Mesenteric Hyaline Vascular Castleman Disease Presenting as Acute Mechanical Intestinal Obstruction

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To the Editor

Castleman disease (CD), also known as angiofollicular lymph node hyperplasia, is a rare lymphoproliferative disorder first described in 1956 by Castleman et al. [1]. Histopathologically, three principal variants are recognized: hyaline vascular, plasma cell, and mixed types. Clinically and radiologically, CD may mimic malignant lesions, rendering preoperative diagnosis particularly challenging. The hyaline vascular subtype is typically asymptomatic and often detected incidentally, although symptoms related to mass effect may occur in advanced disease. Acute mechanical intestinal obstruction as the presenting manifestation of mesenteric CD is exceedingly rare; only a limited number of such cases have been documented in the literature [2,3,4]. Herein, we report a case of unicentric mesenteric CD presenting as acute mechanical small bowel obstruction, and discuss its diagnostic and surgical management.

Patient Report

A 61-year-old male with no significant past medical or surgical history presented to the emergency department with right lower quadrant abdominal pain, abdominal distension, and inability to pass stool for four days. Laboratory evaluation revealed mild leukocytosis (white blood cell count 11.2 × 10^3/μL) and elevated C-reactive protein (CRP: 48 mg/L); tumor markers including carcinoembryonic antigen (CEA) and cancer antigen 19-9 (CA 19-9) were within normal limits. Abdominal computed tomography (CT) demonstrated dilatation of the small bowel loops with multiple air–fluid levels consistent with acute mechanical intestinal obstruction, and a well-defined, homogeneously enhancing soft tissue density mass without calcification adjacent to the terminal ileum causing near-complete luminal obstruction (Fig. 1). The differential diagnosis included lymphoma, gastrointestinal stromal tumor (GIST), carcinoid tumor, and mesenteric tuberculosis.

Emergency surgery was performed. Intraoperative exploration revealed multiple enlarged lymph nodes within the mesentery of both the colon and small intestine, and a mass measuring approximately 3.5 × 4 cm adjacent to the cecum causing luminal obstruction. An extended right hemicolectomy with ileotransverse anastomosis was performed.

Macroscopic examination of the resection specimen revealed an ileum segment measuring 54 cm and a lymph node specimen measuring 4.5 × 3.5 × 1.6 cm showing a dark gray, partially solid nodular structure on sectioning. Microscopic examination at low magnification (×40) demonstrated preserved nodal architecture with regressed and atrophic germinal centers surrounded by expanded concentric mantle zones, imparting the characteristic "onion-skin" appearance. At higher magnification (×200), prominent vascular proliferation with hyalinized vessels penetrating the germinal centers was evident, producing the classic "lollipop" follicle morphology. These features were consistent with the hyaline vascular type of Castleman disease across fourteen mesenteric lymph nodes. Immunohistochemical (IHC) staining was performed to exclude lymphoma. Findings included CD20 positivity in lymphoid follicles, CD3 positivity in paracortical areas, CD23 highlighting the follicular dendritic cell network, increased Ki-67 proliferative activity confined to germinal centers, B-cell lymphoma-2 (BCL-2) negativity in germinal centers (excluding follicular lymphoma), Cyclin D1 negativity (excluding mantle cell lymphoma), absence of a monoclonal kappa/lambda immunoglobulin pattern, and human herpesvirus 8 (HHV-8) negativity (excluding HHV-8-associated multicentric Castleman disease). No evidence of neoplasia was identified (Fig. 2).

The postoperative course was uneventful and the patient was discharged on postoperative day 8. Follow-up evaluations at 1, 3, 6, and 12 months revealed no recurrence or additional pathology.

Discussion

Although Castleman disease has occasionally been reported as a cause of intestinal obstruction [2,3,4], this presenta-

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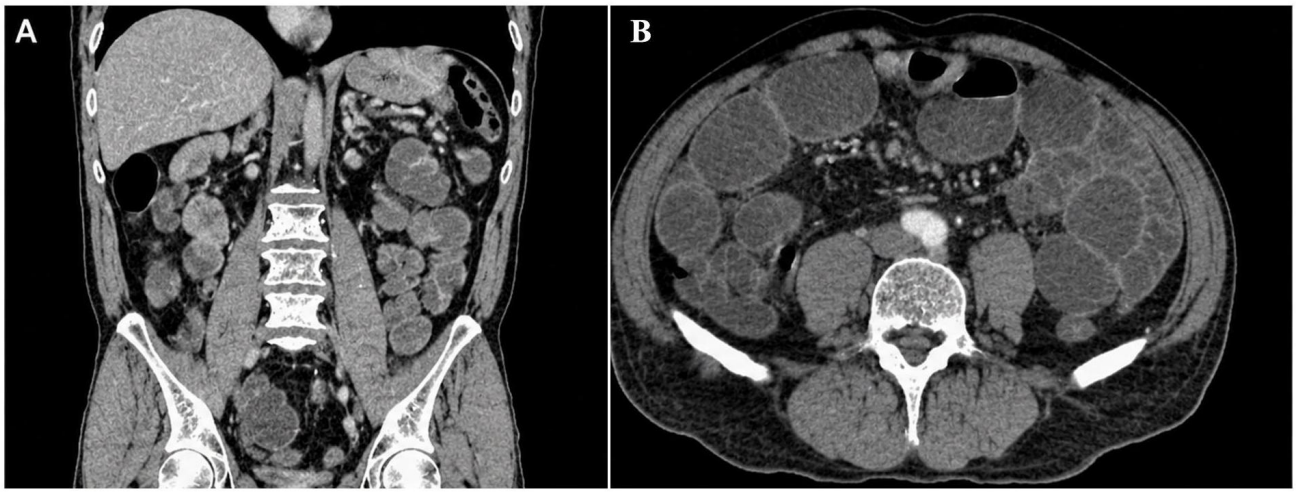

Fig. 1. Abdominal computed tomography (CT) findings demonstrating acute mechanical intestinal obstruction caused by a mesenteric mass. (A) Coronal contrast-enhanced abdominal CT image demonstrating marked dilatation of small bowel loops with multiple air–fluid levels. A well-defined soft tissue density mass is observed adjacent to the terminal ileum, causing near-complete luminal obstruction. (B) Axial CT image showing dilated small bowel loops consistent with mechanical intestinal obstruction. A mesenteric mass lesion compressing the intestinal lumen is also visualized.

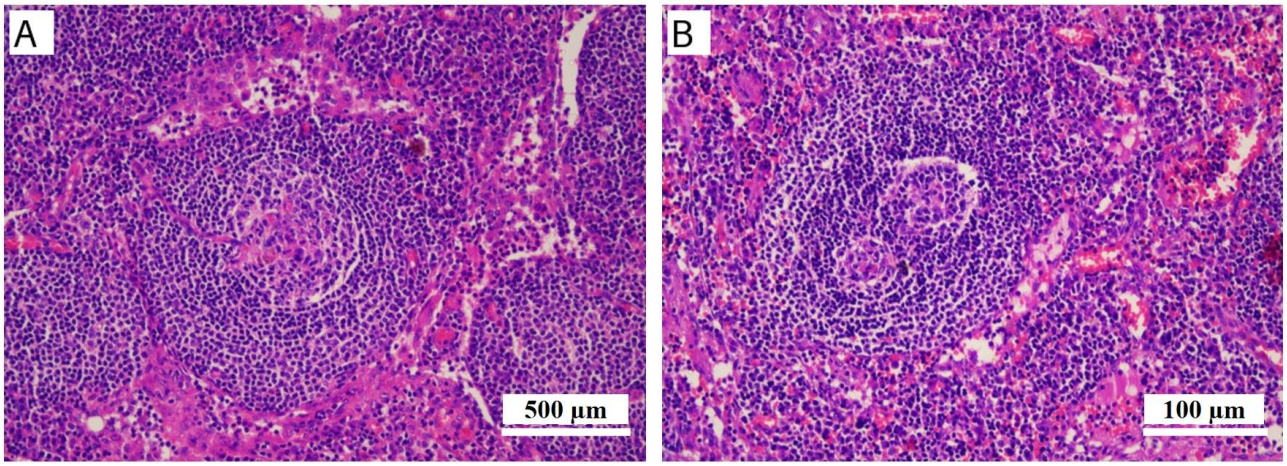

Fig. 2. Histopathological features of the excised mesenteric lymph node consistent with hyaline vascular Castleman disease (hematoxylin and eosin staining). (A) Histopathological appearance of the excised lymph node demonstrating features consistent with the hyaline vascular type of Castleman disease, including regressed germinal centers and prominent vascular proliferation (hematoxylin and eosin staining). Lymphoid follicles showing an expanded mantle zone arranged in a concentric 'onion-skin' pattern surrounding an atrophic germinal center. A hyalinized blood vessel penetrates the follicle, creating the classic 'lollipop' appearance. Scale bar: 500 µm. (B) Higher magnification histopathological image demonstrating characteristic regressed germinal centers with concentric mantle zones ("onion-skin" appearance) and increased vascularity typical of the hyaline vascular variant of Castleman disease. This image displays characteristic germinal center twinning within one shared mantle zone. Scale bar: 100 µm.

tion remains exceptionally uncommon. Kartal et al. [2] reported a case of mesenteric CD causing mechanical obstruction managed with surgical resection, emphasizing the diagnostic challenge in the emergency setting. Similarly, El Demellawy et al. [4] described localized mesenteric CD presenting as recurrent intestinal obstruction, further highlighting the mass-effect potential of this disease. Most reported abdominal cases are either asymptomatic or present with nonspecific abdominal discomfort related to mass effect [5,6]. The present case is notable in that the obstruction was caused by unicentric hyaline vascular CD involving fourteen mesenteric lymph nodes, confirming that even the typically indolent hyaline vascular subtype can produce acute surgical presentations requiring emergency intervention.

Preoperative diagnosis of mesenteric CD remains challenging. Radiological findings are generally nonspecific and may resemble malignant tumors or lymphoproliferative dis-

orders [5]. In our case, the imaging features — a homogeneously enhancing, well-circumscribed mass without calcification — were nonspecific and raised the suspicion of lymphoma, GIST, or carcinoid tumor rather than CD. Normal tumor markers and the absence of systemic symptoms were noted, yet no preoperative finding reliably suggested CD, consistent with previous reports in which definitive diagnosis is established only after surgical excision and histopathological evaluation [3,4,7].

Surgical resection remains the treatment of choice for both diagnosis and definitive management of unicentric CD. Complete excision is associated with excellent prognosis and very low recurrence rates [5,6]. The characteristic histopathological findings in our case — regressed germinal centers, preserved nodal architecture, prominent vascular hyalinization, and a comprehensive immunohistochemical panel effectively excluding lymphoma — were consistent with the hyaline vascular subtype, which is the most frequently reported form in unicentric disease [1,2]. Similar features have been described in previously published cases, further supporting the diagnosis [3,4].

In conclusion, unicentric mesenteric CD should be included in the differential diagnosis of unexplained intra-abdominal masses and mesenteric lymphadenopathy, particularly when clinical findings mimic malignant processes or surgical emergencies. Although preoperative diagnosis is seldom possible, complete surgical resection is both diagnostic and curative in localized disease, with excellent long-term outcomes.

Availability of Data and Materials

All data generated or analyzed during this study are included in this article.

Author Contributions

İES: study conception, guarantor, data collection, surgical management, and manuscript drafting. AT: data collection, surgical management, and manuscript review. FS: study conception, surgical management, and manuscript drafting and revision. SF: data collection and manuscript review. PA: assistance with pathological diagnosis and preparation of histopathological slides, histopathological evaluation, and manuscript review. All authors have been involved in revising the manuscript critically for important intellectual content. All authors gave final approval of the version to be published. All authors have participated sufficiently in the work to take public responsibility for appropriate portions of the content and agreed to be accountable for all aspects of the work in ensuring that questions related to its accuracy or integrity.

Ethics Approval and Consent to Participate

Ethical approval was not required for this study as it represents a single case. Written informed consent was obtained from the patient for publication. This study was conducted in accordance with the Declaration of Helsinki.

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Conflict of Interest

The authors declare no conflict of interest.

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