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## Myofibroblastic infiltration of the bowel: A case report and literature review

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## ABSTRACT

**INTRODUCTION:** Inflammatory myofibroblastic tumours (IMTs), are uncommon tumours which can act with malignant potential. The management of these tumours can be extremely problematic but are often referred to surgical multi-disciplinary team meetings with the intention of surgical and oncological management (Chaudhary [1]).

**CASE REPORT:** A 69-year-old gentleman was admitted with a 2-day history of abdominal pain and vomiting, and a 4-day history of absolute constipation. CT Abdomen Pelvis demonstrated distended loops of small bowel with pronounced fluid levels but no transition point. Intra-operative findings showed a right ileocolic mass adherent to the pelvic side wall and omental caking. Biopsies showed a florid myofibroblastic reaction, not a malignant process.

**CONCLUSION:** IMTs are aggressive lesions but metastases is rare. Abdominal IMTS are difficult to diagnose and manage as they are often initially mistaken for lymphoma or peritoneal metastases. The therapy of choice is surgical resection of the tumour (Firat et al. [3]).

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### 1. Introduction

Inflammatory myofibroblastic tumours (IMTs), are uncommon tumours found most commonly in the lung, mesentery, and omentum [1].

We present the case of a 69-year-old gentleman who presented in intestinal obstruction and diagnosed with inflammatory myofibroblastic tumour of the omentum. This case was reported in line with the SCARE criteria [2].

### 2. Case report

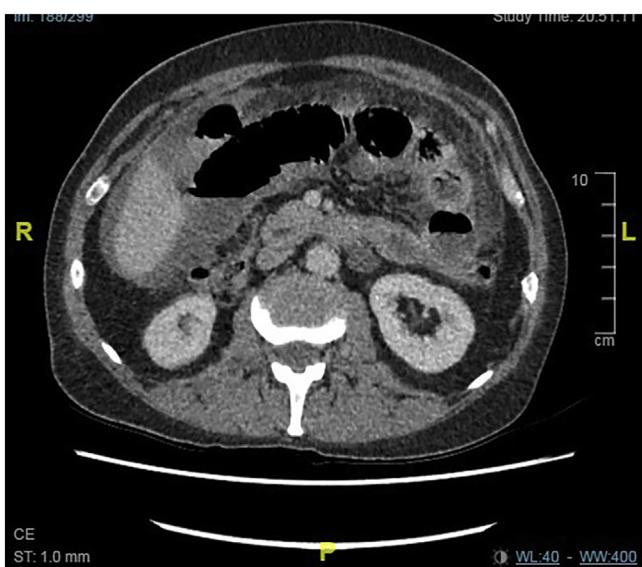
A 69-year-old gentleman presented to the Emergency department with a 2-week history of central and lower abdominal pain, bloating associated with nausea, bilious vomiting, and 3-days of absolute constipation. He presented a week prior to his GP with similar symptoms and was treated for a UTI with a course of oral ciprofloxacin. His past medical history was significant for recurrent UTIs and an underactive bladder for which he self-catheterizes. On examination, abdomen was distended with generalized tenderness. Rectal examination showed an empty rectum. Blood results revealed WCC  $12.8 \times 10^9/L$ , and CRP of 54 mg/L. Erect chest x-

ray showed no evidence of pneumoperitoneum, abdominal x-ray showed dilated small bowel loops.

A contrast CT abdomen/pelvis showed nonspecific free fluid throughout the abdomen, dilated small bowel loops and a segmental jejunal loop with bowel wall thickening and omental stranding (Figs. 1 and 2). An exploratory laparotomy revealed copious straw-coloured ascites, a large ileo-caecal mass adherent to the posterior abdominal wall, heavy serosal and meso-colonic involvement from the caecum to the sigmoid with marked omental caking. Given these findings, the mass was deemed irresectable. A small bowel resection, excision omental biopsy and an end ileostomy was performed. Tumour markers showed a raised Ca-125 and normal CEA and Ca19-9. Histopathology showed extrinsic infiltration by serosa-based spindle and epithelioid tumour. The cells showed prominent nuclear pleomorphism and frequent mitosis. The tumour infiltrated through the muscularis propria and mucosa leading to focal perforation. Immunophenotyping favoured a florid myofibroblastic reaction rather than a malignant process (Figs. 3–7). He recovered well post-operatively and was discharged. He was readmitted a week later with ongoing small bowel obstruction and intra-abdominal sepsis. CT abdomen/pelvis showed 3 fluid collections causing a reactive ileus. He was treated conservatively with nasogastric tube decompression, intravenous fluids and total parenteral nutrition. Unfortunately, during his admission, he vomited, aspirated, followed by cardiopulmonary arrest and he passed away despite attempted resuscitation. Post mortem revealed that the small bowel and colon were encased in thick, dense fibrofatty adhesions also encasing the pancreas, spleen, diaphragm, inferior and

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**Fig. 1.** Axial view of CT abdomen and pelvis showing high density ascites and omental stranding.



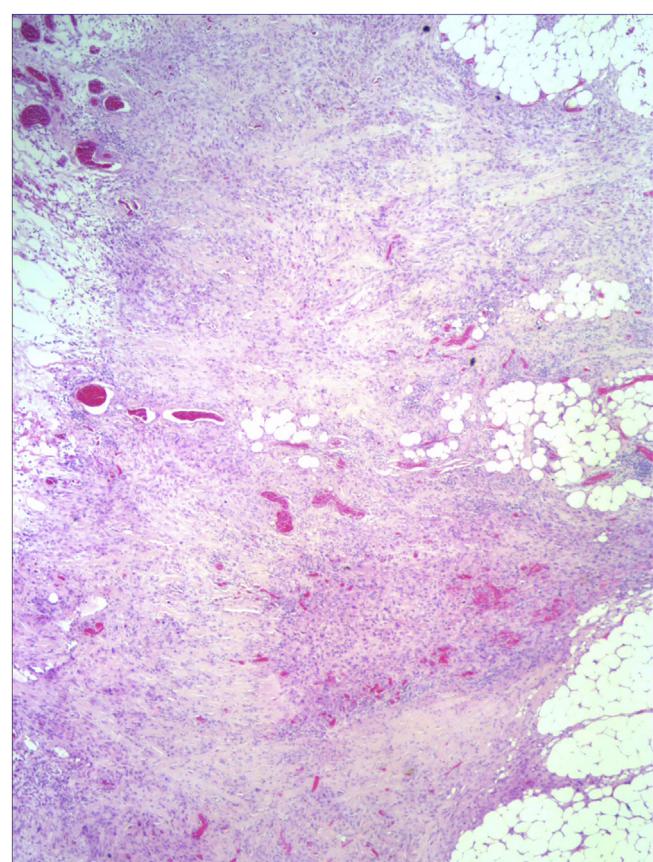
**Fig. 2.** Coronal view of CT abdomen and pelvis showing high density ascites and omental stranding.

posterior aspect of the liver, gallbladder, and the stomach. This had formed one large mass.

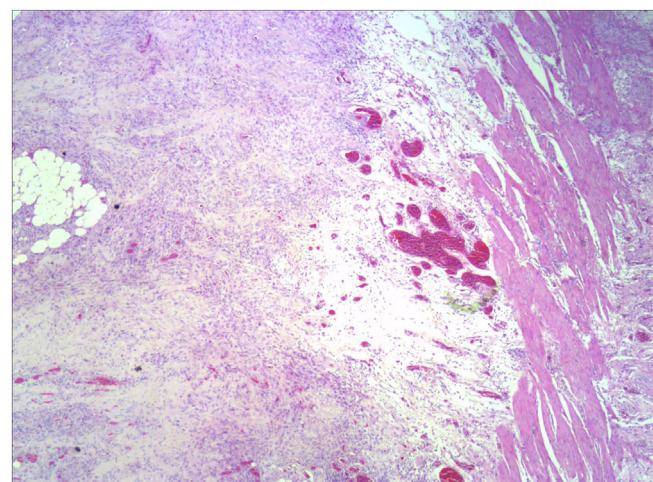
### 3. Discussion

IMTs had previously been accepted as a subtype of the group of tumours called inflammatory pseudotumours [3]. Previously inflammatory fibrosarcomas [1], IMTs are now recognized to comprise their own discrete diagnosis.

The terminology 'inflammatory pseudotumour' was first coined by Umiker and Iverson in 1954 in 4 cases reports involving the lung [4]. IMTs primarily occur in the lung and upper respiratory tract but may affect any organ system with protean manifestations [5] including heart, liver, omentum, mesentery, vagina, and kidneys [4]. Once thought to be reactive, these lesions are now



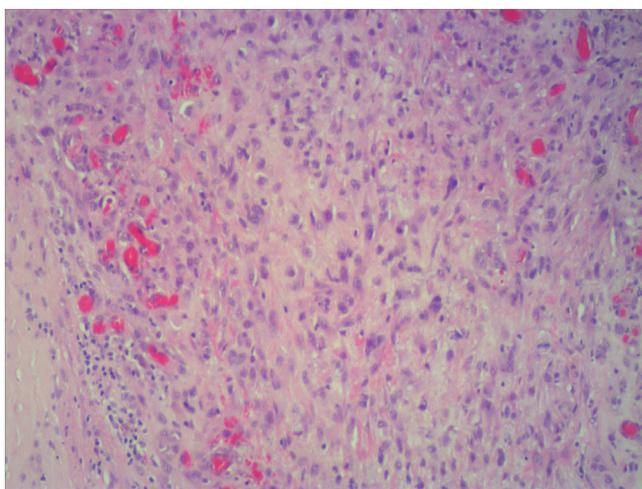
**Fig. 3.** Histopathology of omental biopsies showing benign fibroblastic proliferation.



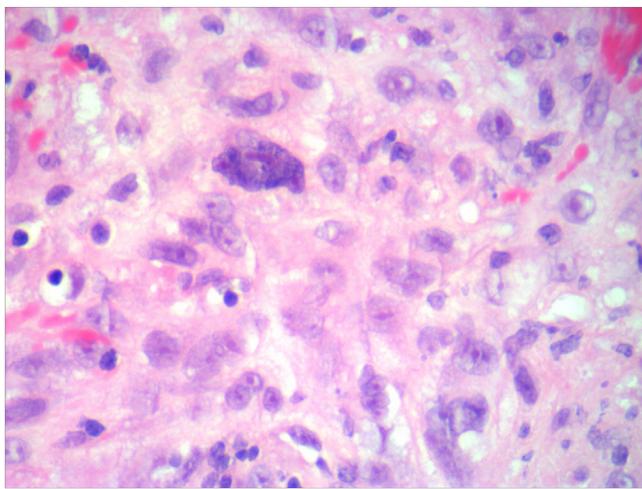
**Fig. 4.** Histopathology of omental biopsies showing benign fibroblastic proliferation.

considered to be neoplastic thus the terminology changed from inflammatory myofibrohistiocytic proliferations or pseudosarcomatous myofibroblastic proliferations to IMTs [1].

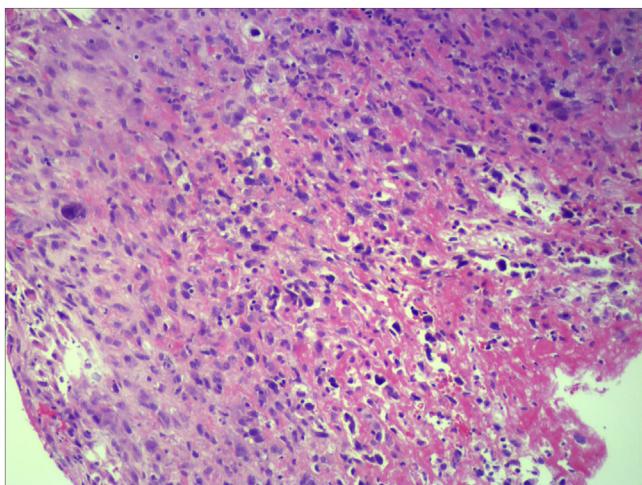
IMTs are most common in children [4] with a mean age of 10 years. They are reported in patients between 3 months to 46 years [1]. IMTs have a slight male predominance but no race predominance [1]. The current histopathological definition of an IMT is a distinctive neoplasm composed of myofibroblastic mesenchymal spindle cells accompanied by an inflammatory infiltrate of plasma cells [3].



**Fig. 5.** Histopathology of omental biopsies showing benign fibroblastic proliferation.



**Fig. 6.** Histopathology of omental biopsies showing benign fibroblastic proliferation.



**Fig. 7.** Histopathology of omental biopsies showing benign fibroblastic proliferation.

Omental IMTs is a rare form of abdominal IMTs. Less than 15 cases have been reported in the literature [6]. Two case reports by Cifti et al. and Aboulafia et al. reported a first presentation of IMT with intestinal obstruction like our case [7,8]. Intra-abdominal IMTs often mimic malignant neoplasms because of its local invasiveness and its tendency to recur [5].

Historically, IMTs were considered benign until Meis and Enzinger published a series of 38 cases, primarily intra-abdominal, with follow up data showing significant recurrences [6].

The aetiology remains unclear [1]. Evidence to support a directly infectious aetiology is scanty [5]. Other potential causes include an immunological response to an infectious or non-infectious agent. Some reports demonstrated that IMTs are true neoplasms and some believe them to be a low-grade sarcoma with inflammatory cells as it has the potential for local infiltration, recurrence, multicentricity, and rarely metastases [5].

There is limited data available on the natural history of IMTs [6]. Features suggesting more aggressive behaviour include multifocal tumours, retroperitoneal location, infiltration of adjacent structures, and incomplete excision. Pathologic features such as tumour cellularity, mitosis and necrosis do not correlate with outcome [6].

CT abdomen/pelvis is the most commonly used radiological investigation. Liu et al reported the most common imaging characteristics were presence of heterogeneity showing varying degrees of contrast enhancement. Two main patterns were individualized. In type 1, the tumour had a distinct boundary without a lobular appearance and displayed hypo-enhanced enhancement in correlation with the mainly histopathologic findings of spindle cells myxoid and hypocellular fibrous. In type 2, the lesions exhibited indistinct boundaries or complete capsule, ill-defined growth patterns or low intralesional attenuation with marked heterogenous or circumferential enhancement, which correlated well with the presence of abundance of micromodule and inflammatory cell infiltration [9]. Interestingly, our patient didn't have a discrete mass, or any features described above.

Treatment of choice in IMTs is non-aggressive surgical resection. In cases where complete excision is not possible, treatment is symptomatic over other modalities of therapy. In cases of local recurrence, re-excision is the treatment of choice. Chemotherapy is reserved for progressive disease after complete resection [1].

Literature about recommended therapy for IMTs in whom complete resection is not possible is limited thus there are no clear-cut treatment guidelines for such patients because most of our knowledge of IMTs comes from individual case reports and case series [1].

Following complete resection, approximately 23% of IMTs locally recur. Rarely IMTs undergo malignant transformation and metastasize. Risk of distant metastases is less than 5%. 5-year survival is approximately 87% [1].

#### 4. Conclusion

IMTs are aggressive lesions but metastases are rare. Abdominal IMTs are difficult to diagnose and manage as they are initially mistaken for lymphoma or peritoneal metastases. The treatment of choice is surgical resection of the tumour [6].

#### Conflicts of interest

No conflicts of interest.

#### Sources of funding

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**Ethical approval**

Ethical approval exempt from Maidstone and Tunbridge Wells NHS Trust as case report.

**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.

**Author contribution**

Munir Tarazi – study design, data collection, writing, editing.  
Caroline Louise English – study design, data collection, editing.  
Katherine Guest – study design, editing.  
Dinesh Balasubramaniam – study design, editing.

**Registration of research studies**

Researchregistry4282.

**Guarantor**

Mr Dinesh Balasubramaniam.

**Provenance and peer review**

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