Case Report

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Sclerosing encapsulating peritonitis (Abdominal Cocoon) associated with Non-Hodgkin's lymphoma of bowel wall: a rare case report

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Introduction: Sclerosing encapsulating peritonitis (SEP) is a rare condition causing intestinal obstruction. A thick fibrous membrane envelops abdominal organs. SEP is classified into two types: abdominal cocoon syndrome (ACS), also called idiopathic or primary SEP, and secondary SEP. Symptoms range from subacute to acute intestinal blockage, and diagnosis is usually confirmed during laparotomy.

Case presentation: The authors present a case of a 32-year-old woman who presented with intermittent per-vaginal bleeding for 1 month, abdominal distension, decreased appetite, and significant weight loss over 5 months. Examination revealed pallor and a palpable 12×12 cm mass in the right lower abdomen. Imaging revealed a lobulated mass in the mid-ileum with bowel wall thickening. She underwent laparotomy, revealing fibrous tissue, and a nodular mass on the distal ileum. Histopathology confirmed non-Hodgkin's lymphoma.

Discussion: Sclerosing encapsulating peritonitis is a rare chronic inflammatory condition affecting the peritoneum. Primary SEP, also known as abdominal cocoon or idiopathic SEP, lacks a definitive cause, while secondary SEP results from conditions like peritoneal dialysis, liver cirrhosis, and malignancies. It involves peritoneal irritation leading to inflammation, fibrin-like material release, and subsequent bowel encasement and fibrosis. Diagnosis is typically made during laparotomy, though contrast-enhanced CT scan aids in presumptive diagnosis.

Conclusion: This case report highlights a rare occurrence of abdominal cocoon syndrome in a patient with non-Hodgkin B-cell lymphoma, presenting with abdominal pain, distension, and bowel obstruction. Diagnosing this condition early is crucial, especially in patients with underlying malignancies. More research is needed to improve management.

Keywords: case report, cocoon, intestinal obstruction, non-Hodgkin's lymphoma, peritonitis

Introduction

Sclerosing encapsulating peritonitis (SEP) is a rare cause of intestinal obstruction characterized by a thick, fibrous, greyish-white cocoon-like membrane covering the small intestine and occasionally other intra-abdominal organs such as the liver, stomach, and colon[1,2]. SEP is categorized into two types: abdominal cocoon syndrome (ACS), also known as idiopathic or primary sclerosing encapsulating peritonitis, and secondary sclerosing encapsulating peritonitis. GI tract malignancy is one of the causes of secondary SEP[3]. The symptoms of ACS vary from recurrent subacute intestinal obstruction to acute intestinal obstruction[3,4]. A CT scan with IV contrast of the abdomen may suggest the diagnosis, but the final diagnosis is generally made after the surgery[4].

HIGHLIGHTS

- Sclerosing encapsulating peritonitis (SEP) is an uncommon chronic inflammatory disorder that affects the peritoneum. It is categorized into primary SEP, also known as abdominal cocoon or idiopathic SEP, which lacks a clearly identified cause. Secondary SEP, on the other hand, is associated with conditions such as peritoneal dialysis, liver cirrhosis, and various malignancies.
- It is challenging to diagnose before surgery because its early clinical signs are nonspecific. Therefore, it is usually identified during laparotomy.
- Patients with mild symptoms or who are asymptomatic are managed conservatively with enteral and parenteral nutrition support. Those with moderate to severe symptoms may require both medical treatment, including corticosteroids and tamoxifen, and surgical intervention, such as bowel exploration and excision of the encapsulating sac.

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We present a case of a 32-year-old female diagnosed with abdominal cocoon syndrome caused by primary intestinal Non-Hodgkin's Lymphoma. This report highlights the diverse clinical features and diagnostic challenges in resource-limited settings. The key learning objectives include understanding the clinical presentation, diagnostic challenges, and limitations of gastrointestinal lymphomas in such environments.

Case presentation

Clinical history

A 32-year-old female with no comorbidities came to the tertiary care center with a history of intermittent per-vaginal bleeding for 1 month and abdominal distension for 5 months. Furthermore, she also had a history of decreased appetite and significant weight loss for the same duration. Moreover, she experienced an evening rise in temperature for the past week. There is no history of neck swelling or masses in the axillary or inguinal regions. Her obstetrics history includes G2P2L2 (Age 14 years and 6 years). Her bladder habits were normal. She had no history of nausea, vomiting, burning sensation of micturition, melena, diarrhea, cough, shortness of breath, and headache. She had no family history of malignancy or other tumors.

Physical examination

Her general condition was conscious, oriented to time, place, and person, coherent, and pale. She had stable vital signs except for a temperature of 100.4°F. On general physical examination, she had a pale conjunctiva and features of mild dehydration. However, lymph node examination showed no neck swelling, axillary, or inguinal masses. Abdominal palpation of the liver and spleen was normal without evidence of hepatosplenomegaly. Moreover, an abdominal examination revealed a 12×12 cm firm, slightly mobile lump palpable on the lower abdomen. Furthermore, Per-rectal examination revealed palpable mass anteriorly, and rectal mucosa was free. The neurological, respiratory, and cardiovascular examination was completely normal.

Laboratory examination

The results of serum laboratory investigations, including complete blood counts (CBC), CA-125, CA19-9, and CEA, were all within the reference range except the hemoglobin level, which was 5.1 gm/dl (Normal: 12–16 gm/dl in females), and packed cell volume (PCV), which was 18% (normal: 36–46% in females). The tuberculin test was also negative.

Imaging examination

With all the history and examination in mind, an ultrasound of the abdomen and pelvis was done, which revealed bowel wall thickening measuring 21 cm in the right adnexal region extending to the left iliac fossa. There were a few enlarged lymph nodes in the periumbilical and left iliac fossa region, ~ 16×13 cm.

For further evaluation, a contrast-enhanced CT scan (CECT) of the chest, abdomen, and pelvis revealed a lobulated mass in the mid ileum with asymmetric exophytic bowel wall thickening measuring 10 cm in length and 4 cm in thickness. No apparent intraluminal narrowing was noted. Mass adhered to adjacent bowel loops and the abutting dome of the urinary bladder. Possible small bowel gastrointestinal stromal tumor (GIST). Heterogeneously enhancing nodular lesion was noted in the parauterine region, the largest one measuring 3 cm×2 cm. A few subcentimeter para-aortic lymph nodes were also seen. The findings mentioned above are shown in Figure 1. However, the chest CECT scan did not show evidence of mediastinal lymphadenopathy, as shown in Figure 2.

Treatment

Initially, the patient was admitted to the gynecology unit, but after CECT of the abdomen and pelvis, she was transferred to the surgery unit. After the collective discussion, her treatment was started by correction of anemia with a transfusion of four pints of pack red blood cells and exploratory laparotomy with En-Bloc resection of the ileal mass with side-to-side anastomosis. The firm mass measuring 12×12 cm from the ileum about 50 cm from the ileocaecal junction was resected, as shown in Figure 3. The mass was adherent to the rectosigmoid junction and dome of the urinary bladder along with the left adnexa, as shown in Figure 3. Therefore, bladder repair with left salpingo-oophorectomy was

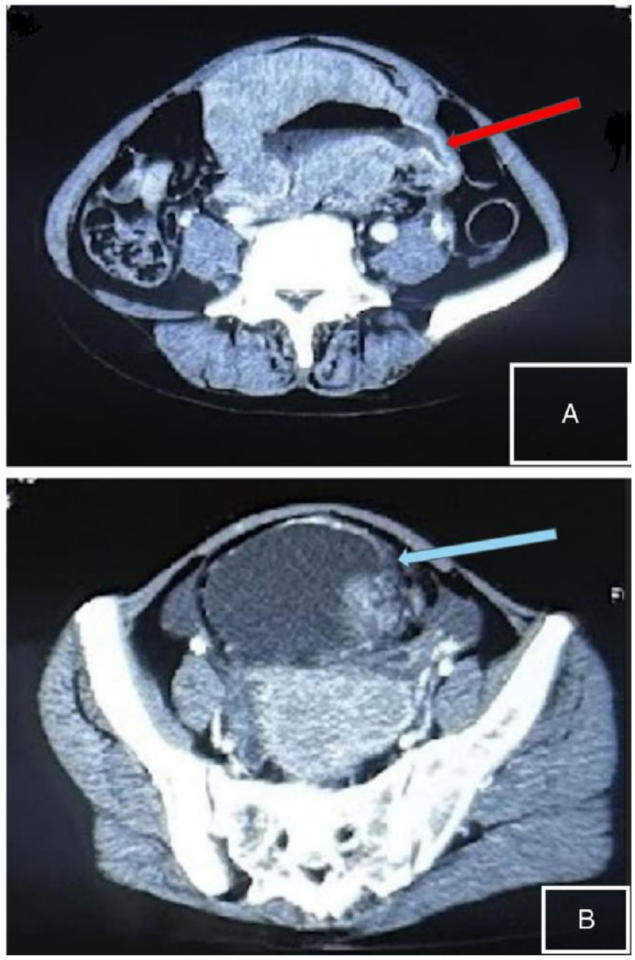

Figure 1. Contrast CT scan of abdomen and pelvis showing (A, red arrow) lobulated mass in the mid ileum with asymmetric exophytic bowel wall thickening. (B, blue arrow) mass adherent to adjacent bowel loops and abutting dome of the urinary bladder.

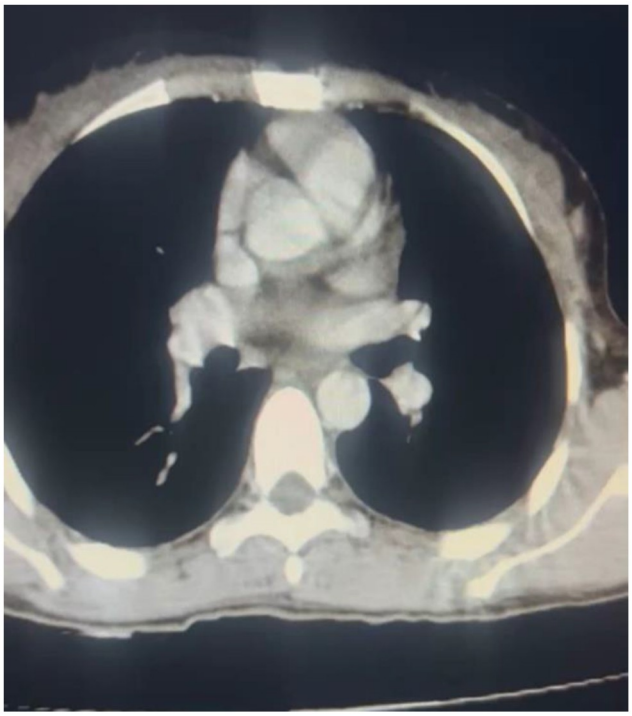

Figure 2. Normal CECT scan of the chest showing no evidence of mediastinal lymphadenopathy.

performed simultaneously. Moreover, enlarged lymph nodes of about 3 cm were noted in the left pelvic region.

Histopathological analysis

After history, examination, radiographic evidence, and intraoperative findings, a specimen of pelvic mass and left adnexa was sent for histopathological analysis.

The specimen of pelvic mass comprised of the bowel and a nodular mass measuring 16 × 7 cm. The bowel margins on either side of the nodular mass measured 6 cm and 5 cm in length. Another lumen was also identified, measuring 3 cm in length. The external surface of the nodule appeared congested. The serial cut surface showed irregular greyish-white to brown tissue extending to the attached part of the urinary bladder and bowel. Altogether, eight lymph nodes were identified around the lesion; the largest measured 1.5 cm in diameter.

The left adnexa specimen consisted of irregular greyish-brown tissue measuring 6×4×3 cm. The external surface appeared irregular and congested. The cut surface showed greyish-white soft to myxoid tissue. The attached tubular tissue measured 5 × 2 cm; the cut surface showed greyish-white tissue.

The histopathological analysis findings include:

1. Multiple sections from the huge nodular mass showed complete effacement of the lymphoid architecture with atypical lymphoid cells arranged in a diffuse pattern. The atypical cells were medium in size, having a moderate amount of basophilic cytoplasm, and were round to oval nuclei with hyperchromatic to vesicular chromatin. Some of the nuclei showed prominent nucleoli. Starry sky patterns with apoptotic bodies, increased mitotic figures, necrosis, and crushing artifacts were also seen, as shown in Figure 4. These tumor cells were seen extending throughout the bowel's thickness and up to the serosal surface.
2. Perineural and lymphovascular invasions were also noted.
3. Sections from the attached part of the urinary bladder showedfeatures of infiltration by tumor cells, as described above.
4. Eight lymph nodes were identified from around the lesion and the attached mesentery, all of which showed similar histopathology as described above.
5. Sections from the bladder and bowel resected margins were, however, free from the tumor.
6. Sections from the left ovarian stroma showed infiltration of the ovarian stroma by tumor cells as described above.
7. Sections from the attached left fallopian tube show normaltubal epithelium; however, the peritubal fibro-collagenous tissue showed infiltration by tumor cells.

Therefore, histomorphological features were suggestive of non-Hodgkin's lymphoma. The differential diagnosis based on histopathological analysis includes non-Hodgkin's Lymphoma and gastrointestinal stromal tumor (GIST). An immunohistochemistry study was recommended for precise diagnosis, categorization, and evaluation. The immunohistochemistry study revealed the tissue with sheets of medium-sized atypical lymphoid cells with fine chromatin, inconspicuous nucleolus, and scant cytoplasm. Karyorrhectic debris was also noted. The tumor cells are positive for CD20 (as shown in Fig. 5), CD10, BCL6, and negative for CK, CD3, TdT, CD30, CD138, BCL2, CD34, MUM1, and SOX11. The Ki67 was ~ 95%, as shown in Figure 5. Furthermore, c-MYC revealed very focal areas with weak staining at the periphery, and the exact percentage could not be commented on due to the poorly preserved nature of the tissue.

The histopathological analysis confirmed the diagnosis as high-grade B cell non-Hodgkin's lymphoma. BCL2, BCL6, and cMYC gene arrangement studies were recommended to confirm/exclude the possibility of Burkitt Lymphoma. Gene arrangement studies could not be performed due to limitations in laboratory resources and technical expertise in our setting. The differential diagnosis includes subtypes of non-Hodgkin's lymphoma such as diffuse large B cell lymphoma (DLBCL), germinal center or nongerminal center lymphoma, Burkitt Lymphoma, etc.

Outcome and follow-up

The patient's postoperative course was uneventful. No complications following surgery were noted. She was discharged on the seventh day after completing the course of antibiotics (ceftriaxone and metronidazole). Her hemoglobin level at the time of discharge was 10 gm/dl. The patient was referred to the oncology department for additional treatment for non-Hodgkin's lymphoma. The patient came for a follow-up after 1 week for suture removal. A follow-up visit after a month of discharge showed a well-healed laparotomy scar and was asymptomatic.

Discussion

Sclerosing encapsulating peritonitis (SEP) is a rare, chronic inflammatory condition affecting the peritoneum with an unknown definitive cause[5]. SEP is categorized into two types: abdominal cocoon syndrome (ACS), also known as idiopathic or

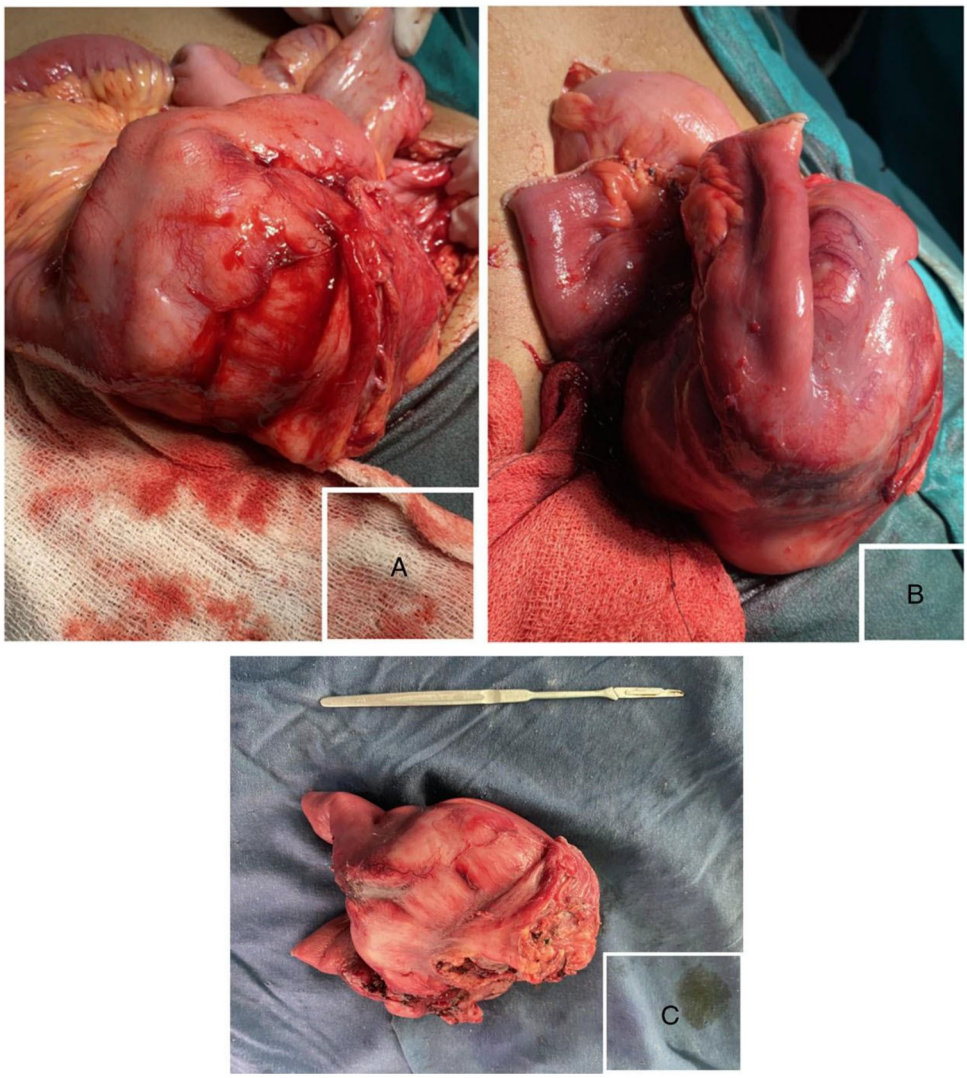

Figure 3. (A, B, C): Resected mass measuring 12×12 cm arising from the ileum about 50 cm from the ileocaecal junction and adherent to the rectosigmoid junction and dome of the urinary bladder along with the left adnexa forming an abdominal cocoon.

primary sclerosing encapsulating peritonitis, and secondary sclerosing encapsulating peritonitis[3]. Cleland described a disease as peritoneal encapsulation in 1968, whereas Foo first introduced the term cocoon syndrome in 1978[6,7]. Idiopathic or primary SEP manifests as an inflammatory condition characterized by recurring low-grade or subclinical peritonitis, initially presenting without specific abdominal symptoms and later progressing to sclerosis, thickened membrane, and cocoon formation with symptoms of acute, subacute, or chronic intestinal obstruction. It can be classified into three categories according to the extent of organ encapsulation: type I, where the membrane encases only a portion of the intestine; type II, where the entire intestine is encased; and type III, where the membrane en cases the intestine as the whole along with additional organs such as the appendix, cecum, ascending colon, and ovaries[2].

Primary SEP, idiopathic SEP, or abdominal cocoon generally occurs in young adolescent girls in tropical or subtropical areas. Though the exact pathogenesis is unknown, it is believed to be related to retrograde menstruation or viral gynecological infection via fallopian tubes. This results in peritoneal irritation, leading to inflammation and release of intraperitoneal fibrin-like material by fibrogenic cytokines, forming thick membranes encasing bowel contents and subsequent fibrosis[7]. Secondary SEP, however, has a definite etiology, such as peritoneal dialysis, liver cirrhosis, GI tract malignancy, pelvic inflammatory disease, tuberculosis, sarcoidosis, use of beta blockers, and practolol therapy[3]. The pathophysiology behind the formation of abdominal cocoon in lymphoma is said to be due to tumor invasion and subsequent perforation of the bowel wall, which can trigger an inflammatory response leading to fibrous tissue formation. This chronic inflammation, either caused by the tumor itself or as a result of chemotherapy, can progressively result in the development of fibrosis. Over time, this fibrosis can lead to the thickening of the peritoneal membrane and the formation of the abdominal cocoon. It is also hypothesized that the immune response against the lymphoma may play a crucial role in cocoon formation. The immune system's effort to combat the tumor can lead to the deposition of fibrin and other proteins within the peritoneal cavity that exacerbates fibrosis and subsequent encapsulating of the intestines and formation of abdominal cocoon[8,9].

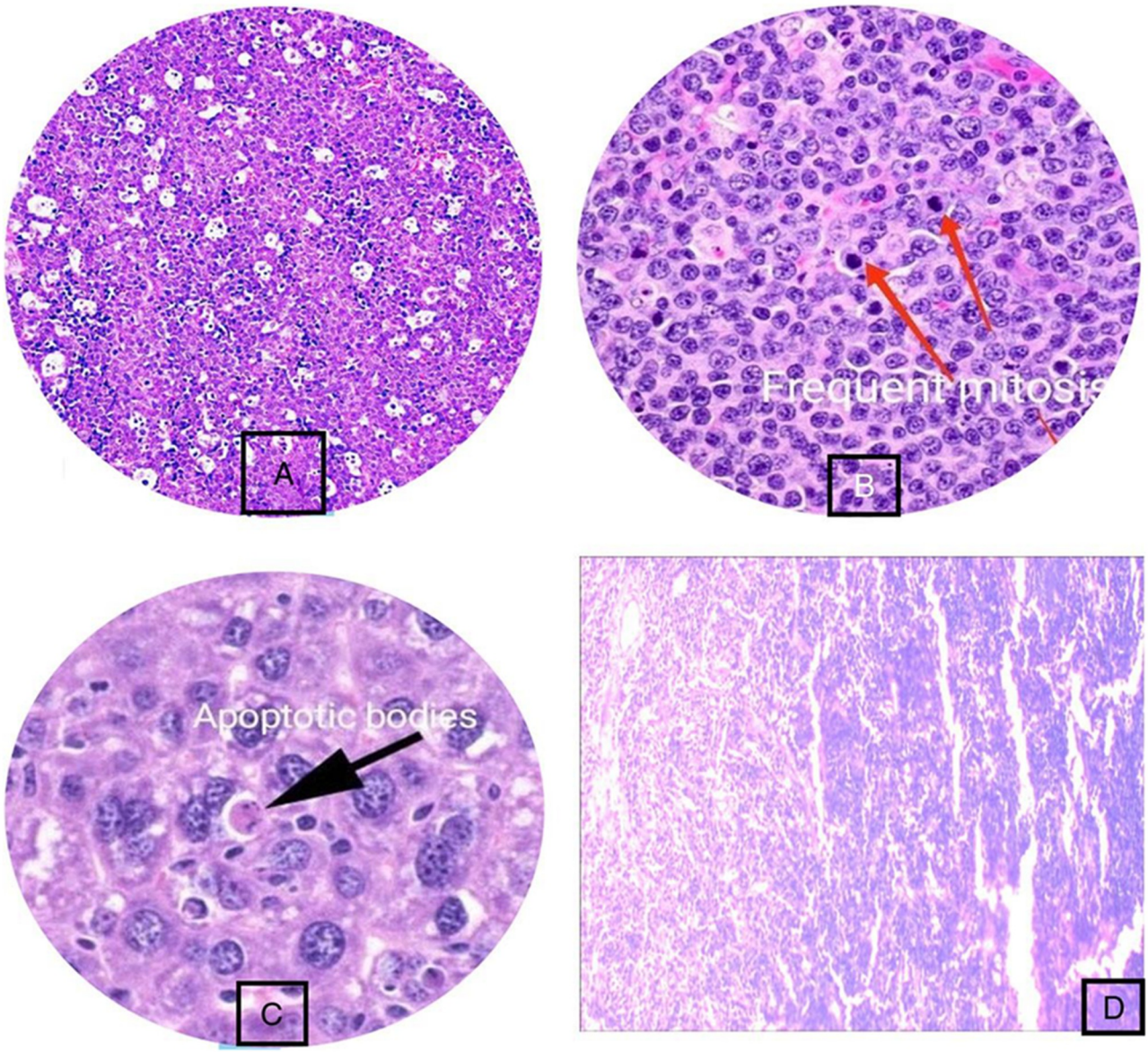

Figure 4. Histopathological analysis of resected pelvicmass and left adnexa. (A, ○) Starry sky pattern (Hematoxylin and Eosin, × 40). (B, ○) Clumped chromatin and prominent nucleoli with frequent apoptosis (Hematoxylin and Eosin,×100). (C, ○) Apoptotic Bodies (Hematoxylin and Eosin,×400). (D, □) Tumor cells extend throughout the bowel's thickness and up to the serosal surface (Hematoxylin and Eosin).

Though GI tract malignancy is a recognized cause of secondary encapsulating peritonitis[3], non-Hodgkin's lymphoma has not yet been identified in the literature as a cause of abdominal cocoon syndrome. The gastrointestinal tract is the most common site for primary extranodal non-Hodgkin's Lymphoma and represents 20–40% of all cases of non-Hodgkin's lymphoma. Nearly half of primary GI lymphomas occur in the stomach, followed by the small intestine and large intestine, accounting for 14–38% and 10–20% of the cases, respectively. Terminal ileum and ileocecal region were most commonly affected with extranodal primary Non-Hodgkin's lymphoma, probably due to a higher proportion of lymphoid tissue in these areas. Histologically, primary extranodal lymphoma of the gastrointestinal tract is of B cell origin and accounts for 80–90% of cases[10,11].Lymphoma accounts for 15–20% of all small intestine neoplasms[12]. The most common clinical symptoms of gastrointestinal non-Hodgkin's lymphoma are abdominal pain, nausea, vomiting, diarrhea, and malabsorption[11].

In our case, the patient's presentation supports a diagnosis of primary gastrointestinal (GI) lymphoma rather than secondary lymphoma with GI tract involvement. The patient experienced abdominal pain, the most common symptom of GI lymphoma, with no or late-appearing systemic B symptoms like fever and night sweats, which are less frequent in primary cases[12,13]. Additionally, the patient did not exhibit lymphadenopathy in the axillary, inguinal, or cervical regions, and the lymphadenopathy was localized to the bowel, a feature of primary gastrointestinal lymphoma. Peripheral blood counts were normal, and

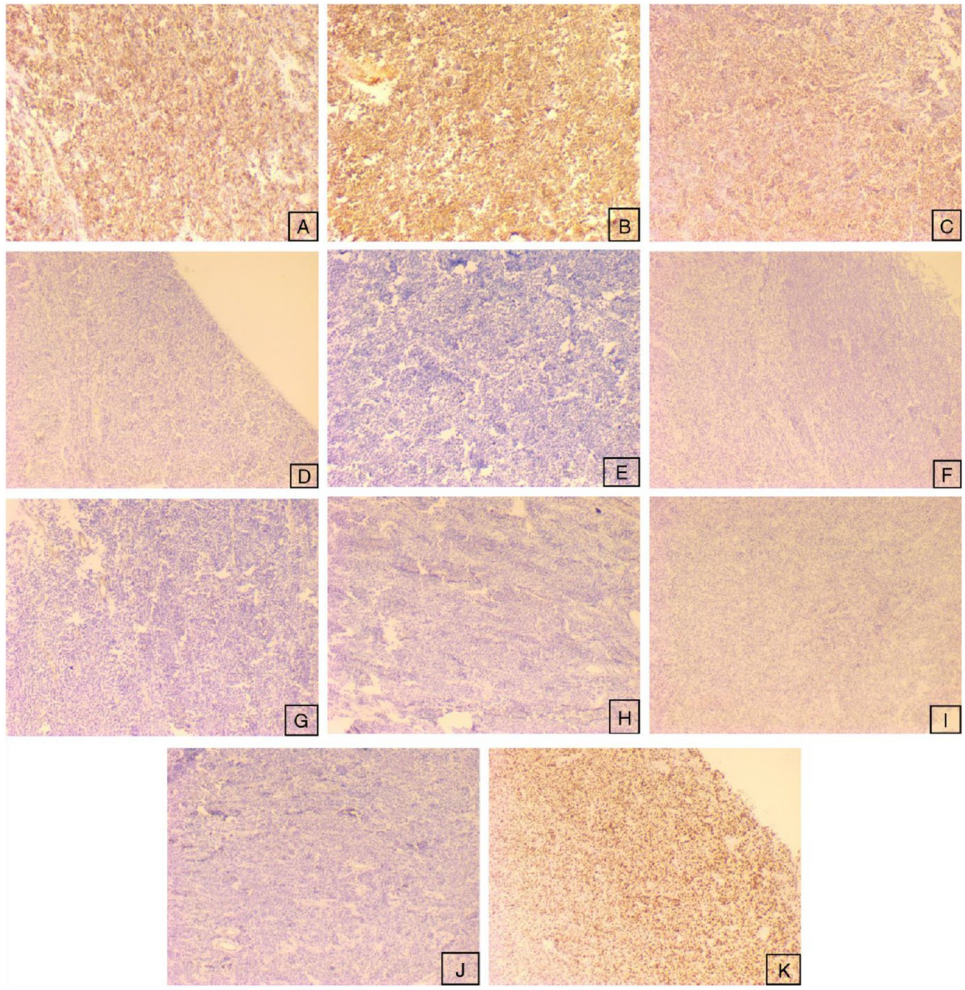

Figure 5. Immunohistochemistry study. (A) Tumor cells with CD20 positive, (B) tumor cells with CD10 positive, (C) tumor cells with BCL6 positive, (D) tumor cells with BCL2 negative, (E) tumor cells with CD3 negative, (F) tumor cells with CD30 negative, (G) tumor cells with CD138 negative, (H) tumor cells with CK negative, (I) tumor cells with CMYC with a very focal areas showing weak staining at the periphery, (J) tumor cells with TdT negative, and (K) tumor cells with ki67 ~ 95%.

there was no evidence of liver and spleen involvement with normal liver and spleen dimensions and echotexture in the CECT abdomen without evidence of hepatosplenomegaly on physical examination. These findings confirm that the lymphoma is primary to the GI tract[14]. Additionally, the symptom of abnormal vaginal bleeding in our case is consistent with the malignant infiltration of tumor cells into the ovary. Typically, patients with ovarian lymphoma present with symptoms such as abdominal or pelvic mass, weight loss, vaginal bleeding, ascites, and weakness[15]. The CECT abdomen revealed heterogeneously enhancing parauterine masse, and histopathological analysis revealed tumor infiltration in the left ovary and fallopian tube. This finding likely accounts for the vaginal bleeding observed in our patient, as the tumor's involvement in these structures can disrupt normal reproductive function and lead to such symptoms.

SEP (abdominal cocoon syndrome) is rarely suspected preoperatively due to a lack of specificity in early clinical manifestations, so the diagnosis is typically made during laparotomy[16].

The features of abdominal pain and vomiting without cardinal symptoms of intestinal obstruction and the presence of a nontender soft mass on abdominal palpation in relatively young females without any apparent cause with a history of similar episodes that resolved spontaneously aid in early preoperative diagnosis of abdominal cocoon syndrome[17]. Besides clinical symptoms, diffuse air-fluid levels and dilated small intestine loops may be visible in abdominal X-rays. Small bowel barium studies show clustered bowel loops in the central abdomen, often called 'cauliflower sign'. Abdominal ultrasonography may reveal dilated bowel segments enclosed within a dense fibrous membrane, free abdominal fluid, and peritoneal thickening. A peritoneal thickening greater than 2 mm is considered significant. Computed tomography with IV contrast (CECT abdomen) is considered the most effective diagnostic tool and typically shows conglomerated small intestine segments at the midline surrounded by a thick capsule with a contrast-free periphery. Histologic diagnosis provides a definitive identification that reveals fibroconnective tissue growth, inflammatory infiltrates, and dilated lymphatics within the peritoneum[2].

The differential diagnosis of SEP includes internal hernia, congenital peritoneal encapsulation, peritonitis carcinomatosa, pseudomyxoma peritonei, and tuberculous peritonitis. Asymptomatic or patients with mild symptoms are treated

conservatively with enteral and parenteral nutrition support. Patients with moderate to severe cases are treated both medically and surgically. Medical management includes corticosteroids and tamoxifen therapy, whereas surgically, patients are managed with bowel exploration and excision of the sac. Surgical excision using a laparoscopic approach is considered optimal, involving the removal of the encapsulating membrane and adhesiolysis, which typically leads to favorable outcomes and an excellent prognosis[7]. The treatment strategy for gastrointestinal lymphoma is influenced by several factors, including the patient's age, clinical presentation, histological subtype, extent and burden of disease, and any comorbidities. Management options include surgery, chemotherapy, and radioimmunotherapy, which may be used in multiple combinations. Surgical resection alone may be sufficient for low-grade B-cell lymphoma of the small intestine. For advanced gastrointestinal lymphoma, surgery, along with a combination of chemotherapy and radioimmunotherapy, is needed. For tumors that cannot be removed surgically, systemic treatment with anthracycline-based chemotherapy is recommended, often followed by radiotherapy[12,18]. Overall, patients with intestinal B-cell Lymphoma who do not undergo surgery tend to have better responses to chemotherapy compared to those with the intestinal T-cell subtype[19].

The prognosis of secondary SEP depends upon the identified cause. For patients undergoing peritoneal dialysis for end-stage renal disease with cocoon formation, the outcome is generally poor, with high mortality rates as high as 69%. With the developments of surgical techniques, recent research suggests a good outcome following surgical management, minimal perioperative mortality, and recurrent obstruction in less than 10% of patients after surgical management[2].

This case report describes a rare occurrence of abdominal cocoon syndrome with non-Hodgkin B-cell lymphoma, presenting a diagnostic challenge due to overlapping symptoms like abdominal pain, distension, bowel obstruction, and lack of specific imaging findingscausing diagnostic dilemma and therapeutic challenge due to the complex interplay between tumor mass and bowel obstruction, tumor infiltration to other organs, risk of dissemination during surgery, and less efficacious to chemoradiotherapy if the tumor has metastasized. This report highlights the successful identification of tumor infiltration through imaging, histopathology, and immunohistochemistry, which contribute to the broader understanding of effective diagnostic strategies in resource constraints.

The limitations of this study include the inability to perform a PET-CT scan to identify metastatic foci and the lack of gene arrangement analysis to further differentiate the tumor subtype, both due to resource constraints. Additionally, the study is limited by the absence of long-term follow-up data, which could provide insights into the long-term efficacy of patient outcomes. The rarity of abdominal cocoon syndrome caused by primary intestinal lymphoma posed challenges in identifying comparable cases and establishing treatment protocols. Ensuring an accurate diagnosis and appropriate management required careful interpretation of available imaging, immunohistopathology, and clinical data in a low-resource setting.

Conclusion

This case report describes a rare occurrence of abdominal cocoon syndrome with non-Hodgkin B-cell lymphoma, presenting a diagnostic and therapeutic challenge. Early and accurate diagnosis is crucial. Clinicians should consider abdominal cocoon in patients with unexplained abdominal symptoms, abnormal vaginal bleeding episodes, and weight loss, especially when there is an underlying malignancy. More research and case reports are needed to better understand and manage this condition.

Methods

This case report was prepared following the SCARE (Surgical Case report Checklist) guidelines[20] to ensure comprehensive reporting.

Ethical approval

Ethical approval is not required for case report in this institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Not applicable.

Author contribution

K.K., A.P., R.B.D.: conceptualization, data curation, writing – original draft, and writing – review and editing; P.L.S.: writing – original draft and writing – review and editing; R.J.: writing – original draft; S.P.: writing – review and editing; P.S.: data curation; S.R.S.: writing – review and editing. All authors approved the final manuscript as submitted.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

Research registration unique identifying number (UIN)

1. Name of the registry: not applicable.
2. Unique identifying number or registration ID: not applicable.
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): not applicable.

Guarantor

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Data availability statement

All the relevant data have been included in the manuscript itself.

Provenance and peer review

Yes.

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