Inflammatory Myofibroblastic Tumor Presenting as Ileocolic Intussusception: A Case Report

Nischal Khanal¹, Rupak Subedi², NIrajan Shrestha², and Shristi Shrestha²

¹Tribhuvan University Institute of Medicine ²Madan Bhandari Academy of Health Sciences

June 30, 2023

Title:

Inflammatory Myofibroblastic Tumor Presenting as Ileocolic Intussusception: A Case Report

- 1. Dr. Nischal Khanal, MBBS, MS General Surgery, Madan Bhandari Academy of Health Sciences (corresponding author).
- 2. Dr. Rupak Subedi, MBBS, Madan Bhandari Academy of Health Sciences.
- 3. Dr. Nirajan Shrestha, MBBS, MS General Surgery, Madan Bhandari Academy of Health Sciences.
- 4. Dr. Shristi Shrestha, MBBS, Madan Bhandari Academy of Health Sciences.

Consent: Written informed consent was obtained from the patient to publish this report.

Conflict of Interest Statement : There are no conflict of interest regarding this manuscript.

Abstract

We present a case of a 36-year-old male who presented with abdominal pain and vomiting. Inflammatory markers were elevated in routine investigations, while other laboratory parameters were within normal limits. Ultrasound imaging revealed a target lesion, which required further evaluation with a CT scan and confirmed ileocolic intussusception. An exploratory laparotomy showed a solid lesion measuring 5 by 6 cm in the cecum, along with evidence of ileocolic intussusception. A standard right hemicolectomy was performed, and the postoperative course was uneventful. Histopathological examination showed an inflammatory myofibroblastic tumor with nodular hyperplasia. The report highlighted the importance of assessing rare neoplastic causes in patients with intussusception.

Keywords: Inflammatory Myofibroblastic tumor, intussusception, hemicolectomy, spindle cell proliferation, abdominal pain, vomiting

Introduction: Inflammatory Myofibroblastic tumors (IMTs) are rare neoplasms characterized by proliferative Myofibroblastic spindle cells accompanied by a prominent inflammatory infiltrate. [1] IMTs predominantly affect children and young adults but can occur at any age. [2] The most common site of occurrence is the lung, followed by the extrapulmonary sites like liver, pancreas, intestine, and bones [3]. Intestinal involvement is relatively rare, and ileocolic intussusception caused by IMT is an even rarer presentation [4]. We report a case of a 36-year-old male who presented with abdominal pain, vomiting, and subsequent diagnosis of IMT leading to ileocolic intussusception.

Case presentation: A 36-year-old male presented to the emergency department with a three-day history of diffuse abdominal pain, followed by vomiting. The pain started from the right lower quadrant and later generalized. On physical examination, mild tenderness was present on deep palpation in the right lower

quadrant of the abdomen. The patient had no significant medical history and denied any recent weight loss or changes in bowel habits.

Laboratory investigations revealed raised inflammatory marker. A complete blood count revealed elevated WBCs $(17,100/\mu L)$ with neutrophilia (80%). Blood urea nitrogen and creatinine level were normal and other hematological and biochemical parameters were within normal limits. An ultrasound examination of the abdomen showed a target lesion in the right lower quadrant, raising the suspicion of intussusception. A subsequent CT scan confirmed segmental, circumferential thickening of the terminal ileocolic junction showing a target stratification pattern (Fig 1). The protrusion of terminal ileum into ascending colon gave an intraluminal pseudomass appearance (Fig 2). The small bowel loops proximal to ileocolic junction was dilated with outer-to-outer diameter of 3.2cm and mottled appearance giving small bowel feces sign.

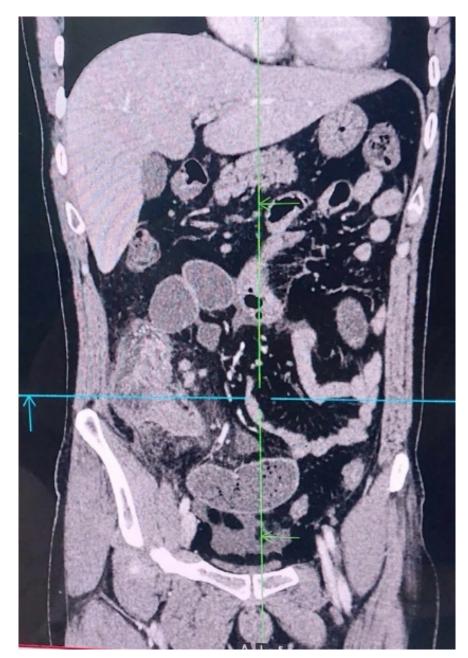
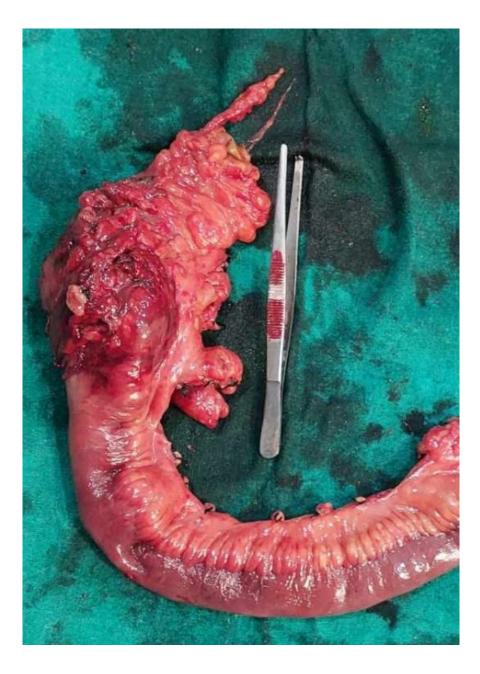




Fig 2: Coronal image showing intraluminal

pseudomass appearance with perlesional

fat strandings Fig 1: Axial image showing thickened ileocolic junction with target stratification pattern



The patient underwent an exploratory laparotomy. Intraoperatively there was a solid lesion measuring approximately 5×6 cm, well-demarcated smooth mass involving the cecum, approximately 2 cm proximal to ileocolic junction (Fig: 3 and 4). There was no evidence of ascites, no mesenteric lymphadenopathy and liver surface appeared normal. Lead point of ileocolic intussusception was the mass and right sided standard hemicolectomy performed. On cut surface, the lesion had diffuse grey to yellowish appearance. The procedure completed without complications and the specimen was marked and sent for histopathological examination.

Fig 3: Tumor mass removed along with segment of ileum



Fig 4: Intraoperative image with tumor insitu

The post-operative course was unremarkable, and after 24 hours of observation in Intensive care Unit (ICU) the patient was transferred to the surgery ward. He recovered well and discharged after fifth Post-operative day.

Histopathological examination of the resected specimen showed an Inflammatory Myofibroblastic tumor. The tumor showed nodular hyperplasia with diffuse eosinophilic infiltrate, subserosal bland spindle to stellate cells proliferation along with lymphoid aggregates, lymphocytes, plasma cells, and eosinophils.

Discussion: Inflammatory Myofibroblastic tumors being rare neoplasms with diverse clinical presentations can affect various organs like lungs, liver, endolarynx, maxillary sinus, oral cavity etc.[5] However, their involvement in the gastrointestinal tract is particularly rare and their expression as intussusception is even rarer. [4]. Their exact etiology remains uncertain but they can even mimic various inflammatory conditions and their behavior ranges from benign to locally aggressive or rarely malignant, making the diagnosis challenging.

Some common causes of adult ileocolic intussusception are adhesions, polyp, inflammatory bowel disease,

gastro intestinal tumor (GIST) etc. [6]. Inflammatory Myofibroblastic Tumor Presenting as Ileocolic Intussusception is a rare diagnosis, so it is even difficult for a resources equipped centers to diagnose. Diagnosing this rare condition in Hetauda Hospital, a district hospital located in sub-urban region with inadequate resources is quite challenging. The diagnosis of this condition at our center either reflects the inadequacies in studies or not frequently reported.

In our case, the patient's presentation with abdominal pain, vomiting, and imaging findings of intussusception prompted further investigation, leading to the diagnosis of an IMT. Surgical resection followed by histopathological examination of the resected specimen performed.

Immuno histochemical staining (IHC) which helps to identify specific marker expressed by tumor cells like Smooth muscle actin (SMA), anaplastic lymphoma kinase (ALK Protein), vimentin, and CD34. These IHC features can help in distinguishing benign or malignant nature of the tumor. Together in conjunction with histopathological features, they can help to establish confirmatory diagnosis. However, in sub-urban area like ours, patient presents with their complaints and after getting relief from their issues, they do not come up to hospital for follow up. Therefore, in our case we are unable to give Immunohistochemistry (IHC) reports as he was lost to follow up.

Further studies, research and case reports are necessary to understand the etiology, behavior, and optimal management strategies for IMTs, particularly those involving gastrointestinal tract. Additionally, increased awareness among clinicians is also essential to ensure early recognition, prompt diagnosis, and appropriate surgical intervention, as demonstrated in this case, are crucial for optimal management and favorable patient outcomes of these rare tumors.

Conclusion :

This case report shares a very rare condition of inflammatory myofibroblastic tumor (IMT), presenting with ileocolic intussusception in a middle-aged man, which is extremely uncommon and can be confused with other inflammatory conditions however, through diagnostic evaluation and multidisciplinary approach can result in early diagnosis, optimal management and favorable patient outcomes of these rare tumors.

References

1. Koyuncuer, A., Inflammatory myofibroblastic tumor of the small-bowel mesentery: A case report of nonspecific clinical presentation and a review of the literature. Int J Surg Case Rep, 2014.5 (12): p. 1214-7.

2. Camela, F., et al., Pulmonary Inflammatory Myofibroblastic Tumor in Children: A Case Report and Brief Review of Literature. Front Pediatr, 2018. 6: p. 35.

3. Khalil, S., T. Ghafoor, and A.K.F. Raja, *Inflammatory Myofibroblastic Tumor: A Rare Presentation and an Effective Treatment with Crizotinib.* Case Reports in Oncological Medicine, 2020.2020 : p. 6923103.

4. Hameed, T., et al., Acute Intestinal Obstruction Due to Ileocolic Intussusception in an Adult; A Rare Presentation of Inflammatory Myofibroblastic Tumor. Am J Case Rep, 2020. **21** : p. e920438.

5. Palaskar, S., et al., Inflammatory myofibroblastic tumor.Contemp Clin Dent, 2011. 2 (4): p. 274-7.

6. Tarchouli, M. and A. Ait Ali, Adult Intussusception: An Uncommon Condition and Challenging Management. Visceral Medicine, 2020.37 (2): p. 120-127.







