolapse (Parfitt & Shepherd 2008). Mucosal prolapse accompanies 50% of reported ICP cases (Levey et al. 1994) and therefore it has been proposed that ICPs is due to mucosal prolapse (Levey et al. 1994; Saul 1987; Du Boulay et al. 1983). ICPs may mimic adenomas and even adenocarcinomas, both endoscopically and histologically (Parfitt &

Shepherd 2008). There are reports of squamous cell carcinoma *in situ* arising in ICPs (Saul 1987; Hanson & Armstrong 1999; Jaworski et al. 1969) and in some of these cases HPV type 16 was demonstrated in the areas of squamous carcinoma *in situ* in the polyps by polymerase chain reaction (PCR) (Jaworski et al. 1969). A rare case of anal intraepithelial neoplasia has been reported in literature to be associated with ICPs. As such it has been attempted to use the p53 gene, Ki67 gene, and proliferating cell nuclear antigen (PCNA) as a marker to be used in identification of these polyps. ICP did not show a consistent pattern of immunoeexpression of PCNA¹³ which is also used as a DNA damage marker. The p53 and Ki67 (MIB-1) have shown strong presence in dysplasia and is suggested to be used in challenging histopathological cases where interpretation is difficult. Although there are cases showing squamous carcinoma *in situ* arising from ICPs the literature pool on ICPs are rather shallow to make a definitive assumption that the occurrence of carcinoma in these lesions is more common than not.

Differential diagnosis

The differential diagnosis of lesions of the colon involves malignant and benign lesions. Malignant lesions include carcinomas of the anus and rectum. The differential diagnosis of benign submucosal lesions of the colon includes fibroblastic polyps, adenomas with secondary prolapse, and Solitary Rectal Ulcer Syndrome (SRUS), inflammatory cap polyps, and inflammatory myoglandular polyps (Parfitt & Shepherd 2008).

Endoscopic appearance

Endoscopic findings of the ICPs may show polypoid growth low in the rectum. It may be pedunculated (Saul 1987) or more commonly sessile (Mathialagan et al. 2000). ICP may materialize as a villiform, tumorous mass at the anorectal junction, and to the histopathologist the incidence of villiform epithelial hyperplasia and noticeable regenerative 'atypia' may cause bewilderment with a villous adenoma (Mathialagan et al. 2000). It may also mimic anorectal cancer or in some cases have squamous cell carcinoma *in situ* (Saul 1987; Hanson & Armstrong 1999; Jaworski et al. 1969). Further, the occurrence of misplaced glands, with or without mucus lakes, inside the submucosa may produce mural thickening, which may imitate carcinoma radiologically, as well as causing histopathological confusion with invasive adenocarcinoma (Parfitt & Shepherd 2008).

Treatment and management

Literature has reported that some patients have decided ultimately not to resolve the ICPs taking solace that it is not something more sinister. Thus a conservative high fiber diet may be suggested in helping to alleviating some of the complaints of the patient. However literature (Mathialagan et al. 2000) and the experiences of these cases suggest that by resolving the prolapse through surgical resection ultimately resolves the pathologic changes. In the case of the two patients reported in this manuscript, at one year follow up, there has been gradual resolution of pathologic changes following resection of lesions and correction of prolapse with stapled techniques. This surgical resection of the prolapse should not be a major nosocomial event for the patient as the procedure is usually outpatient, and post surgery complaints usually resolve themselves in a short period of time. Surveillance should be continued even after resection of the lesions and prolapse, especially if future lesions are abundant and signal severe dysplasia. HPV should be tested for to ensure that the lesions are not viral in origin.

Conclusions

With the increasing utilization of modern medical technology and in the course of investigations of symptoms and regular preventative screenings for colorectal cancer ICPs may be discovered incidentally. These submucosal lesions may be associated with nonspecific symptoms and have a low risk of malignancy, though risk of degeneration to cancer is both real and difficult to quantify. Treatment of these lesions may involve conservative surveillance, polypectomy, or in view of these findings here correction of prolapse is recommended. The decision of whether to treat and choice of modalities to use may be dependant on the size of the lesion, histologic features, and the risks of procedure. However it would seem by these cases reported that correction of the prolapse ultimately leads to the resolution of the ICPs and the complaints of the patient. In the female patient sequential histologic examinations showed gradual resolution of dysplasia following correction of prolapse which is suggestive of causality.

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Contributions

Dr. Morfesis was responsible for the treatment and management of the patients. And contributed to the treatment and management section, as well as provided information on the patients.

Dr. A. Georgakilas provided commentary and analysis of the molecular aspects of the polyps regarding the p52 and Ki67 genes, and PCNA. He also participated in the preparation of the manuscript and had the overall editing responsibilities.

Mr. Kalogerinis was responsible for the overall preparation of this manuscript and provided the literature review.

Dr. S. Georgakila provided insights into the clinical aspects of the findings and participated in the preparation of the manuscript.

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