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Case Report

Small-intestinal Castleman disease mimicking a neoplastic lesion: a case report

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ABSTRACT

Castleman disease (CD) encompasses a group of rare lymphoproliferative disorders with diverse clinical presentations, histopathological features, and treatment approaches. This most commonly involves the mediastinal lymph nodes. Involvement of the small intestine is exceedingly uncommon and it often mimics more prevalent pathologies such as gastrointestinal stromal tumours (GIST), lymphoma, or neuroendocrine tumours. Early recognition is essential as surgical excision is typically curative in unicentric disease. A 38-years old male presented with complaints of dull aching pain in left upper abdomen for 1 year. CECT abdomen showed a soft tissue mass in the left lumbar region suggesting of neoplastic etiology. He underwent laparoscopic jejunal mesenteric soft tissue tumour excision with resection and anastomosis of jejunum. Post operative biopsy was suggestive of CD. CD of the small intestine is exceptionally rare and it is often misdiagnosed as other small bowel tumours. Pre-operative identification is extremely challenging due to non-specific clinical and radiological findings. Complete surgical excision remains the cornerstone of management for unicentric disease and is associated with excellent outcomes.

Keywords: Castleman's disease, Lymphoproliferative disorder, Small intestine tumour, Jejunum

INTRODUCTION

Castleman disease (CD) which is also known as angiofollicular lymph-node hyperplasia or giant node hyperplasia is a rare nonclonal lymphoproliferative disorder. Although CD commonly involves mediastinal, cervical, and abdominal lymph nodes, primary involvement of the small intestine is exceedingly uncommon, with only a limited number of cases described in the literature.[1]

This atypical anatomical location often leads to non-specific clinical features, delayed diagnosis, and often misinterpretation as neoplastic or inflammatory bowel pathology.

Given its rarity, especially within the small bowel, every documented case significantly contributes to the understanding of its clinical spectrum, diagnostic challenges, and optimal management. We present a case of CD arising from the small intestine, highlighting its clinical presentation, radiological findings, operative management, and histopathological features.

CASE REPORT

A 38-year-old male presented to our department with complaints of dull aching pain in the left upper abdomen. No history of loss of weight /loss of appetite. No h/o vomiting or altered bowel habits. He underwent open appendicectomy 20 years back.

Investigations

Routine laboratory tests were within normal limits. Serum chromogranin A: 17 ng/ml.

Contrast-enhanced CT showed (Figure 1) a well-defined homogenously enhancing soft tissue dense lesion measuring 3.5×4.4×3.7 cm (AP×TR×CC) noted in the left lumbar region with surrounding mild haziness. No evidence of calcification/fat components. Sub centimetric mesenteric lymph nodes noted.

MRI abdomen (Figure 2) showed a well-defined T2 hypointense/T1 hyperintense homogenous lesion showing diffusion restriction measuring 3×3.1×3.4 cm (AP×TR×CC) noted in the mesentery in left lumbar region.Few sub centimetric lymph nodes noted around the lesion. -likely neoplastic etiology-suggested HPE correlation

Upper GI endoscopy and colonoscopy showed normal study

Operative findings

The patient underwent laparoscopic jejunal mesenteric soft tissue tumour excision with resection and anastamosis of jejunum.

Intraoperatively

Encapsulated mass of size 6×5×4 cm noted over the jejunal mesentery, between its two leaves /folds. Mass appears in close contact with the jejunal wall. Multiple feeding vessels from jejunal mesentery noted.

Surgical technique

Initial assessment included laparoscopic exploration revealed an encapsulated mass of size 6×5×4 cm between the two folds of jejunum. (Figure 3 A). After giving adequate margin, the soft tissue tumour was separated from the jejunum using thunder beat (Figure 3 B). All the feeding mesenteric vessels were cauterised and divided using ligature. On posterior dissection the mass seems to be extending beyond posterior leaf of jejunum-About 15 cm of jejunum seems to be congested. Entire tumour was excised into with 15 cm of jejunum and adjoining node containing mesentery. Small midline laparotomy made and the specimen was extracted. Side to side jejunojejunal anastomosis was done in 2 layers.

Post operative HPE

Mesenteric soft tissue tumour-suggestive of (Figure 5), CD23-Positive in dendritic cells, CD10, CD20, BCL6, BCL2-Positive in follicles. CD3-Positive in T cell zone and HHV8-Negative.

Postoperative course

The recovery was uneventful. The patient was discharged on postoperative day 7. At 1 month follow up, FDG PET was done which showed no significant lesions. At 6-month follow-up, the patient remains asymptomatic with no recurrent lesions on imaging.

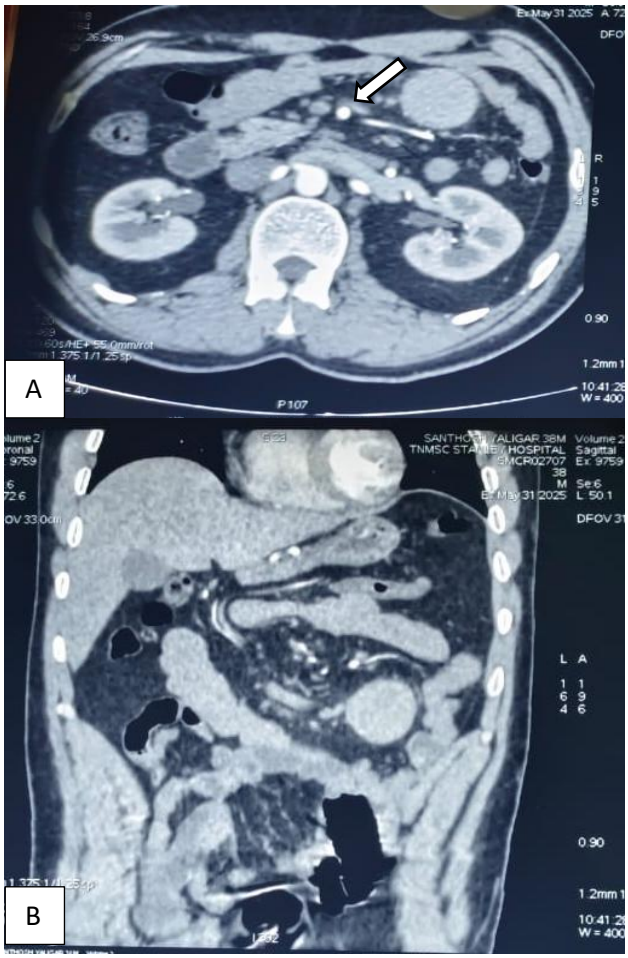

Figure 1 (A and B): Diagnostic imaging of CD-CECT abdomen.

*CECT: A well-defined homogenously enhancing soft tissue lesion measuring 3.5×4.4×3.7 cm noted in left lumbar region with surrounding mild haziness. Neoplastic etiology

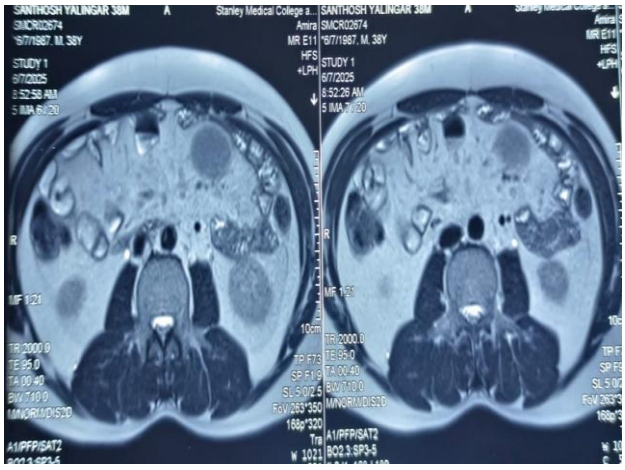

Figure 2: Diagnostic imaging of CD-MRI abdomen.

*MRI-Well defined T2 hypointense/T1 hyperintense homogenous lesion showing diffusion restriction measuring 3×3.1×3.4 cm noted in the mesentery in left lumbar region.

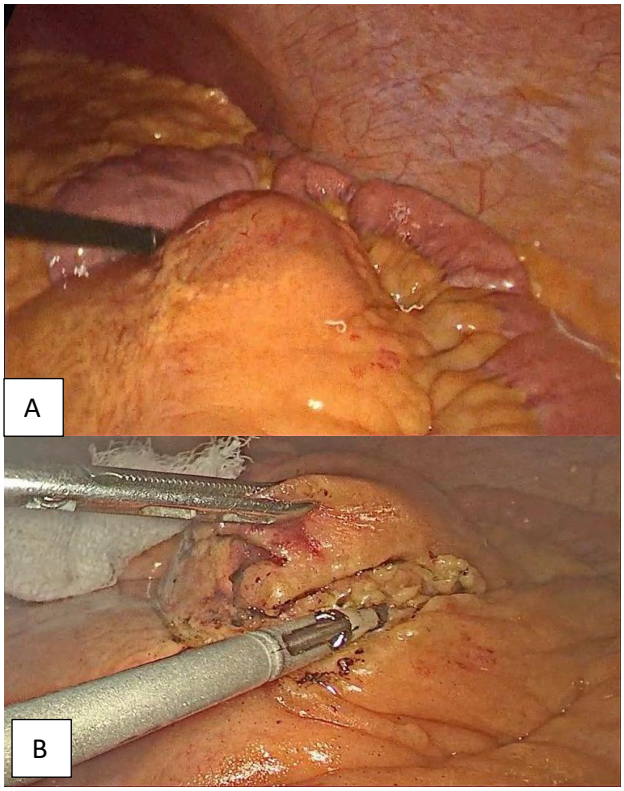

Figure 3 (A and B): Intraoperative surgical steps.
A-intra operative: Encapsulated mass of size 6×5×4 cm noted between the two folds of jejunal mesentery. B-Soft tissue tumour was separated from jejunal mesentry using thunderbeat.

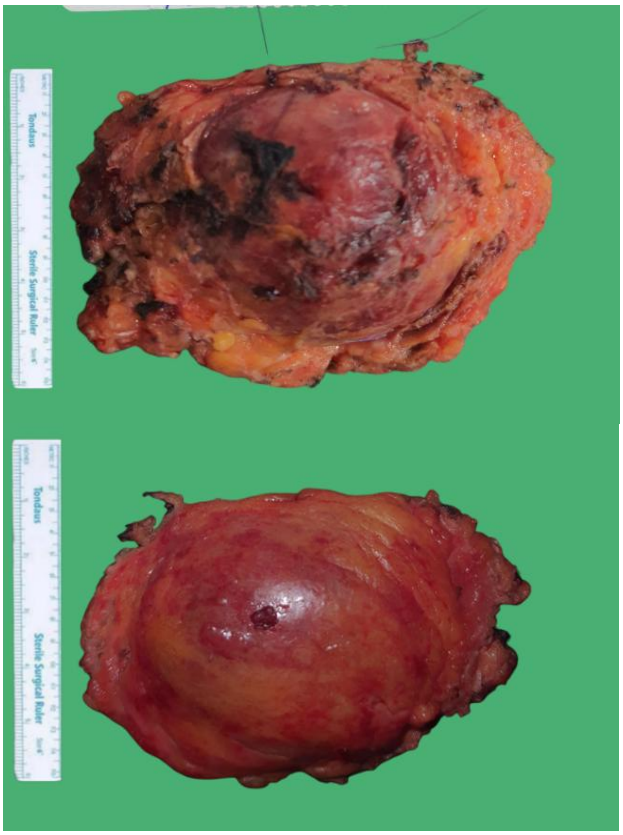

Figure 4: Postoperative specimen.

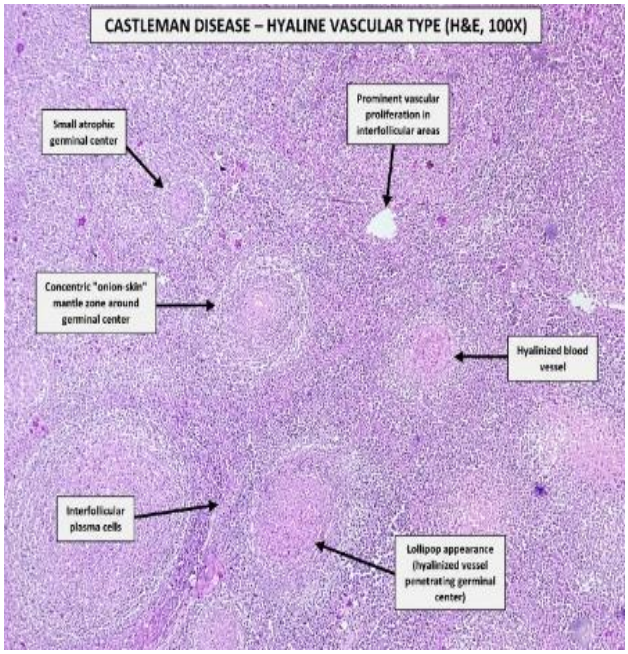

Figure 5: Post op HPE.

DISCUSSION

The CD was first reported in 1954. And in 1956 it was again described by Benjamin Castleman, in a case series of localized mediastinal lymph-node hyperplasia.[1] CD is broadly classified into unicentric CD (UCD), involving a single lymph node or region, and multicentric CD (MCD), which affects multiple lymph node regions. MCD has been further categorized into human herpesvirus 8 (HHV-8)-associated, idiopathic MCD (iMCD) and polyneuropathy, organomegaly, endocrinopathy, monoclonal plasma cell (PC) disorder, skin changes (POEMS)-associated MCD (POEMS-MCD).[2]

Unicentric CD (UCD) usually seen as a benign, localized lymphoid hyperplasia and often presenting as a solitary, indolent lymph node swelling. In contrast, multicentric CD (MCD) presents with multifocal lymphadenopathy, systemic inflammation, and constitutional symptoms and is potentially life threatening.[2] CD consists of several lymphoproliferative subtypes and all shares some histological features in the lymph nodes.[3] On the other hand, numerous clinical findings and aetiologies make the diagnosis of the disease challenging Histopathologically, CD is categorised into hyaline-vascular, plasma-cell, and mixed variants, with the hyaline-vascular subtype being the most frequent, particularly in UCD.[4]

Radiologically, CD may mimic gastrointestinal stromal tumours, neuroendocrine tumours, lymphoma, or mesenteric desmoid tumours, reflecting the non-specific nature of imaging features. As a result, definitive diagnosis is almost always dependent on histopathological examination, which shows

characteristic features such as regressed germinal centres, concentric mantle-zone layering ("onion-skin" pattern), and prominent vascular proliferation.[5,6]

The management of UCD is well established, with complete surgical excision being both diagnostic and curative in the majority of patients.[7] The prognosis is excellent, with recurrence being exceedingly rare following R0 resection. In contrast, MCD requires systemic therapy and carries a more variable clinical course.[3]

In the context of small-intestinal involvement, surgical resection is usually unavoidable due to the difficulty in obtaining pre-operative tissue diagnosis and the frequent suspicion of malignancy.[8]

Our case adds to the limited literature describing small-bowel CD and it reinforces key clinical messages: Small-intestinal CD should be considered a differential diagnosis for well-vascularised, solitary small-bowel masses; Histopathology remains the cornerstone of diagnosis; and surgical excision provides both diagnosis and definitive treatment for unicentric disease.

CONCLUSION

CD of the small intestine represents an exceptionally uncommon entity that can present as an isolated bowel mass and pose a significant diagnostic challenge. When encountered in this location, it is often indistinguishable from other small-intestinal neoplasms until histological evaluation is performed. Surgical resection remains central to management, both for establishing the diagnosis and achieving definitive treatment in unicentric disease. This case contributes valuable clinical insight into an unusual presentation and underscores the need for heightened awareness of this rare pathology among surgeons, radiologists, and pathologists.

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