


Combination of ectopic pancreas and intestinal malrotation presenting as non-specific right iliac fossa pain in a SARS-CoV-2 positive patient

Woochan Hwang ,¹ Kristof Nemeth,¹ Annabelle White,¹ Luca Bonomo^{1,2,3}

¹General Surgery, Guy's and St Thomas' NHS Foundation Trust, London, UK

²General Surgery, University Hospitals Dorset NHS Foundation Trust, Bournemouth, UK

³Honorary Lecturer, King's College London, London, UK

Correspondence to

Dr Woochan Hwang;
woochan.hwang@gstt.nhs.uk

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SUMMARY

We describe the case of a 31-year-old man who presented with a 3-day history of right iliac fossa pain with associated nausea and vomiting. He denied any previous incidents of abdominal pain and had no relevant medical history or family history to note. Given the typical history, examination findings of localised peritonism and infection risk, he was taken to theatre for laparoscopic appendicectomy without diagnostic imaging. Intraoperatively, we noted gut malrotation and an inflammatory jejunal mass which was resected after converting to a mini-laparotomy. The inflammatory mass was reported to be an ectopic pancreatic tissue from histology. Given that this patient had tested positive for SARS-CoV-2 on admission, we propose a possible case of SARS-CoV-2 infection triggering inflammation of the ectopic pancreatic tissue.

BACKGROUND

Ectopic pancreas (also known as annular pancreas or heterotopic pancreas) is defined as pancreatic tissue found in the body, lacking vascular or anatomical connection to the pancreas.¹ The incidence ranges from 0.5% to 13.7% on autopsy studies, where the vast majority are found on the stomach, duodenum and jejunum.² The reported frequency of jejunal involvement is between 0.5% and 35%. Ectopic pancreas are usually incidental findings, but rarely present with abdominal pain, obstruction, bleeding or inflammatory changes.^{1,2}

Intestinal malrotation has been estimated to occur in approximately 1 in 500 live births, predominantly presenting within the first month of life.³ Because it is rarely suspected in older children and adults, it is often an incidental finding on imaging or on laparotomy, but may present acutely with complications such as volvulus. Non-contrast radiography is of little diagnostic benefit, but the position of the small and large bowel may be suggestive. Contrast studies, especially CT scans, are used most commonly with a sensitivity of 97.5%⁴ and may contain additional signs such as the vertical positioning of the superior mesenteric vein and artery.⁵ Although controversial there is growing evidence on the benefits of surgical correction of malrotation in the adult population.^{3,4}

The combination of ectopic pancreas and malrotation is exceedingly rare and has been reported mostly as a congenital malformation diagnosed in the paediatric population.^{6,7} We found one case⁸ reported in the adult population where a 39-year-old man presented acutely with volvulus.

CASE PRESENTATION

A 31-year-old man presented to the emergency department in October 2020 with a 3-day history of gradually worsening right iliac fossa pain associated with nausea and vomiting. He had been to the emergency department of another hospital 12 hours before, where he was discharged with analgesia. He denied any previous episodes or urinary symptoms, but described that he had been constipated for 3 days but opened his bowels just prior to presenting to us. There was no significant medical history or family history of cancer, inflammatory bowel disease and situs inversus. He does not have any regular medications. He denied any smoking history or use of recreational drugs and reported occasional alcohol consumption.

INVESTIGATIONS

On examination, there was right iliac fossa tenderness on palpation and rebound tenderness. There was no renal angle tenderness and the abdomen was soft with no palpable mass. His basic observations were normal. Blood on admission showed white cell count of $11 \times 10^9/L$, C-reactive protein of 55 mg/L and normal liver function test including amylase of 70 mg/L. No D-dimer or fibrinogen levels were sent, but international normalised ratio (INR) and activated partial thromboplastin time (APTT) were both normal. Venous blood gas showed lactate of 0.8 mmol/L.

We note that his SARS-CoV-2 nasal and oropharyngeal swab taken as a standard procedure during admission came back positive on PCR testing despite having no respiratory symptoms on admission. The patient provided us with a negative test result from a SARS-CoV-2 nasal swab taken 6 days before his abdominal pain started.

His chest X-ray showed clear lung fields, with normal anatomical position of the visible visceral organs, including the liver and stomach. No further abdominal imaging was performed at the time as it would have delayed intervention in a patient developing peritonism, but also to minimise risk of nosocomial infection.

DIFFERENTIAL DIAGNOSIS AND TREATMENT

Given the typical history and examination findings, the patient was consented and taken to theatre for a laparoscopic appendicectomy. The operation was performed in full personal protective equipment as per local trust policy. Following port insertion, we noted the organs from the ligament of Treitz to



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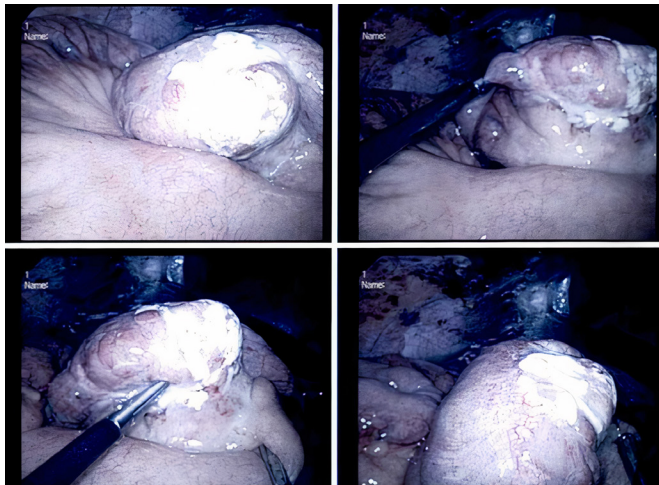


Figure 1 Intraoperative images of the ectopic pancreatic mass on the jejunum.

the descending colon and rectum were transposed, with normal anatomy of the liver, stomach and proximal duodenum. The appendix was located in the left lower quadrant and was mildly inflamed with significant amount of surrounding free pus. We suspected that the appendix inflammation may be secondary to the free pus rather than the primary cause given the relatively mild degree of inflammation.

While scanning the bowel for a potential primary cause, an inflammatory mass (figure 1) was noted on the jejunum 20 cm distal to the duodenal jejunal flexure with free pus, pelvic fluid and adhesions. The appendix was dissected first and removed through the umbilical port. The procedure was then converted to a mini-laparotomy for resection of the inflammatory mass found on the jejunum. The resected mass was removed through a small Alexis retractor and was later reported to be an inflamed pancreatic tissue from histology.

OUTCOME AND FOLLOW-UP

The patient made good postoperative recovery but had mild (<2 L) oxygen requirement for 2 days with breathlessness on exertion. He did not have any temperature spikes and the oxygen requirement was deemed likely due to SARS-CoV-2 infection.

He was kept nil by mouth for 3 days until he passed a water-soluble contrast swallow test. The patient tolerated oral consumption well and opened his bowels. He was discharged on day 5 following his operation.

Histology confirmed acute inflammation of Heinrich type 1 ectopic pancreatic tissue (figure 2) with extensive necrosis of the underlying fat, with fibrinous serositis. The background small bowel and the appendix were within normal histological limits and did not show any unexpected findings. The peritoneal fluid was negative for acid-fast bacilli culture. Unfortunately, we did not have any SARS-CoV-2 testing performed on the histology or peritoneal fluid sample.

At the 6-week follow-up appointment, he reported good recovery with no further complications. However, the patient attended the emergency department with increased abdominal pain around the surgical wound 2 weeks after the follow-up appointment. He did not report any nausea or vomiting and had been opening his bowels normally. Examination showed a hypertrophic scar which was tender to light touch but was otherwise normal. Blood tests and CT imaging of the abdomen were all non-significant. The case was discussed in the benign

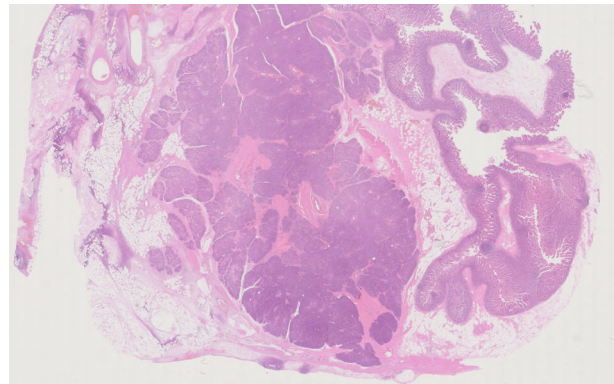


Figure 2 Histology slide of the Heinrich type 1 ectopic pancreas showing extensive inflammation.



Figure 3 CT of the abdomen and pelvis performed 2 months postoperatively in the emergency department. (Left) Coronal view demonstrating small bowel loops on the right and large bowel on the left side of the abdomen. (Right) Axial view demonstrating vertical orientation of the superior mesenteric artery (A) and superior mesenteric vein (V).

hepatobiliary multidisciplinary meeting, where they noted that the CT scan (figure 3) was consistent with partial malrotation of the gut rather than situs inversus. He was referred for an MRI small bowel investigation given the persisting pain.

MRI small bowel study (figure 4) performed 4 months postoperatively showed small bowel adhesions on the background of small bowel malrotation. No small bowel pathology that explains the patient's persisting pain was found in the study. The patient had clinically improved at this point, and we explained the MRI findings to the patient with a plan for follow-up in 6 months. We have provided imaging studies including an abdominal radiograph (figure 5), which demonstrates small bowel malrotation with small bowel loops lying on the right side of the abdomen and the colon lying on the left.

DISCUSSION

There are few reported cases where inflammation of the ectopic tissue was identified either in isolation⁹ or simultaneously¹⁰ with inflammation of the anatomical pancreas. During the COVID-19 pandemic, we have also seen cases of SARS-CoV-2 infection associated with acute pancreatitis,^{11 12} with growing evidence to suggest that the novel coronavirus could be a cause of acute pancreatitis. We believe that the abundant ACE2 expression in pancreatic cells and SARS-CoV-2 using ACE2 to enter human cells is a plausible mechanism. The current evidence base is insufficient to show causality,¹³ and our purpose is not to prove any mechanism but to describe a relevant case as there is currently no reported case of SARS-CoV-2 infection associated with inflammation of an ectopic pancreas.

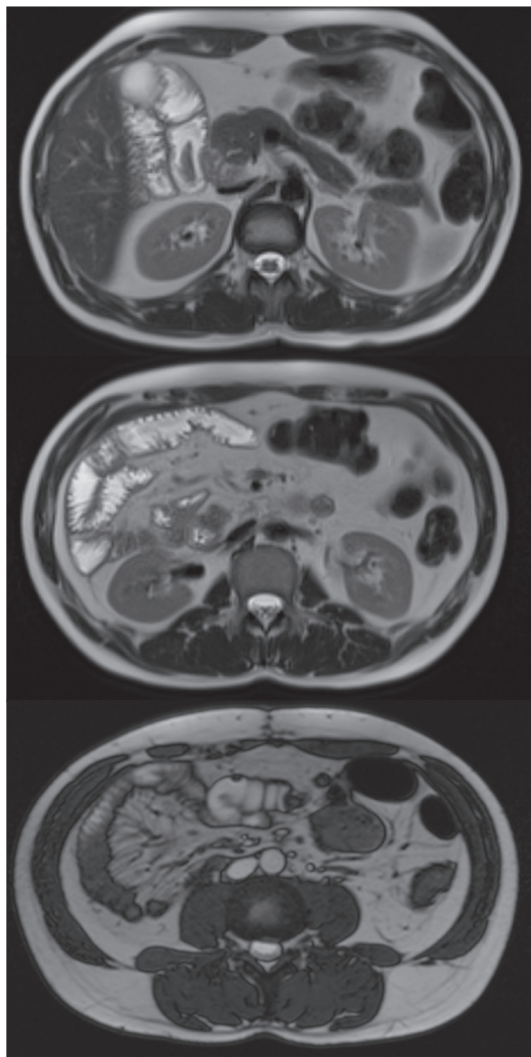


Figure 4 MRI small bowel study demonstrating the anatomy of partial malrotation. T2-HASTE (half-Fourier acquisition with single-shot turbo spin-echo) protocol used to acquire the top two images and T2-TRUFI (true fast imaging with steady-state free precession) was used to acquire the bottom image.

Assuming that the SARS-CoV-2 swab results that the patient produced are valid, we can narrow down the window of infection to 9 days prior to presenting to our emergency department. Given the lack of previous episodes and the fact that the vast majority of ectopic pancreas tissue are incidental findings in asymptomatic patients, this may be the first reported case of SARS-CoV-2 infection causing inflammatory changes in an ectopic pancreatic tissue.

Learning points

- ▶ SARS-CoV-2 infection could be associated with inflammatory changes in an ectopic pancreas.
- ▶ Ectopic pancreas can present with non-specific abdominal pain following inflammatory changes.
- ▶ Undiagnosed malrotation can be present in the adult population and may complicate assessment of the acute abdomen.

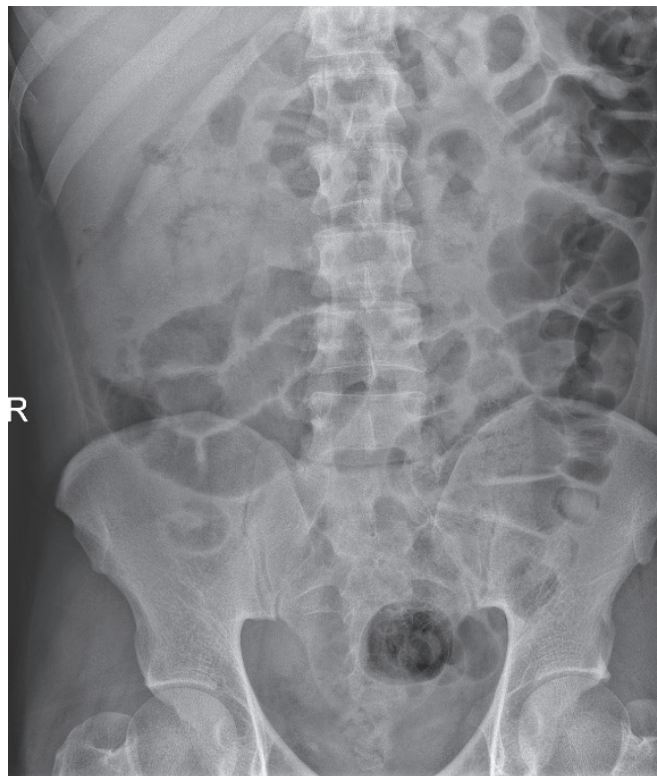


Figure 5 Abdominal radiograph taken on the same day of the MRI small bowel.

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ORCID iD

Woochan Hwang <http://orcid.org/0000-0002-9874-7772>

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