CASE REPORT

Gastric necrosis and perforation in a patient with Asperger's syndrome

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SUMMARY

Acute gastric necrosis is a very rare but potentially fatal condition which has been reported in patients with abnormal eating behaviours. We describe the case of a 24-year-old female with a background of Asperger's syndrome, who presented with abdominal pain and gross distension. She underwent an emergency exploratory laparotomy and was found to have a massively distended, necrotic stomach. A total gastrectomy was performed with interval reconstruction planned. This case reports the surgical management of a rarely seen condition and highlights the importance of recognising gastric necrosis and its causes, which include patients with abnormal eating behaviours, the majority of whom are young females. This is the first report highlighting gastric necrosis in a patient with Asperger's syndrome and coincides with a growing recognition of the association between eating disorders and the autistic spectrum. It is also a rare example of patient survival following total gastric necrosis with perforation.

BACKGROUND

Gastric necrosis is very rare due to the abundant vasculature of the stomach. It has been reported that, in rare cases, abnormal eating behaviours such as overeating after a period of starvation and binge eating, can lead to massive gastric dilatation and necrosis.¹⁻³ These abnormal eating behaviours are associated with eating disorders such as anorexia and bulimia but are also seen in diagnoses such as Prader-Willi syndrome and autistic spectrum disorders.

There is a growing recognition of the association between eating disorders and the autistic spectrum.⁴⁻⁷ The incidence of eating disorders is also increasing and consequently there is a need for greater awareness of the potential surgical complications.

Gastric necrosis may occur in young patients and an 80%–100% mortality has been reported; therefore, early recognition and emergency surgical management are essential.^{8,9}

CASE PRESENTATION

A 24-year-old female, with a history of Asperger's syndrome was brought by ambulance to a district general hospital. She was confused with evidence of poor personal care and reported diffuse abdominal pain over the preceding day. The diagnosis of Asperger's syndrome was made in 2009 by her local adolescent mental health service based on

her significant difficulties with social situations, preoccupation with her bowel habit and an unusual interest in numbers. She had also previously been known to have a low weight and reportedly only ate on alternate days.

On examination, she appeared very thin and unkempt. Her weight was 43 kg and her height was 157 cm giving a body mass index of 17.4 kg/m². Her abdomen was hugely distended with generalised tenderness and no audible bowel sounds.

Her temperature was 37.1°C, blood pressure was 113/55 mm Hg, heart rate was 121/min and respiratory rate was 16.

INVESTIGATIONS

Blood results showed a creatinine of 146 µmol/L and urea of 10 mmol/L with a reduced estimated glomerular filtration rate at 38 mL/min. Other laboratory values were in normal range.

Venous blood gas findings included pH 7.03, lactate 11.2 mmol/L, base excess -12.4, bicarbonate 13.3 mmol/L and glucose 27.1 mmol/L.

Plain chest radiograph showed gross pneumoperitoneum (figure 1).

DIFFERENTIAL DIAGNOSIS

The initial preoperative diagnosis was a perforated hollow viscus, most likely a stercoral perforation of the colon or a perforated peptic ulcer.

TREATMENT

Aggressive initial fluid resuscitation and nasogastric decompression were instituted. A decision was made to proceed with emergency exploratory laparotomy, performed jointly by the general and upper gastrointestinal surgical teams.

At laparotomy, she was found to have a grossly dilated stomach to a volume of over 5 L (figure 2). The stomach was entirely necrotic apart from the pylorus, with palpable crepitus throughout, a large perforation of the posterior wall and frank contamination of the lesser sac with stomach contents. There was four-quadrant peritonitis, with gas evident in the epiploic appendages, the greater omentum and in the lesser omentum tracking towards the porta hepatis.

The arterial blood supply appeared intact, with normal pulsation of the major gastric vessels: right and left gastric, right and left gastroepiploic and short gastric. The small bowel and colon appeared normal.

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Figure 1 Presenting plain chest film showing gross pneumoperitoneum.

A total gastrectomy was performed with the stomach stapled distal to the lower oesophageal sphincter and just proximal to the pylorus using an Ethicon linear cutter TLC 75. Foley catheters were inserted into the oesophageal (12-French) and gastric antral (30-French) stumps. A feeding jejunostomy was inserted 60cm distal to the duodenal–jejunal flexure, as well as three drains in the perisplenic, periduodenal and subphrenic spaces.

OUTCOME AND FOLLOW-UP

Macroscopic examination of the resected specimen in the pathology department confirmed massive distension of the

Figure 2 Resected stomach showing dilatation and necrosis.

Figure 3 Resected stomach showing mucosal surface.

stomach with widespread serosal and mucosal necrosis and a posterior perforation (figure 3). Inside the stomach, there was a large quantity of minimally digested chicken. Histological examination revealed diffuse ischaemic changes of the gastric wall and full thickness necrosis consistent with an acute gastric infarction (figure 4).

The patient was managed on the intensive care unit postoperatively, with her recovery complicated by pneumonia, reintubation and a slow respiratory wean necessitating tracheostomy. Ventilatory and inotropic support were withdrawn as sepsis was controlled and haemodynamic parameters normalised, enabling step-down to ward care after 4 weeks.

The patient developed a leak from her gastric antral stump and required a period of total parenteral nutrition in addition to careful enteral feeding. She remained in hospital for a number of weeks during which plans were made for referral to a tertiary oesophagogastric centre for reconstruction.

DISCUSSION

The stomach enjoys a rich blood supply, derived principally from the three branches of the coeliac axis, with collateralisation of supply and contribution from the superior mesenteric circulation. Adequate arterial perfusion of the stomach has been demonstrated in cadavers with only one patent major artery¹⁰ and necrosis in animal models only achievable with occlusion of right and left gastric and gastroepiploic arteries as well as 80% of collateral vessels.¹¹ For this reason, gastric necrosis is a very

Figure 4 Histological gastric mucosa and submucosa necrosis.

rare pathology with references in the literature limited to case reports.

There are a few case reports describing different causes of gastric necrosis, including gastric volvulus,¹² caustic substance ingestion,¹³ acute necrotising gastritis,¹⁴ pancreatitis¹⁵ and vascular thrombosis.^{16 17}

Abnormal eating behaviours such as binge eating and starvation followed by overeating have also been known, in very rare cases, to cause necrosis of the stomach. There are a few case reports describing this in the context of anorexia,^{4 6 18 19} bulimia^{5 20 21} and Prader-Willi syndrome.^{22 23}

Asperger's syndrome, an autistic spectrum disorder, is a neurodevelopmental condition characterised by difficulties with social interaction and communication, restricted interests and repetitive behaviour. Eating disorders occur frequently and with a poorer prognosis in conjunction with Asperger's syndrome.^{1 2} Research also confirms that girls with anorexia nervosa have elevated autistic traits.³ We know that the patient in this case had a low body mass index and a history of eating only on alternate days. The presence of a large amount of undigested food in her stomach is in keeping with the findings of other patients with eating disorders and gastric necrosis.^{6 18} Her postoperative psychiatric evaluations revealed an abnormally selective and restrictive pattern of eating, as is often seen in autistic spectrum disorders.

The most widely recognised mechanism for acute gastric dilatation in patients with eating disorders is that the stomach becomes atonic and atrophied by periods of starvation. Following ingestion of a large meal, the weak stomach then overdistends. Intragastric pressure rises as the stomach distends; when this exceeds venous pressure, a decrease in venous outflow causes ischaemia.^{7 18 24} This mechanism of necrosis may start to occur at gastric volumes of 3 L and distension can increase to 5–9 L sometimes without perforation.^{6 7 21 25}

Plain chest and abdominal radiographs may assist with rapid diagnosis of gastric dilatation and pneumoperitoneum. CT may also be useful in identifying gastric distension and perforation but should not delay emergency surgery. In the context of surgical subspecialisation, these imaging results provide an indication for upper gastrointestinal surgical referral, as gastrectomy is mandated.

Initial treatment should include fluid resuscitation and nasogastric decompression, although the latter may be impossible in the presence of a large quantity of particulate matter. An inability to vomit has been reported in other cases of gastric necrosis in patients with eating disorders,^{7 25} further highlighting the need for early nasogastric decompression.

Urgent laparotomy, as was performed in the majority of cases in the literature, is essential if perforation is suspected. Operative management for gastric necrosis reported includes partial,⁶ subtotal²¹ and total gastrectomy.^{18 26} Conservative management is also reported in necrosis without perforation.⁷ In this case, the stomach was all necrotic apart from the pylorus, necessitating total gastrectomy. We would advocate against a one-stage procedure with primary anastomosis in the absence of favourable conditions, as this has been reported with poor outcomes.^{24 27 28} We would also advise use of a feeding jejunostomy, enabling adequate nutritional recovery before gastrointestinal reconstruction.

On initial assessment of young patients with abdominal pain and distension, it is important to take a thorough history of any abnormal eating behaviours, in particular if the patient looks malnourished, gastric necrosis should be considered. Following recovery, we suggest psychological evaluation to consider

potential underlying eating disorders which may also complicate recovery after gastrointestinal reconstruction.

In conclusion, we describe the first case of gastric necrosis in a patient with Asperger's syndrome and one of only a few cases of total gastric necrosis with survival of the patient. Patients on the autistic spectrum represent a category at increased risk of eating disorders. Early suspicion and appropriate expedient management will reduce mortality in this young group of patients.

Patient's perspective

'Before my admission I had not been eating normally since about 2013 after I started to live on my own. It was not because I wanted to be thin, although I know I did not want to be fat. Initially I started skipping mealtimes and I only ate one meal a day. Then I started only eating every other day. The longest I went without eating properly was about 8 days. I would eat nothing and then eat a whole loaf of bread for example. One day I ate too much in one go and I just kept eating even though my stomach hurt – I ate a lot of chicken and potato. I do not understand why this happened to my stomach when some people are able to eat so much and gain weight'.

Learning points

- ▶ Although rare, clinicians should have a high suspicion of gastric necrosis in patients with eating disorders. Early recognition and optimal management are essential as mortality from necrosis and perforation is very high.
- ▶ Patients on the autistic spectrum represent a category at increased risk of abnormal eating behaviours which can cause gastric dilatation and necrosis.
- ▶ A thorough history to elicit eating disorders should be taken during assessment as well as psychological evaluation during recovery.
- ▶ Gastrointestinal reconstruction following gastrectomy should be delayed in the context of sepsis and haemodynamic instability, where prolonged inotropic therapy and intensive care support is anticipated.

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