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Leiomyosarcoma of the Recto-Sigmoid Junction Prolapsing out of the Anus: A Case Report

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Abstract

Leiomyosarcoma is a rare mesenchymal neoplasm in the gastrointestinal tract which has similar microscopic and macroscopic characteristics to GISTs. In this article we present an unusual presentation of this rare tumour which posed us a management challenge. A 73-year-old male presented with bleeding per rectum and was found to have an obstructing growth at recto-sigmoid. While awaiting tissue diagnosis, this tumour prolapsed out of the anus with bleeding. To control bleeding and to relieve pain trans-anal excision of the tumour was done. Subsequently the patient underwent laparoscopic anterior resection for a recurrent tumour. Histology and immunohistochemistry confirmed the diagnosis of a leiomyosarcoma. Leiomyosarcomas carry a poor prognosis and should be always differentiated from GISTs using immunohistochemistry. Radical surgery is the main stay of management.

Keywords: Leiomyosarcoma; Prolapse; GIST

Introduction

Leiomyosarcoma is a mesenchymal neoplasm which can be found mainly in the uterus, retroperitoneum and rarely in the gastrointestinal tract [1-3]. Leiomyosarcoma share similar macroscopic and microscopic characteristics with Gastrointestinal Stromal Tumours (GIST). Accurate differentiation of the two is important in their management and prognosis which became possible only after the discovery of their immune-histochemical identities. Since then, diagnosing a primary leiomyosarcoma in the gastrointestinal tract became so rare to the extent that there is limited data on demography, clinical features and gross features of these tumours [1,4,5].

Bleeding per rectum and alteration of bowel habits are the commonest presentations of tumours of the le t sided colon and rectum [6]. Rare cases of colorectal adenocarcinomas presenting with rectal prolapse has been reported in literature [6-8]. There have been some case reports of patients with polyps and polyposis syndromes presenting with a prolapsed polyp [9-11]. In this background where tumour prolapse is an atypical presentation of cancer, a patient with a recto-sigmoid leiomyosarcoma causing tumour prolapse is an extreme rarity **(Figure 1)**.

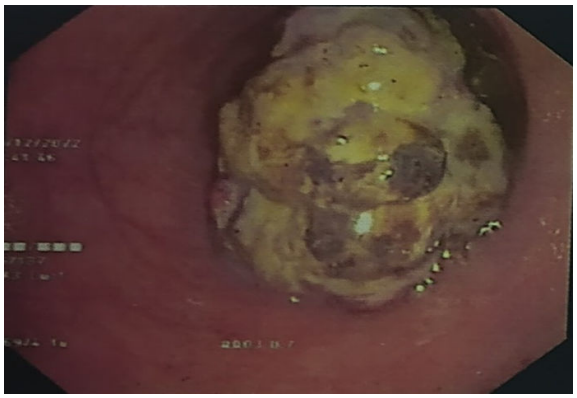

Figure 1: At flexible sigmoidoscopy before prolapse.

Case Presentation

A 73-year-old male patient with hypertension and stage II chronic kidney disease presented with bleeding per rectum and discomfort while passing stools for 3 months.

On examination, he was not pale and the abdominal examination was unremarkable. On digital rectal examination an apex of a mass was felt 5 cm from the anal verge. Flexible sigmoidoscopy showed a malignant looking polypoidal growth at 15 cm from the anal verge. The scope could not be negotiated beyond the lesion. Multiple biopsies were taken and the patient was admitted for further investigations for diagnosis and work up for surgery in suspicion of a rectal carcinoma.

Contrast Enhanced Computed Tomography Scan (CECT) demonstrated a large polypoidal growth almost completely obliterating the rectosigmoid junction with possible infiltration

into the serosa. There was no evidence of regional lymphadenopathy or distant metastases.

During the ward stay he developed several episodes of per rectal bleeding with his bowel motions. One day when passing stools, he had a severe bout of pain with a significant amount of blood and a mass prolapsing out of the anus. On the first occasion this was manually reduced without much difficulty. On the next day it prolapsed again and could not be reduced manually. As it was painful and the prolapsed mass was bleeding continuously, the patient was taken to theatre for examination under anaesthesia.

While the patient was in the lithotomy position, we noted a 10 × 10 cm sized mass with bleeding projecting out of the anus **(Figure 2)**. It was connected by a narrow pedicle. Our plan was to divide this mass from the pedicle while ensuring haemostasis. We divided the pedicle using an ultrasonic dissector and by transfixing with 3-0 polyglactin sutures. Subsequent digital rectal examination did not reveal any active bleeding from the divided pedicle. The specimen was sent for histology.

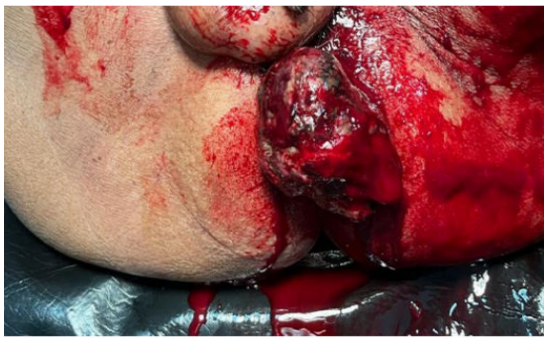

Figure 2: Prolapsing tumour out of the anus with bleeding just before trans anal excision.

Post-operatively he was asymptomatic and was discharged with a plan to review with histology. The histology reported a leiomyosarcoma with its spindle cell architecture and supportive immunohistochemical studies. With our available resources, Smooth Muscle Actin (SMA), *Desmin*, S100 and CD34 studies were done. It was found to be SMA and *Desmin* positive and CD117, S100 and CD34 negative. It was reported independently by two consultant pathologists and final conclusion of a leiomyosarcoma was made. The resection margin was involved by the tumour **(Figure 3)**.

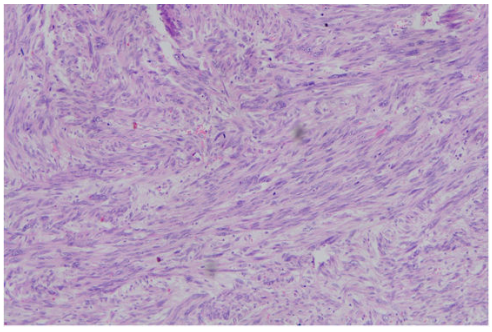

Figure 3: Hematoxylin and Eosin stained sections showing spindle cell architecture.

One month after the trans-anal excision, a flexible sigmoidoscopy was performed to see the status of the tumour bed and to plan further management. Sigmoidoscopy revealed a recurrence of the tumour at the recto-sigmoid junction. Following a multi-disciplinary team meeting, the patient was planned for an anterior resection **(Figure 4)**.

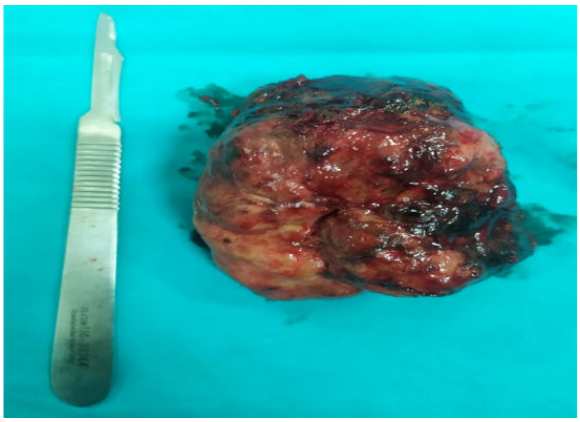

Figure 4: Tumors after trans-anal excision.

After a brief period of prehabilitation, he underwent a laparascopic anterior resection. Post-operative recovery was uneventful and the patient was discharged home on day five following surgery. Histology confirmed the diagnosis of leiomyosarcoma with negative resection margins and two out of four lymph nodes showing tumour deposits. Tumour was of grade 3 under French Federation Nationale des Centers de Lutte Contre le Cancer (FNCLCC) system.

Subsequently the patient was referred to the oncologist for adjuvant therapy. While waiting for chemotherapy the patient developed a myocardial infarction from which he recovered. However, he was deemed unfit for further adjuvant therapy.

Result and Discussion

Cancers arising in the gastro-intestinal tract are mainly epithelial in origin and out of colorectal malignancies commonest is adenocarcinoma [12]. Primary mesenchymal sarcomas are so rare in the gastrointestinal tract out of which commonest is leiomyosarcoma [4]. However, following advances in immunohistochemistry, it was realized that most of the mesenchymal neoplasms previously identified as leiomyosarcomas turned out to be GISTs. Since then, diagnosis of a leiomyosarcoma in the gastrointestinal tract became a rare entity [4,5].

Individual case reports and case series identify abdominal pain, lower gastrointestinal bleeding and abdominal mass as common presentations of colorectal leiomyosarcomas [13-15]. Intestinal obstruction, perforation and acute suppurative peritonitis are rare presentations reported in individual case reports [4,14,15].

Tumour prolapse with or without full thickness rectal prolapse is a rare presentation even for colorectal adenocarcinomas [6-8]. Tumour prolapse percise is almost unheard of except for case

reports of prolapsing polyps [10,11]. A clinical note published in Saudi Medical Journal in 2007 reports a rectal GIST presenting with prolapse [16]. Rectal and rectosigmoid carcinomas presenting with full thickness rectal prolapse have been reported. Presence of tumour at the apex of the prolapse in these cases indicate the fact that these tumours might have acted as the lead point for intussusception and subsequent prolapse [6–8].

The novelty of this presentation posed us a management challenge. This painful and bleeding tumour outside the anus needed to be attended to as soon as possible. Given the patient's age and his comorbidities and fact that we did not have a proper tissue diagnosis at that time, relieving his symptoms with least morbidity was our objective. This trans-anal excision relieved of his symptoms but a definitive management strategy was needed after the tissue diagnosis **(Figure 5)**.

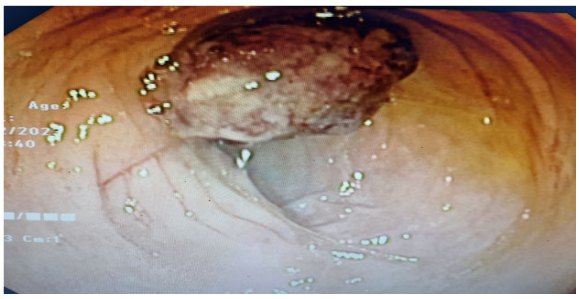

Figure 5: Follow up sigmoidoscopy after trans-anal excision showing recurrent tumor.

GISTs and leiomyosarcomas are macroscopically and microscopically similar [17]. GISTs originate from the interstitial cells of Cajal and leiomyosarcomas has its origin from smooth muscle cells [1]. Owing to this difference in origin they have distinct immunohistochemical identities and prognosis [1,3-5,17,18].

Leiomyosarcomas carry a poor prognosis by being aggressive tumours with high chance of local recurrence and metastasis, poor overall survival, absence of a targeted therapy and poor response to chemotherapy [1,2,4,18]. In 1998, Hirota et al., discovered the oncogenic role of c-kit mutations in GISTs and it paved way for the place of targeted therapy with tyrosine kinase inhibitors [3,5,17]. Subsequently KIT (CD117), CD34 and DOG1 were identified as immunohistochemical markers with higher sensitivity and specificity in the diagnosis of GISTs. SMA, *Desmin* and h-caldesmon are immunohistochemical markers which indicate smooth muscle cell origin of leiomyosarcomas and these markers are absent in GISTs [3].

In this patient SMA and *Desmin* were positive while CD117 and CD34 were negative. S100 is a schwannoma marker protein which was also negative supporting the diagnosis of a leiomyosarcoma.

As the initial resection margin was positive and the subsequent endoscopic examination showed the presence of recurrent tumour, definitive surgery to achieve an oncological resection was warranted. According to the current literature radical resection is the main mode of management. The role and the effectiveness of chemotherapy and radiotherapy in management is still not established [4,17].

Conclusion

Leiomyosarcomas are a rare type of mesenchymal tumours occurring in the gastrointestinal tract which should be differentiated from GISTs. They are aggressive tumours with poor prognosis and surgery remains the mainstay of management.

Ethics Approval and Consent to Participate

Not applicable.

Consent for Publication

This case report was prepared for publication with the informed written consent of the index patient and relatives. All images were taken and included with consent.

Conflicts of Interests

Non declared.

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Not applicable.

Author's Contributions

GBKDB collected relevant information and formulated the case report with guidance from AA and MIMDA. MIMDA did the final review. All authors read and approved the final manuscript.

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