



Multifocal Small Bowel Adenocarcinoma Presenting as Multiple Strictures Mimicking Tuberculosis – A Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aims: This article tries to highlight the importance of suspecting Small bowel adenocarcinoma in a patient with multifocal small bowel strictures in tuberculosis endemic regions like India.

Presentation of Case: A 54 year old male presented with colicky central abdominal pain for 4 weeks. Initial evaluation suggested the possibility of Intestinal tuberculosis. He presented again 2 weeks later with acute intestinal obstruction and was taken up for emergency surgery which revealed multiple non passable strictures in jejunum and ileum which were resected and multiple anastomoses were done. Biopsy revealed multifocal primary small bowel adenocarcinoma without any evidence of predisposing factors for small bowel adenocarcinoma.

Discussion: Acute intestinal obstruction is one of the commonest indications for emergency laparotomies and obstruction secondary to small bowel strictures in the Indian subcontinent is most commonly due to Intestinal Tuberculosis with Crohn's disease being a distant second entity. Small bowel adenocarcinoma is not only rare but also presents with nonspecific findings making it difficult to diagnose. Surgical resection is the cornerstone of management and standard chemotherapy regimens are still evolving. Small bowel adenocarcinoma most commonly presents as a solitary

infiltrating mass and hence our case is very unique in its presentation as multiple malignant strictures in jejunum and ileum. Despite extensive histological and radiological evaluation of our patient, there were no known predisposing factors like Crohn's disease or polyposis that can explain the multifocal presentation of small bowel adenocarcinoma.

Conclusion: Even with all the latest advancement in the field of diagnostics, preoperative diagnosis of small bowel adenocarcinoma is still uncommon. We are reporting this case to stress the importance of having a differential of small bowel adenocarcinoma in intestinal obstruction patients with multiple strictures of small bowel.

Keywords: Multifocal small bowel adenocarcinoma; multiple strictures; Intestinal Tuberculosis; Case Report.

ABBREVIATIONS

SBA : Small Bowel Adenocarcinoma

ITB : Intestinal Tuberculosis

IBD : Inflammatory Bowel Disease

SBO : Small Bowel Obstruction

TB : Tuberculosis

1. INTRODUCTION

Small bowel obstruction (SBO) is one of the commonest indications for visiting the surgical emergency and it accounts for around half of all the emergency laparotomies done each year in the United Kingdom [1]. The aetiology of SBO differs between countries, with adhesive SBO being the most common cause in the Western countries [1]. But in the Indian subcontinent the most common causes are equally distributed between Intestinal Tuberculosis (ITB) [2], obstructed hernia [3] and adhesive SBO [4]. Malignancies are a rare cause of SBO all across the world.

Small bowel malignancies are most commonly adenocarcinomas (30 – 40%) and carcinoids (35-42%) followed by lymphomas and sarcomas [5]. Small intestine cancer represents 0.6% of all new cancer cases [6] and 2.3% of all digestive tract cancers [7]. Duodenum is the most commonly involved segment with jejunal or ileal involvement being rare [8]. Most Small Bowel Adenocarcinomas (SBAs) are solitary lesions, presenting with vague abdominal symptoms, leading to a delayed diagnosis.

The present case report is of a similar patient who presented late with intestinal obstruction due to multiple strictures of jejunum and ileum initially diagnosed as ITB. Postoperative histopathology proved to be a surprise revealing primary de novo multifocal small bowel adenocarcinoma with no evidence of Tuberculosis or Crohn's

disease. Hence the current report is made to highlight considering SBA as a differential in patients presenting with small bowel obstruction even in the absence of classical risk factors.

2. PRESENTATION OF CASE

A 54 year old man from northern India presented to the surgical emergency with complaints of colicky central abdominal pain for 4 months with exacerbation of symptoms for the last one week. He did not have any significant past medical condition or family history. He was moderately built with BMI of 19.2 kg/m². Clinical examination revealed normal hemodynamics and an ill-defined lump in right iliac fossa with raised bowel sounds. Rest of the examination was normal. The patient was evaluated as partial bowel obstruction secondary to ileo-caecal tuberculosis.

Routine biochemical investigations were unremarkable. Initial ultrasound abdomen showed dilated small bowel loops. Examination of tumour associated antigens revealed normal values for carcino-embryonic antigen (CEA) and carbohydrate antigen 19-9 (CA 19-9). CECT of abdomen and chest revealed dilated distal jejunal and proximal ileal loops, with thickened, matted distal ileal loops adherent to the anterior abdominal wall with peripherally enhancing hypo dense thickening of ileo-caecal region and multiple subcentimetric mesenteric lymph nodes (Fig 1). The rest of the abdominal organs and the lungs were normal. Colonoscopic evaluation was normal. Diagnosis of Intestinal tuberculosis was suggested based on clinical and radiological evaluation.

He underwent expectant management and was discharged a week later in stable condition with a plan for small bowel enteroscopy and biopsy to be done at a higher specialised centre to confirm

ITB. But two weeks later he again presented to our emergency with acute intestinal obstruction. Repeat CECT abdomen showed short segment abrupt concentric narrowing in distal ileum with proximal bowel loops dilated. Patient was taken up for emergency laparotomy which revealed multiple non-passable strictures (10 strictures totally) in small bowel starting from 50 cm distal to ligament of Treitz to 20 cm proximal of ileo-caecal junction (ICJ), suggestive of Intestinal

Tuberculosis with second differential of Crohn's disease (Fig 2). With these possibilities in mind, two segments of jejunum measuring 20 cm and 8 cm each (located 50 cm and 90 cm distal to ligament of Treitz respectively) and one segment of ileum measuring 10 cm (located 30 cm from ICJ) were resected and anastomosed. Peritoneal and omental biopsies were also taken. Large bowel, liver and rest of the abdomen were normal.



Fig. 1. Contrast enhanced CT scan of the abdomen (coronal view) showing thickening of ileo-caecal region (arrow) with mesenteric lymph nodes

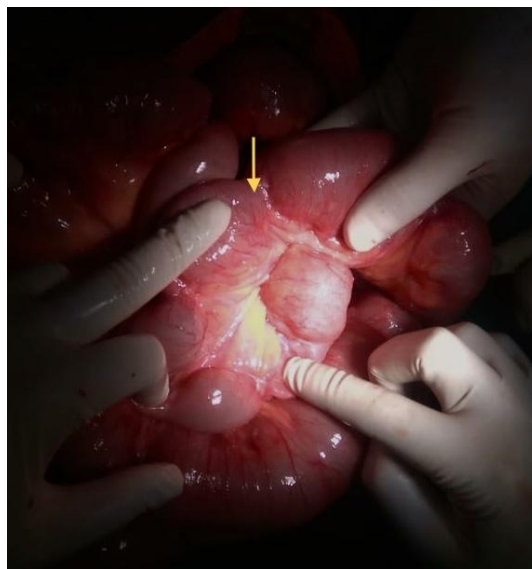


Fig. 2. Intraoperative photograph of involved bowel segment (arrow showing stricture in jejunum)

Histopathological report confirmed a primary de-novo multifocal adenocarcinoma small bowel (grade 2- moderately differentiated) in all the three resected segments with largest focus of 2 x 1.5 x 1 cm (Fig 3). No nodal metastasis was found in 5 resected lymph nodes with final tumour stage being T4N0M0 (Stage II B). No histological evidence of Tuberculosis, Crohn's disease, Celiac disease, polyps or adenoma was found in any of the specimen. The patient developed anastomotic site leak with intra-abdominal sepsis post operatively and was planned for re exploration but unfortunately he developed multiple organ dysfunction syndrome and he died on post-operative day 5.

3. DISCUSSION

In the Indian subcontinent if a SBO patient comes to the emergency, without any hernia or a previous history of abdominal surgery, then the first diagnosis is ITB. The most common mode of acute presentation of abdominal TB is Intestinal obstruction, with TB accounting for 10% of all surgical emergencies in India [9]. The most common pathology causing obstruction in TB patients is small bowel strictures, often multiple in number and most commonly located in Ileum followed by jejunum [2], [9].

The aetiology of small bowel strictures differ starkly between developed and developing countries with Crohn's disease being one of the leading causes in the former and ITB in the later (Table 2) [2], [10], [11], [12]. Malignant strictures

are extremely rare in both the regions of the world.

SBA is a remarkable disease, not only in its rarity but also because it presents a challenge both in diagnosis and treatment. Due to patients presenting with non-specific abdominal symptoms and lack of any screening investigation, diagnosis of SBA occurs at advanced stages; with 39% of patients having lymph node metastasis at the time of diagnosis and 35% presenting with distant metastasis [13]. Many patients have no symptoms until they develop obstruction (40%) or bleeding (24%) warranting emergency surgery [13]. SBA commonly presents a solitary lesion with most frequently affected site being duodenum followed by jejunum with ileum being the least common site of involvement [8], [13].

Increased risk of SBA has been seen among smokers, alcoholics, those who consume large amounts of sugar, refined carbohydrates, red meat and smoked food, while a reduced risk was observed with higher intakes of coffee, fish, fruit, and vegetables [8]. According to latest research regular consumption of high lipid diet may also cause such anomalous situation and increase the incidence of Crohn's disease-like inflammatory bowel disorders [14]. Conditions which predispose to SBA include genetic causes like Familial Adenomatous Polyposis, Lynch syndrome and Peutz-Jegher's syndrome and inflammatory conditions like Crohn's disease and Celiac disease [8].

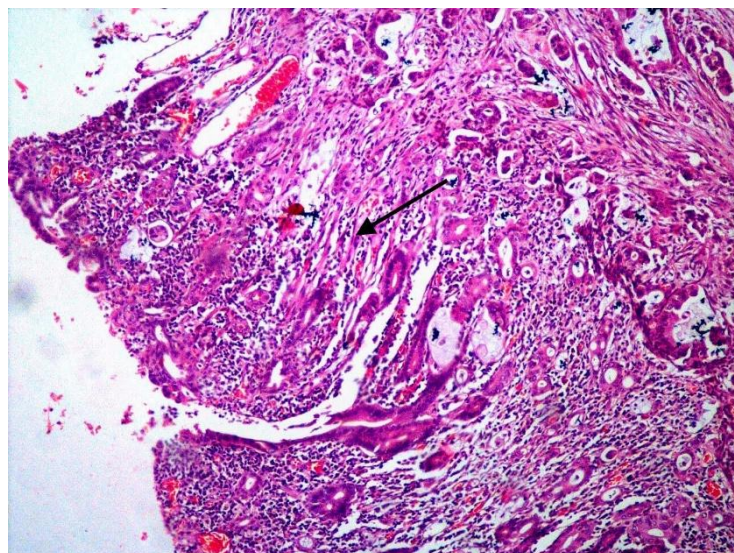


Fig. 3. Haematoxylin and eosin stained sections showing (black arrow) invasive adenocarcinoma (original magnification x20)

Table 1. Timeline

Day	Events
0	Patient presents to the hospital with complaint of colicky central abdominal pain.
7	Patient was fully evaluated and after a period of expectant management patient symptoms improved and was discharged with plan for further evaluation and follow up
22	Patient presented to emergency in a state of acute intestinal obstruction and was operated on the same day – Exploratory laparotomy with resection of multiple strictures in jejunum and ileum with anastomosis of bowel segments
26	Patient developed signs of anastomotic leak and intra-abdominal sepsis and was planned for re-surgery
27	Patient developed multiple organ dysfunction syndrome and his clinical condition deteriorated quickly resulting in emergency surgery being not feasible. Patient expired on post-operative day 5

CT scans have an overall accuracy of only 47% in detecting SBA [8] and newer modalities like Capsule endoscopy and double balloon enteroscopy have a diagnostic yield of only 20-30% and 60-70% respectively [15]. In 57% of patients with distal SBA, none of the preoperative investigations picked up the disease and the diagnosis was established only by laparotomy [13].

Multifocal SBA is unique with only handful of cases reported, with even fewer cases in patients without predisposing factors like long standing Crohn's disease, Celiac disease, Peutz-Jeghers syndrome, Lynch syndrome or Familial adenomatous polyposis syndrome [16], [17], [18]. Despite extensive search we were able to find only two case reports published on misinterpretation of SBA as ITB [16], [19]. Our case is unique on two counts, one is that SBA presented not as a mass but as stricture, which is a rare mode of presentation of SBA. The other unique feature is that malignant strictures were found in multiple locations in both jejunum and ileum which to the best of our search has never been reported before (Table 3). The fact that the most common pathological finding on laparotomy in ITB being multiple small bowel strictures further complicated the differential diagnosis in this patient [9].

New protocols must be framed urgently for early diagnosis of SBA and to distinguish it from ITB and Inflammatory Bowel Disease (IBD). Despite extensive evaluation, no clinical, genetic, radiological or histological evidence of any of above predisposing diseases was found in our patient, making us wonder whether any hitherto unknown factor was involved in the multiplicity of the neoplastic foci.

Surgical resection with adequate clear margins and regional lymphadenectomy remains the treatment of choice in localised disease, with similar surgical principles required even in metastatic SBA presenting with obstruction or bleeding [8]. Standardised chemotherapy regimen against SBA is lacking and is a topic of continuing research. The prognosis of SBA is poor with 5 year overall survival rate being 50-60% for stage I, 39-55% for stage II, 10-40% for stage III, and 3-5% for stage IV [8].

In resource scarce places like the Indian subcontinent where 24 hour frozen section are not available for emergency surgeries and capsule endoscopy or double balloon enteroscopy are done only in a handful of premier institutions, clinical judgement plays a major role in diagnosing diseases and managing the patient, especially in emergency surgeries. Even with all the above mentioned services being available, SBA is not diagnosed commonly in early stages and most patients present in emergency with obstruction or bleeding, like it was in our patient. Surgeons should have high index of suspicion of this rare but sinister diagnosis of SBA in such patients so that timely and appropriate management can be done. Even if the presentation is very highly atypical as it was in our patient having a differential of SBA will help in making better surgical decisions and ultimately results in better patient outcomes. Despite all the latest diagnostic modalities preoperative differentiation of SBA from ITB or from Crohn's disease is not straightforward and further research into this field might help us to diagnose this disease early and to differentiate it with other benign conditions in the future.

Table 2. Worldwide comparison of similar small bowel stricture cases

Study	Number of Patients	Study location	Most common causes of small bowel strictures
Baloch N A et al. 2011 [2]	252	Pakistan	Intestinal tuberculosis (30.6%). Crohn's disease was not found in any of the patients.
Fukumoto A et al. 2007 [10]	179	Japan	Crohn's disease (31.8%) Postoperative or post inflammatory adhesions (17.3%) Tuberculosis was found only in 3.4%
Kroner P T et al. 2016 [11]	71	United States of America	NSAID induced stricture (32%) Crohn's disease (21%) Tuberculosis was not found in any of the patients.
Sonika U et al. 2017 [12]	89	India	Intestinal Tuberculosis (26.9%) Crohn's disease (23.5%)

Table 3. Review of salient findings of articles similar to the current case

Case reports	Age/Sex	Clinical Scenario	Intra-operative Findings	Outcome
Li Q et al. 2020, China [16].	70/Male	Misdiagnosed as Intestinal Tuberculosis (Non-Emergent Presentation).	Multiple infiltrating masses at jejunum with largest 5 cm in diameter.	Fatal outcome
Tang CQY et al. 2019, Singapore [18].	59/Male	Acute intestinal obstruction (Emergency Presentation)	Multiple strictures in terminal ileum.	Follow up data unavailable.
Aggarwal M et al. 2021, India [19].	60/Male	Acute intestinal obstruction (Emergency Presentation)	Solitary 5 cm stricture in distal ileum.	Patient was doing well on follow up.
Present case	54/Male	Acute Bowel obstruction (Emergency Presentation)	Multiple strictures in both jejunum and ileum.	Fatal outcome

4. CONCLUSION

We report this case to highlight how a rare diagnosis of multifocal SBA can easily be overlooked in a patient presenting with acute intestinal obstruction due to multiple strictures. Our patient is further unique in the absence of all known predisposing factors that might explain the multifocal nature of the adenocarcinoma. Pre-operative diagnosis of SBA continues to be challenging and hence attending surgeons should have a high index of clinical suspicion in patients presenting with obstruction, especially in tuberculosis endemic regions like India. There is a critical need for further research to evaluate newer diagnostic and treatment strategies to improve the overall outcome.

CONSENT

All authors declare that written informed consent was obtained from the next of kin of the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

Ethical approval not applicable /not necessary at our institution to publish an anonymous case report.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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SUMMARY OF THE CASE:

1	Patient (gender, age)	54 year old male
2	Final diagnosis	Multifocal primary de novo small bowel adenocarcinoma of jejunum and ileum
3	Symptoms	Central colicky abdominal pain
4	Medications	N/A
5	Clinical procedure	Exploratory Laparotomy with Resection of multiple strictures in jejunum and ileum with anastomosis of bowel segments
6	Specialty	Surgical oncology

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