Hell J Surg. 2023 Oct-Dec;93(2):94–96
doi.: 10.59869/23018

Bowel Perforation and Peritonitis Caused by a Jejunal Leiomyoma.

A rare complication of jejunal leiomyoma

Iraklis Perysinakis[1], Kallirroi Spanou[2], Dimitrios Tamiolakis[3], Penelope Korkolopoulou[2], Georgios Stamatakis[4]

[1]Department of Surgical Oncology, University Hospital of Heraklion, Heraklion, Greece, [2]1st Department of Pathology, National and Kapodistrian University of Athens, Greece, [3]Department of Pathology, University Hospital of Heraklion, Crete, Greece, [4]Department of Surgery, General Hospital of Rethymno, Crete, Greece

ABSTRACT

Small-bowel leiomyomas are rare, usually indolent, benign neoplasms, sporadically associated with complications, mainly including bleeding and obstruction. Herein we report a case of a 78-year-old female patient under high-dose corticosteroid treatment for systemic disease, presenting with clinical signs suggestive of peritonitis. Urgent exploratory laparotomy revealed an intraluminal jejunal mass complicated by bowel perforation and multiple enlarged mesenteric lymph nodes. Microscopic examination of the surgical specimen suggested the presence of a jejunal leiomyoma as the cause of perforation and lymph nodes' infiltration by Hodgkin lymphoma. The patient died on the 15th post-operative day due to pulmonary complications. To our knowledge, this is the first reported case of a jejunal leiomyoma complicated by bowel perforation and peritonitis. A high index of suspicion for the presence of underlying neoplastic disease should be maintained in cases of non-traumatic small-bowel perforation. Additionally, the potentially detrimental effect of comorbidities on a patient's outcome should be considered even in the context of successful management of primary disease.

Key Words: *Small bowel leiomyoma; jejunal leiomyoma; bowel perforation; Hodgkin lymphoma*

CASE REPORT

A 78-year-old Caucasian female patient presented to the Emergency Department complaining of worsening acute abdominal pain over the previous eight hours. Patient's past medical history was significant for dyslipidaemia, type 2 diabetes mellitus and arterial hypertension. Additionally, the patient was on high-dose corticosteroid treatment for the past month due to generalised lymphadenopathy and a recent diagnosis of sarcoidosis, based on lymph node pathological examination.

Upon presentation, the patient was haemodynamically unstable and lethargic. Vital signs were BP=80/40mmHg, 150bpm, 37°C, SpO_2=96%. Abdominal examination revealed generalised rigidity and rebound tenderness to palpation. The clinical picture was compatible with septic shock and sepsis protocol was initiated. Laboratory tests revealed elevated WBCs, increased CRP levels, thrombocytopenia, prolonged INR and APTT and impaired renal function. Abdominal computed tomography scan indicated the presence of an abnormal jejunal loop with an intraluminal calcified lesion and mural thickening surrounded by regional inflammatory alterations, pneumoperitoneum, free intraperitoneal fluid and generalised thoracoabdominal lymphadenopathy (Figure1). Explora-

Corresponding author:
Perysinakis Iraklis, MD PhD FEBS FACS
27, Damaskinou Str., P.C. 71305, Heraklion, Crete, Greece
Tel.: +30 6973 621867, e-mail: iraklisper@gmail.com

Submission: 26.09.2023, Acceptance: 27.11.2023

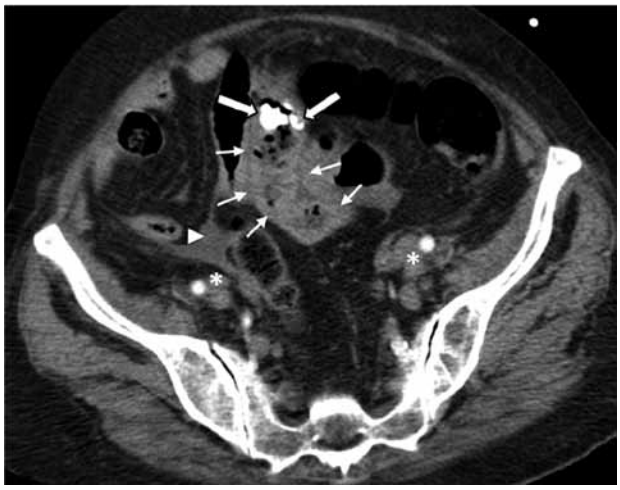

FIGURE 1. Contrast-enhanced axial computed tomography image of the lower abdomen in the arterial phase. Coarse calcifications (thick arrows) are identified along a jejunal loop (thin arrows) with wall thickening and homogenous attenuation, findings that possibly imply the presence of intestinal leiomyoma. Also note the presence of marginally enlarged external iliac lymph nodes (*) and free intra-peritoneal fluid (arrowhead).

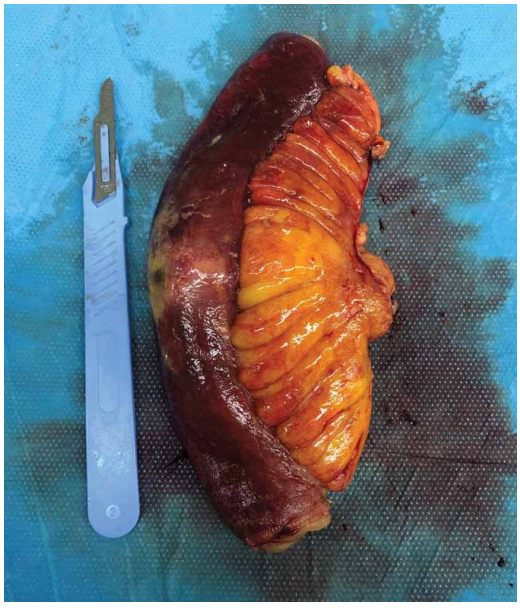

FIGURE 2. Surgical specimen of perforated jejunum. Palpation of the perforated jejunal loop revealed the presence of a firm intraluminal mass.

tory laparotomy was urgently performed confirming the presence of generalised peritonitis due to small-bowel perforation. Palpation of the perforated jejunal loop revealed a firm intraluminal mass. A 12-cm bowel segment was resected with its associated mesentery and the surgical specimen was sent for pathological examination (Figure 2). Bowel continuity was restored with a stapled side-to-side anastomosis and the abdominal cavity was copiously irrigated with warm saline. Temporary abdominal closure with vacuum pack technique was decided based on the patient's clinical status. Postoperatively, the patient was transferred to the intensive care unit (ICU). Re-exploration was performed 48 hours later, confirming the integrity of the anastomosis and significant regression of the peritoneal inflammation, thus permitting definite abdominal wall closure. During hospitalisation, the patient's general condition progressively deteriorated and she finally died on the 15th postoperative day due to pulmonary complications.

Gross pathology revealed the presence of a 2-cm small bowel tumour, occupying the full thickness of the intestinal wall and causing perforation together with six mesenteric six lymph nodes (Figure 3). Microscopic examination of the specimen showed mesenteric lymph nodes' infiltration by mixed cellularity classical Hodgkin lymphoma (Figure 4a). The small bowel tumour was characterised as a benign mesenchymal neoplasm showing positive immunohistochemical staining for desmin and calponin. Stains against CD34, cKIT, DOG1, S100, SOX10 and CKAE1/AE3 were negative and Ki67 expression was low (1-2%). These features were consistent with leiomyoma of the small bowel (Figure 4b).

DISCUSSION

Small-bowel leiomyomas are rare, benign tumours even among the smooth muscle cell tumours of the small intestine. It has been reported that there is one leiomyoma for every 100 GISTs of the small bowel. Clinically relevant leiomyomas occur predominantly in the oesophagus, colon and rectum. On the other hand, small bowel leio-

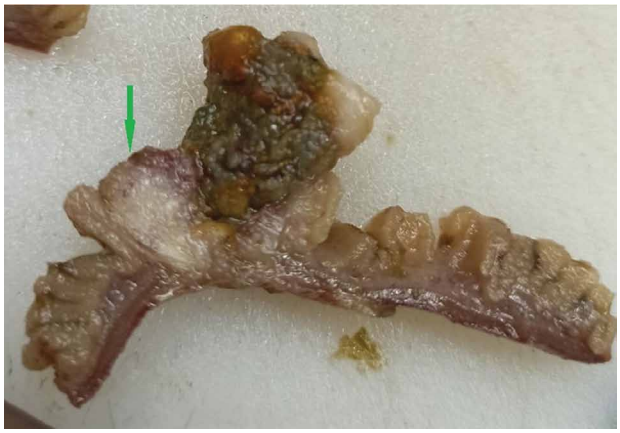

FIGURE 3. Gross examination of perforated tumour-bearing jejunum. A tumour was found occupying the full thickness of the intestinal wall and causing perforation (arrow).

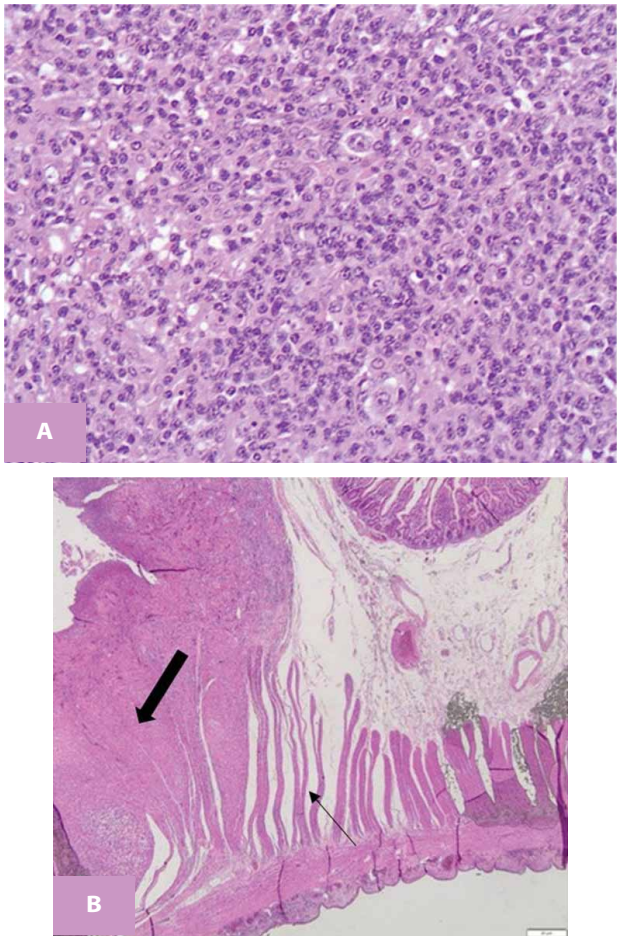

FIGURE 4. A. Microscopic image of an intestinal lymph node involved by mononuclear Hodgkin cells with single round to oval nucleus and prominent eosinophilic nucleolus. B. Microscopic image of a small bowel leiomyoma (thick arrow) arising from muscularis propria (thin arrow).

myomas are typically asymptomatic and usually found incidentally through imaging or during unrelated abdominal procedures. Symptomatic leiomyomas along the small intestine have been sporadically reported [1-4]. In this regard, complications may occur either in case of significant extraluminal growth resulting in central necrosis and bleeding into the bowel lumen, or in case of excessive intraluminal growth associated with bowel obstruction. Given that complicated small-bowel leiomyomas may clinically resemble duodenal ulcer or diverticulosis, leiomyoma of the small intestine has been characterised as one of the "great imitators" [1]. Additionaly, Laibangyang et al. reported a novel case of bowel perforation caused by two giant parasitic leiomyomas, adhered to the bowel, in a patient with previous abdominal myomectomy for fibroids [5]. Despite their admittedly low prevalence and complications rate, to the best of our knowledge, this is the first reported case of primary small bowel leiomyoma resulting in bowel perforation and peritonitis.

This case emphasises that a high index of suspicion for the presence of underlying neoplastic disease should be maintained in cases of non-traumatic small-bowel perforation. Interestingly, the patient presented herein was finally diagnosed with two distinct clinical entities, one benign and one malignant. Although morbidity and mortality would reasonably be expected to be cancer-related, the benign condition was ultimately proven to be fatal. Lastly, despite timely and successful control of the patient's urgent condition, immunosuppression related to the concomitant malignancy and corticosteroid treatment may have posed a significant challenge on recovery. This strongly highlights that, as comorbidities may have a determinant adverse effect on outcome, they should always be thoroughly appraised.

Conflict of Interest: *The authors declare that they have no conflict of interest. The authors have full control of all primary data and they agree to allow the journal to review their data if requested.*

REFERENCES

1. Smith LE. Acute GI bleed due to leiomyoma of the jejunum: A case report. J Surg Case Rep [Internet]. 2017;2017(7):rjx138. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5534017/
2. Sunamak O, Karabicak I, Aydemir I, Aydogan F, Guler E, Cetinkaya S, et al. An intraluminal leiomyoma of the small intestine causing invagination and obstruction: A case report. Mt Sinai J Med. 2006 Dec;73(8):1079-81.
3. Konstantinidis K, Theodoropoulos GE, Spanomihos G, Sambalis G, Vorias M, Georgiou M, et al. Laparoscopic-assisted small bowel resection of a leiomyoma causing recurrent obscure gastrointestinal bleeding. J Laparoendosc Adv Surg Tech A. 2005 Aug;15(4):396-9.
4. Abbas MA, Al-Kandari M, Dashti FM. Laparoscopic-assisted resection of bleeding jejunal leiomyoma. Surg Endosc. 2001 Nov;15(11):1359.
5. Laibangyang A, Law C, Gupta G, Da Dong X, Chuang L. Parasitic leiomyoma causing small bowel perforation: A case report. Case Rep Womens Health [Internet]. 2021 Aug; 32:e00349. Available from: https://www.sciencedirect.com/science/article/pii/S2214911221000679?via%3Dihub