Clinics in Surgery

Case Report
Published: 08 Jul, 2016

Fistula in Ano and Carcinoma: Primary or Secondary?

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Abstract

Because of rarity and depth of the lesion, the carcinoma arising in fistula in ano is often diagnosed late resulting in need for radical surgery and poor outcome. We at our institute analyzed the records for last 10 years and found 3 such cases. In 2 patients the fistulae were long standing of more than 10 years duration and in one patient the fistula of 1 year duration was reported to have a focus of adenocarcinoma in the excised specimen. He received adjuvant radio chemotherapy and is on follow up. All the three were reported to have mucinous adenocarcinoma. One patient underwent abdominoperineal resection (APR) followed by radio chemotherapy, the others received neo adjuvant therapy and is on a diversion colostomy waiting for APR. A long standing and recurrent fistula should raise a high index of suspicion and be examined and biopsied preferably under anesthesia. MRI provides a precise assessment of extension of the lesion. The treatment should be aggressive and radical.

Introduction

Carcinoma arising in a fistula in ano is a rare complication. No more than 200 cases have been reported in the literature. It accounts for only 2-3% of large bowel cancers [1]. Malignant changes are supposed to occur in long standing fistula due to chronic inflammation and irritation. This is an aggressive cancer usually of mucinous variety and often misdiagnosed clinically. Diagnosis is usually made late because the tumor develops outside bowel wall deep into the ischiorectal fossa and perineum [2]. Sometimes one is confused as to whether cancer has developed denovo or secondary to fistula. We present our cases and experience in our teritiary care institution with review of relevant literature for readers as a guide for early diagnosis and differentiating the two entities.

Case Presentation

45 years old male, presented with recurrent fistula in ano with four attempts at repair in previous 10 years. Last surgery was done 3 years back. This time patient presented with perianal discharge. On examination patient was found to have multiple skin tags & scars in the perianal region. Digital examination revealed a sinus at 4 O'clock position going deep into ischiorectal fossa and discharging mucinous material with previous scars around (Figure 1). Lower GI Endoscopy was normal. MRI revealed a deep cavity in the region of right rectal fossa opening in the perianal region at 8 o'clock position containing some echogenic material. Intra operative findings revealed a deep cavity in the region of right perianal region with 1 cm opening at 4 O'clock position at anal verge containing mucinous material and soft granulation tissue which was curetted out and sent for histopathological examination. Histology revealed mucinous adenocarcinoma with some skin cells. Patient refused abdominoperineal radical resection and was subjected to diversion colostomy at some different place. Two months later patient presented once again with continued perianal discharge. Chemo-radiotherapy was planned. Three months after completing neo adjuvant therapy, biopsy from the cavity revealed no residual tumor. Repeat biopsy 3months later showed presence of cancer cells. Wide local excision was done (Figure 2 and 3). Patient awaits reconstruction of wound by local flap subjected to clearance of disease. A 38 year old male patient presented with recurrent fistula in ano. He has started a perianal abscess about 11 years back. The abscess was drained that time but persisted with perianal discharge on and off for about 2 years when he underwent fistulectomy for a transsphincteric fistula as per the records. The wound healed in about a months' time. Three months later he developed perianal swelling at the site of scars which burst with discharge of pus. He continued with recurrent episodes of perianal discharge for another 3 years and was operated by the same surgeon. Same procedure was done. The wound healed in about 6weeks time. Patient did not have any complaint for another 9 months when he noticed again a discharging sinus just near the anal verge. The discharge would be mucoid and sometimes purulant. However it did not give him any discomfort and it continued for about 3 years, when he consulted another doctor who

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Received Date: *06 Jun 2016*
Accepted Date: *04 Jul 2016*
Published Date: *08 Jul 2016*

Citation:
Chowdri AN, Fazl NS, Parray Q, Wani AR, Thakur N. Fistula in Ano and Carcinoma: Primary or Secondary?. Clin Surg. 2016; 1: 1062.

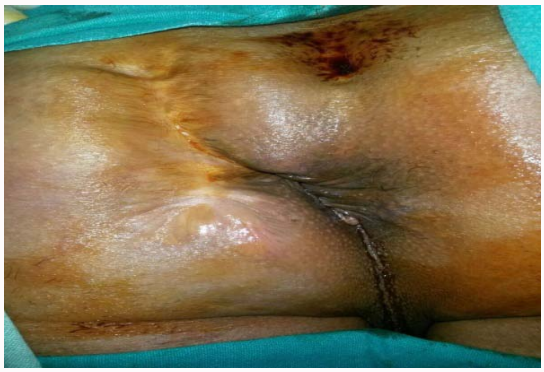

Figure 1: Scars of previous surgeries.

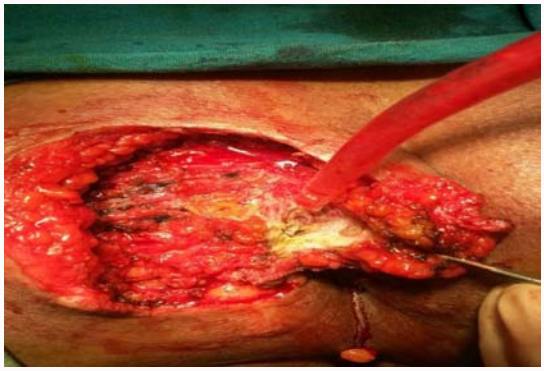

Figure 2: Wide local excision of cancerous lesion.

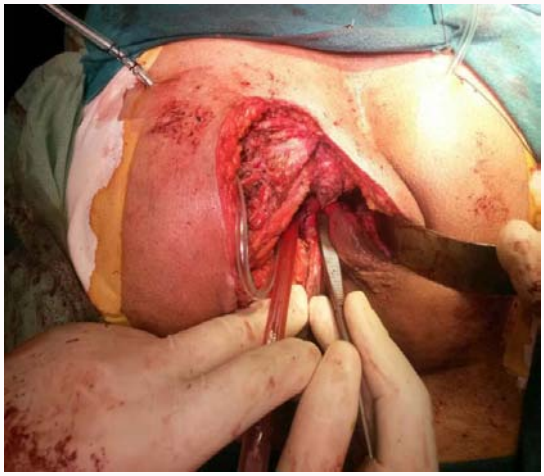

Figure 3: Showing defect with multiple cavities after wide excision.

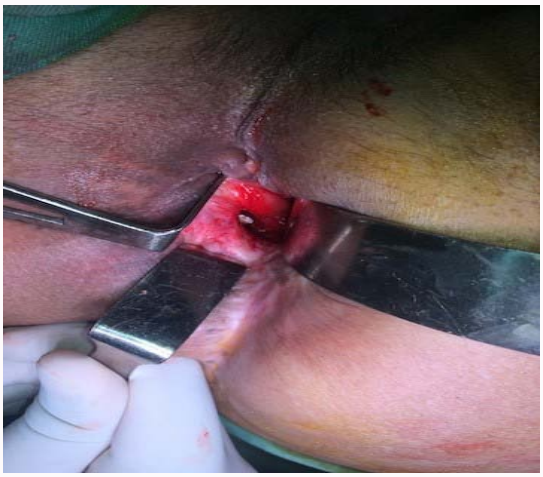

Figure 4: Showing external opening of cavity.

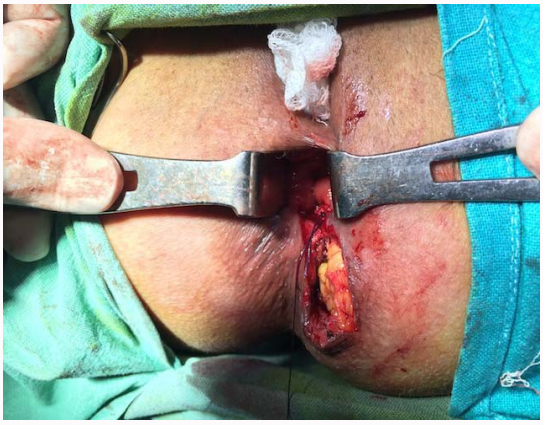

Figure 5: Revision excision of the fistula site.

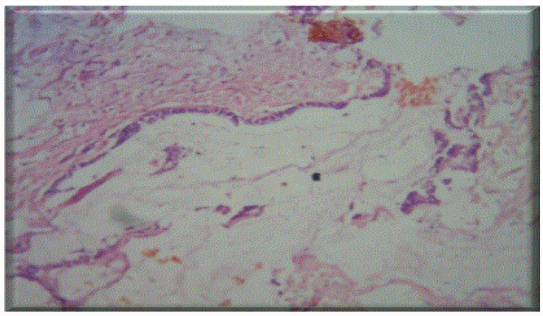

Figure 6: Showing mucinous adenocarcinoma.

advised surgery which he refused but he continued with this draining sinus in the perianal region for 2 more years when he presented to us. He had few attacks of pain, discomfort and indurations which would get relieved spontaneously with discharge of pus. When patient presented to us there was about 7-8 mm opening at 8 O'clock position just 1cm from anal verge discharging mucoid material with some tenderness in the perianal region (Figure 4). No additional internal opening was detected on digital rectal examination. Sigmoidoscopy was normal. MRI revealed a cavity in the area of right ischio-rectal fossa containing echogenic material communicating with the anus just near anal verge on right side. Curratings form the cavity revealed mucin secreting adeno carcinoma with apparently no pelvic or groin lymph adenopathy. Patient was advised radical excision of the lesion with possibility of permanent stoma. However he refused permanent stoma. The cavity was explored and excised. Following excision the wound collapsed and healed in 3 weeks time. Patient was subjected to chemo radiotherapy and he is apparently disease free at 2 year follow up.

Another female patient aged 55 years was referred for further management. She was operated for fistula in ano of one year duration. She had a history of recurrent perianal discharging sinus. Fistulectomy was done and specimen was reported as moderately differentiated Adenocarcinoma. There was no inguinal adenopathy. CECT did not reveal any distant disease. Revision wide excision of healed scar was done (Figure 5). However there was no residual

tumor in the respected specimen. Patient was subjected to adjuvant therapy. She is disease free at a follow up of 15 months.

Discussion

Mucinous adenocarcinoma associated with perianal fistula is a rare cancer & constitutes 3-11% of all anal cancers [3]. The cancer may arise de novo and present as fistula or it may arise from a long existing perianal fistula or an abscess cavity. This sometimes becomes a controversial issue as to whether the fistula is the source of tumor or the presenting feature of the anorectal carcinoma. Obviously the carcinomatous changes are due to long standing sepsis and chronic inflammation and irritation leading to metaplasia but unfortunately due to its rarity etiological relationships is not well documented in the literature [4]. Although it is a known fact that perianal fistula and abscess arise from the anal glands and ducts but the origin of carcinoma arising from these ducts and glands is debatable. Some are of the opinion that carcinomatous changes are due to malformation of intestinal tract with nests of cells being source of the disease [5]. Fistula in ano presenting in perianal discharge has also been reported to develop from a pre- existing carcinoma [6]. Kline et al. [7] documented that carcinoma from perianal fistula was associated with carcinoma elsewhere in the colon. Perianal fistula associated adenocarcinoma has also been reported in Crohn's disease [8]. Yamada K et al. [9] stressed the need for special attention to be given to similar conditions associated with malignant disease. Our first patient had a long standing recurrent fistula with a deep cavity in the perianal region which was ignored by the patient and probably missed by the physician as well. Stasis with chronic irritation and inflammation in this cavity could be the reason for metaplasia and development of cancer. The second patient had a chronic history of recurrent fistula with formation of cavity in the ischio rectal fossa which was left untreated for years together. Again the chronicity, stasis and irritation could be the reason for development of cancer in this cavity. The cavity being deep in the perianal region misleads the patient and the surgeon about the progress and nature of the disease. The other patient had a simple fistula for which she underwent fistulectomy. In this patient probably fistula had develop secondary to underlying carcinoma as chronic inflammation and irritation is must for metaplasia to occur in an underlying fistula or cavity. Early diagnosis is difficult in carcinoma associated with fistula in ano due to insidious nature of the disease and masking of symptoms. Malignancy should be suspected in a chronic perianal fistula which shows early in duration, continued discharge especially mucinous and which does not disappear following surgery. There may be isolated areas of malignant transformation separated by inflammatory zone. Therefore it is advisable to take multiple biopsies from suspected lesions. Malignant changes usually occur in deep seated cavities as was the case in our first patient. The pathology is usually of mucinous adenocarcinoma (Figure 6). Some have reported low grade adenocarcinoma in about 44% cases [10]. Once diagnosed the treatment should be radical i.e., abdominoperineal resection with the permanent stoma. Large defects following wide excision sometimes need to be reconstructed by myocutaneous flaps. Depending on the stage of the disease, adjuvant or neo-adjuvant therapy should be recommended.

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