

Case Report

Mesenteric cysts: a rare cause of abdominal pain

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Gastrointestinal stromal tumours (GIST) constitute the most important group of primary mesenchymal tumours of the gastrointestinal tract. They may be difficult to diagnose because of their non-specific presentation. We report a young man who presented with right upper quadrant pain due to a GIST, diagnosed following surgical exploration.

CASE REPORT A 27 year-old man was admitted with a history of right upper quadrant pain, occurring five days after flying home to the United Kingdom from Australia via Singapore. The pain was constant and throbbing in nature, and was exacerbated by deep inspiration. He had a poor appetite. He gave a history of passing pale stools and dark urine for one week, a number of months previously. He had been investigated two years previously for melaena and bleeding per rectum. Colonoscopy, gastroscopy, small bowel series, ultrasound of abdomen and isotope Meckel's scan were all negative. There were no risk factors for hepatitis, and he had received immunisation against Hepatitis A and B prior to his journey to Australia.

On examination, the abdomen was soft with mild tenderness in the right upper quadrant. Murphy's sign was negative. There was one finger-breadth of hepatomegaly noted. There were no other abnormal examination findings. He remained afebrile following admission. The provisional diagnosis on admission was acute cholecystitis.

Investigations revealed a slightly raised WCC at $13.4 \times 10^9/l$ with neutrophils of $10 \times 10^9/l$. Liver function tests were normal apart from an elevated γ -glutamyl transferase 253 U/l. D-dimer was elevated at 0.62 $\mu g/ml$, CRP was 67 mg/l. Serological tests for *Legionella*, *Leptospirosis* and *Brucella* were

all negative. No malaria parasites were seen on peripheral blood smear. Complement showed a raised C3 with a normal C4. Immunoglobulins, autoimmune screen and hepatitis serology were negative.

In view of the pleuritic nature of the pain, the raised D-dimer level and the long aeroplane flight, a ventilation perfusion scan was performed, which was normal. Chest X-ray and abdominal X-ray were also normal. Ultrasound of abdomen demonstrated a contracted gallbladder with no calculi.

The patient continued to complain of persistent abdominal pain, and CT scan abdomen and pelvis was performed. This revealed two areas of decreased attenuation anteriorly within the abdominal cavity, suggestive of mesenteric cysts, the largest measuring 7.7×5.8 cms. and the smaller 5×3 cms (*Figure 1*). The lesions were surrounded by a well defined wall. There was no adjacent inflammation or lymphadenopathy and no abnormality was seen within the liver. The spleen was enlarged measuring 16.7 cms. in bipolar length. Subsequently hydatid serology was performed and was negative.

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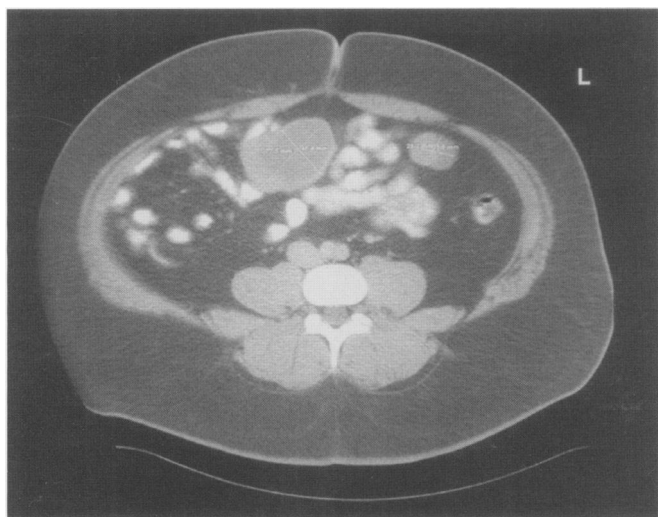


Fig 1. CT scan of abdomen demonstrating two mesenteric cysts.

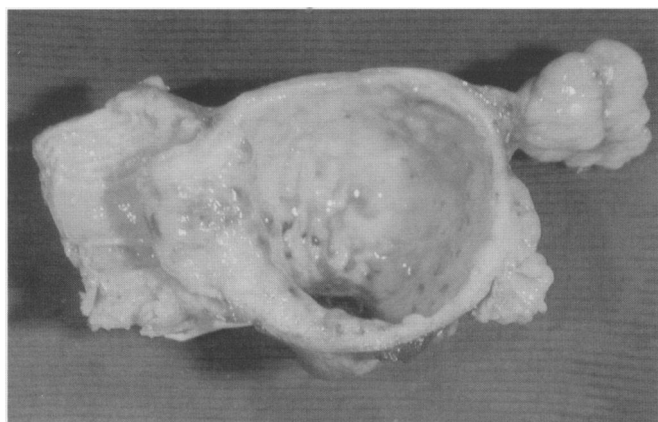


Fig 2. Resected surgical specimens.

Surgical excision of the mesenteric cysts was performed without complication (*Figure 2*). Histological examination revealed both lesions to be gastrointestinal stromal tumours (GIST). Both tumours stained strongly positive for C-kit (CD 117) and also expressed CD34. A low mitotic activity was noted. No further oncological treatment was required. He made a full recovery and remains well at 6-month follow-up.

DISCUSSION

Gastrointestinal stromal tumours (GISTs) constitute the most important group of primary mesenchymal tumours of the gastrointestinal tract. They are known for their diversity in clinical behaviour. Two problems may arise for the clinician. Firstly, the diagnosis may be difficult to establish, and secondly, it may

be difficult to determine the future behaviour of the tumour and the necessary choice of further chemotherapy, if required.¹ One large study by Langer *et al*² aimed to identify prognostic markers of tumour progression in 48 GISTs resected from 39 patients in a single tertiary referral centre over an eleven year period. GISTs were classified as low or high risk on the basis of tumour size, mitotic rate and/or proliferation. This was confirmed by Wong *et al*³ who recently reported a study aimed at determining whether immunohistochemical markers increase the accuracy of predicting prognosis for GISTs, and found that the mitotic count remains the best predictor of outcome following surgical resection of gastric GISTs. Ki67 immunohistochemistry and p53, Bcl-2 and cyclin D1 immunohistochemistry provide no additional prognostic features. Our patient's tumours had a low mitotic activity suggesting that the tumours were benign and no further oncological treatment was required.

Cyto-genetic and comparative genomic hybridisation (CGH) studies have indicated characteristic chromosomal patterns in GISTs.⁴ This technique has been combined with immunohistochemical analysis. Bergmann *et al* reported that the most common and characteristic alteration in GISTs were loss of chromosomes 14 and 22.⁴

In our case both tumours were strongly positive for C-kit (CD 117), and also expressed CD34. Interestingly, Nishida and Yasumasa⁵ report that GISTs are composed of KIT-positive mesenchymal-origin spindle - or polygonal-shaped tumour cells in the gastrointestinal tract. The gain-of-function mutations in the C-kit gene (90%) or platelet-derived growth factor receptor alpha (PDGF-R alpha) gene (5%) are now considered to be causative for GISTs. Therefore, treatments such as ST 1571 (Glivec), a molecule designed to selectively inhibit Bcr-Abl, KIT and PDGF-R activity, has recently been introduced in the management of these tumours. It shows high response-rate and efficacy for non-resectable and/or relapsed GIST (partial response 60%).⁵

In summary, we present a young man with a GIST as a rare cause of abdominal pain. His tumour had a low mitotic index and his prognosis would appear to be good following successful surgical excision.

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