Rare disease

Spontaneous gastric perforation in an 11-year-old boy with anorexia nervosa: rare presentation with right iliac fossa pain

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Summary

Spontaneous gastric perforation is rare in children. It is usually associated with prematurity, foreign bodies and trauma. Anorexia nervosa (AN) can be an underlying condition although no cases are reported in the literature. We are reporting a rare case of 11-year-old boy with AN who presented with right iliac fossa (RIF) pain mimicking acute appendicitis. Intraoperative findings proved a gastric perforation. Pathophysiology of this condition in AN is poorly understood. Gastric smooth muscle atrophy and ischaemia can be the possible underlying mechanisms. The case discussed here highlights the fact that any abdominal pain in patients with underlying eating disorders (EDs) should raise suspicion of perforation as diagnosis can be far more complicated.

BACKGROUND

Gastric perforation is a potentially fatal condition that is rare in infants and children.¹ Most reported cases in the paediatric population are in neonates in association with prematurity, gastric volvulus, foreign body ingestion, trauma or iatrogenic injuries.^{2–6}

It has rarely been reported in association with psychiatric disorders, for example, Rapunzel syndrome^{7 8} (Trichophagia ie, eating hair). AN is one of the most serious psychiatric disorders found in children having the highest mortality of all psychiatric disorders at 5.6% per decade.⁹ It is frequently associated with medical complications such as osteoporosis, malnutrition, hepatic and endocrine abnormalities.¹⁰ Surgical complications are generally rare but are usually catastrophic. Acute surgical emergencies include acute gastric dilatation secondary to impaired gastric emptying or related to occasional very large meals, oesophageal perforation, gastric necrosis, superior mesenteric artery syndrome, multiple fractures and spontaneous gastric perforation.¹¹ Cases of gastric dilatation with associated necrosis are frequently reported in the adolescent group with AN.^{12 13} Spontaneous gastric perforation alone in children with AN is extremely rare.^{14 15} We report a case of spontaneous gastric perforation occurring in an 11-year-old boy with AN.

CASE PRESENTATION

An 11-year-old boy presented to the emergency department complaining right iliac fossa pain of 36 h duration. The child had been suffering from poor body image for last 6 months and had been treated for early onset AN in the community. His mother had previously been treated for bulimia nervosa and was currently asymptomatic.

On examination the child was 40 kg (50th percentile of his ideal body weight) and generally looking healthy. Routine observations revealed tachycardia and fever of 38°C. Abdominal examination revealed tenderness in the right iliac fossa region with guarding. Blood tests

showed a white cell count of 6.3 and C reactive protein (CRP) of 98.

INVESTIGATIONS

Blood tests showed a white cell count of 6.3 and CRP of 98. Urea and electrolytes, liver function tests and serum amylase levels were within the normal range.

TREATMENT

The overall clinical picture favoured the diagnosis of acute appendicitis. Appendicectomy was planned via a right iliac fossa incision. This revealed large amount of free gas and free purulent fluid in right iliac fossa and right paracolic gutter with a normal-looking appendix. The incision was extended transversely and inspection of the abdominal organs revealed prepyloric anterior wall gastric perforation. The perforation was repaired using vicryl sutures in two layers and peritoneal lavage was performed with saline and a drain was left in the subhepatic space. Serum *Helicobacter pylori* antibody turned out to be positive so the child was started on eradication therapy. The patient had an uneventful recovery and was discharged home on the fourth postoperative day.

OUTCOME AND FOLLOW-UP

On follow-up visit the wound was healing well but the child was still suffering from poor eating habits although the weight remained stable. Currently, the patient is being managed in community by joint paediatric and psychiatric teams for AN.

DISCUSSION

EDs are very common among children yet very little is known about the epidemiology of the condition. Subsyndromal presentations of AN, typically referred to as EDs not otherwise specified, have higher rates of occurrence but often are under-reported. According to a recent study in Canada, the incidence of early-onset restrictive

EDs in children aged 5–12 years seen by paediatricians was 2.6% (2.1–3.2) cases per 100 000 person-years.¹⁶ AN is usually diagnosed in adolescents and young adults. Patients with AN and bulimia nervosa, approximately 60% of whom will have gastric dysmotility, are at increased risk for acute gastric dilatation and subsequent possible perforation due to decreased gastric motility, increased gastric capacity and decreased gastric emptying.^{17 18} The pathophysiology of gastric dysfunction in AN is poorly understood. Possible mechanisms include gastrointestinal smooth muscle atrophy, diminished release of cholecystokinin, abnormalities in the autonomic nervous system and gastric rhythm abnormalities.¹⁷ As the intragastric pressure rises above 30 cm H₂O, a decrease in venous outflow may result in ischaemia and infarction of the gastric wall which can rupture.¹⁹ Spontaneous gastric perforation without gastric dilatation in relation to AN has not been reported in this age group before as far as authors are aware.

In our case, the presentation was atypical with right iliac fossa pain. Findings of free gas and clinically normal-looking appendix raised suspicion and a formal inspection of abdominal organs revealed the pathology which was a surprise. In typical presentations, these patients often deny history of ED and become unwell in a very short period with abdominal distension. Abdominal radiographs are very helpful in diagnosis. CT scan is usually diagnostic in doubtful cases. Early surgical intervention is the corner stone of management.

Learning points

- ▶ Aforementioned example highlights the fact that clinical presentation can be often misleading in children with eating disorders (EDs).
- ▶ Focus on history is vital in reaching an accurate diagnosis.
- ▶ History of ED in a child with acute abdomen should alert a careful clinician to the possible gastrointestinal perforation and should prompt appropriate investigations.

Competing interests None.

Patient consent Obtained.

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Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Hashmi KS, Ellul T, Leopard DC, Woodward A. Spontaneous gastric perforation in an 11-year-old boy with anorexia nervosa: rare presentation with right iliac fossa pain. *BMJ Case Reports* 2012;10.1136/bcr-2012-006512, Published XXX

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