CASE REPORT

Atypical presentation of a spontaneous coeliac artery dissection resulting in jejunal ischaemia

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SUMMARY

We describe a case of a 59-year-old man with no significant medical history apart from a hiatus hernia and depression who presented with periumbilical pain which woke him at night. Before this he had 6 weeks of generalised abdominal pain. Blood tests were relatively normal and CT revealed some ill-defined stranding around the coeliac artery. He was diagnosed with a spontaneous coeliac artery dissection. Given the complexity of the case, a multidisciplinary team approach was adopted. He was managed conservatively and improved significantly over the next few days. Further investigations confirmed ischaemic changes to the distal duodenum and proximal jejunum. He has since been followed-up with CT scans and has had no further episodes 12 months from his initial admission.

BACKGROUND

Spontaneous dissection of the visceral arteries is relatively uncommon.¹ Coeliac artery dissection is very rare.² The coeliac artery supplies the proximal duodenum.³ To the best of our knowledge, this is the only case published, which involves coeliac artery dissection causing distal duodenal/proximal jejunum ischaemia.

In the documented literature, the main presentation for coeliac artery dissection is from upper abdominal pain to epigastric pain.⁴ There are associated risk factors involved such as hypertension and smoking, but the exact pathogenesis of the disease is not well known. There are a multitude of causes of periumbilical pain and this case could have mimicked appendicitis. The imaging in this case initially was not clear. These patients are difficult to manage and it is not clear what the best form of follow-up is after discharge.

We present a case of generalised abdominal pain that was present for 6 weeks which culminated in an acute presentation of periumbilical pain.

CASE PRESENTATION

We report a 59-year-old man who presented with a 2-day history of worsening periumbilical pain, sharp in nature, with no radiation that woke him from sleep. Before this, he had been experiencing generalised abdominal pain for 6 weeks. This was not associated with eating.

Medical history included a hiatus hernia and depression. There are no known drug allergies and he takes omeprazole and fluoxetine. He is a non-smoker and drinks minimal alcohol.

On examination his heart rate was 68 bpm, blood pressure 162/90 mm Hg, respiratory rate 18 and saturations 99% on air. He was generally tender around the periumbilical region and mildly tender suprapubically. Rectal examination was normal and no blood present on retraction.

INVESTIGATIONS**Blood tests**

Summarised in [table 1](#).

CT of the abdomen and pelvis with contrast (day of admission)

There was some ill-defined inflammatory stranding around the coeliac artery and the superior mesenteric artery ([figure 1](#)). This was causing mild narrowing of the origin of the common hepatic artery. The vessels were patent. A hiatus hernia was also present. Degenerative changes in the spine were noted at the L4/L5 level.

CT of the abdomen and pelvis with contrast (5 days postpresentation)

Track passes were made with the previous scan. There was now marked localised dilation and inflammation affecting the proximal small bowel from the level of the DJ flexure down to midjejunum ([figures 2 and 3](#)). The previously noted non-specific inflammation around the mesenteric vessels was no longer present. The mesenteric vessels appear patent. A small amount of peritoneal fluid was present. There were no features to suggest ischaemia and the appearances were unusual for infection or peptic acid disease.

Upper GI endoscopy (2 weeks postpresentation)

There was ulceration in the fourth part of the duodenum with scalloping, oedema and superficial ulceration for at least 15 cm. Histology showed this to be non-specific.

Capsule endoscopy (6 weeks postpresentation)

There was evidence of small bowel ulceration and a proximal stricture.

Balloon enteroscopy (2 months postpresentation)

A stricture was present at the fourth part of the duodenum/proximal jejunum. Ulceration and fissuring were also present. Histology was suggestive of ulceration/necrosis secondary to very focal ischaemia and likely of vascular origin. There was no

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Table 1

<i>Blood tests (on admission)</i>		
Haemoglobin	140 g/L	(131–166 g/L)
White cell count	$9.5 \times 10^9/L$	($3.5\text{--}9.5 \times 10^9/L$)
Platelets	$285 \times 10^9/L$	($150\text{--}400 \times 10^9/L$)
Mean cell volume	81.9 fL	(81.8–96.3 fL)
Neutrophils	$7.54 \times 10^9/L$	($1.7\text{--}6.5 \times 10^9/L$)
Sodium	140 mmol/L	(133–146 mmol/L)
Potassium	4.9 mmol/L	(3.5–5.3 mmol/L)
Urea	5.2 mmol/L	(2.5–7.8 mmol/L)
Creatinine	77 $\mu\text{mol/L}$	(62–106 $\mu\text{mol/L}$)
Albumin	46 g/L	(35–50 g/L)
Total bilirubin	7 $\mu\text{mol/L}$	(0–21 $\mu\text{mol/L}$)
Alkaline phosphatase	86 IU/L	(30–130 IU/L)
ALT	23 IU/L	(0–41 IU/L)
Amylase	53 IU/L	(28–100 IU/L)
Prothrombin time	11.1 s	(9.5–11.0 s)
APTT (s)	22.8 s	(19.9–28.0 s)
Fibrinogen	5.0 g/L	(2.0–4.0 g/L)
<i>Selected test</i>		
Enterocyte Ab: IgG		Negative
Anti-Goblet Cell Ab: IgG		Negative
Endomysium antibody: IgA		Negative
IgA t-Transglutaminase Ab		Negative
Antinuclear antibody-Hep2		Negative
Centromere antibody		Negative
Cytoplasmic-ANCA		Negative
Perinuclear-ANCA		Negative
Complement C3	1.67	(0.75–1.65 g/L)
Complement C4	0.28	(0.14–0.54 g/L)
HIV Ag/antibody screen		Not detected

Figure 1 CT coronal image showing (A) coeliac artery dissection.

evidence of a chronic inflammatory process, including inflammatory bowel disease.

CT of the abdomen and pelvis with contrast (2 months postpresentation)

Two months postpresentation several abnormalities were noted on CT of the abdomen and pelvis with contrast, such as extensive residual soft tissue around the root of the coeliac, marked narrowing of the origin of the coeliac with complete obstruction 2–3 cm beyond, and refilling from other vessels.

Figure 2 CT axial image showing (B) ischaemic distal duodenum/jejunum. **Figure 3** CT axial image showing (C) ischaemic distal duodenum/jejunum.

CT of the abdomen and pelvis with contrast (6 months postpresentation)

There was no interval change in appearances since the last scan. Aneurysmal distal coeliac artery with an inflammatory rind of tissue with luminal narrowing and possible dissection was seen. The hyperaemic segment of the proximal jejunum which was visible on the previous scan is no longer discernible.

Vascular MDT review (7 months postpresentation)

Within the main coeliac trunk, there is an area of dissection (with contrast filling of the true and false lumen), the total diameter being 12 mm (~2 cm long) (figure 4). This appears to compromise the true lumen supplying the common hepatic artery and splenic artery. The left gastric artery is seen filling, and arises from the affected segment. The gastroduodenal arcade is well developed, with clear communication from the superior mesenteric artery. The origin of the inferior

Figure 4 CT axial image showing (D) true lumen of the coeliac artery and (E) false lumen of the coeliac artery.

pancreaticoduodenal artery is complex, and there is possibly a dissection in this region also. There does not appear to have been significant progression between previous scans.

Vascular radiology team review of all images (13 months postpresentation)

From an imaging perspective, there is definite evidence of the distal duodenal and proximal jejunal ischaemia in all scans. The inflammatory stranding in the initial scans is markedly significant in the coeliac territory as opposed to the superior mesenteric artery. There is contrast in the wall of the vessel, a large segment followed by a small segment which represents the false and true lumen, respectively (figure 4). This could be due to atheromatous ulceration, but there is no evidence of atheromatous disease on the patients scans. We concur with a diagnosis of a coeliac artery dissection.

Medical team input

This is unlikely to be due to inflammatory bowel disease or a vasculitic picture, given results of our investigations.

DIFFERENTIAL DIAGNOSIS

- ▶ Appendicitis
- ▶ Cholecystitis
- ▶ Pancreatitis
- ▶ Vasculitis
- ▶ Atheromatous ulceration

TREATMENT

He was treated conservatively with fluids, pain relief and improved with time.

OUTCOME AND FOLLOW-UP

The patient was discharged from hospital 26 days following admission. He did very well and is currently undergoing routine follow-up and CT scanning and is under treatment with our vascular and hepatobiliary multidisciplinary team.

DISCUSSION

A review in 2007 identified 24 published cases of coeliac artery dissection, the presentation included 10 with abdominal pain, 6 with epigastric pain, 2 with upper abdominal pain, 1 with lower abdominal pain, 3 were asymptomatic and 2 classified as other.⁵ To date, our case is the only published case to report

periumbilical pain as the presenting symptom. Pain can be subjective and it is thought that patients who are asymptomatic can be explained due to having good collateral flow from the superior mesenteric artery.⁵

This patient developed ischaemia as evidenced by radiological, endoscopic and histological findings in the distal duodenum/proximal jejunum. This area is supplied by the superior mesenteric artery.³ To date, our case is the only published case to report these findings.

Most patients with visceral artery dissection present with acute or chronic abdominal pain.¹ Some of the causes of arterial dissection include atherosclerosis, trauma, iatrogenic, pregnancy, infection, polyarteritis nodosa and other collagenous associated diseases such as Marfan's disease or Ehler's Danlos syndrome.⁶

A recent systematic literature review performed by Cavalcante *et al*⁴ in 2016 found 60 publications over the course of 1987–2015 with 169 patients identified. Ninety-nine patients were men and 17 women. Hypertension and smoking were the associated conditions seen.⁴ The average age of presentation was 53.1 years.⁴ Abdominal pain was the most common presenting symptom, but there was no specific breakdown of which region the pain localised to.

The rise in cases identified has been thought to be due to the increased use of multidetector CT.^{7 8}

There is no consensus on treatment of these patients. The majority of patients of published cases are treated conservatively (~80%).⁴ The majority of patients do not receive specific medications, a proportion receive anticoagulants and antiplatelet therapy.⁴ There are surgical options, but these tend to be used when medical therapy has failed and include open surgery or an endovascular approach.⁹

How long to follow-up patients is unclear. Aneurysmal degeneration occurs at significantly varied rates with some published cases showing occurrences from 1 to 44 months postdissection.^{10 11}

A protocol by Galastris *et al*¹¹ suggested imaging and clinical evaluation at 30 days following the event, every 3 months for the first year and then annually following this time frame.

Most of the case reports we sourced did not indicate how often patients with coeliac artery dissections are scanned. We have discussed this patient in two of our multidisciplinary teams and have imaged him at regular intervals. He will continue to have follow-up. This case highlights the difficulties in diagnosing a spontaneous coeliac artery dissection but the importance of adopting a multidisciplinary approach in the management of these patients. While it is rare, new imaging techniques are allowing us to identify more of these patients and manage them appropriately.

Learning points

- ▶ Periumbilical pain can be a presentation of spontaneous coeliac dissection, but epigastric pain, upper abdominal pain and abdominal pain are more common in the literature.
- ▶ To date, spontaneous coeliac dissection causing proximal jejunal and fourth part duodenal ischaemia has not been described.
- ▶ More cases are being found and this could be due to multidetector CT.
- ▶ The majority of these patients can be treated conservatively and benefit from a multidisciplinary approach to their management.
- ▶ There is no consensus as to how often these patients should be reimaged following discharge.

Patient's perspective

The experience was a difficult one at the time. I knew something was wrong leading up to the day I came into hospital, but I did not think much of it. Everyone gets pain from time to time in the tummy. I had never experience pain like it and I was very worried. I was fully supported by the team that looked after me and the care I have gotten since leaving the hospital has been excellent.

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Contributors JG is responsible for preparing the manuscript, analysing the data, editing and proofing. HN is responsible for editing, concept development and proofing. AH is responsible for editing, concept development and proofing. AA-M is responsible for supervisor, editing, concept development and proofing.

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