CASE REPORT

Stomach infarction in an ex-premature infant

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SUMMARY

Gastric pneumatosis and thickened gastric wall are rare radiological findings that may be indicative of severe gastrointestinal tract ischaemia or necrosis; we report a case with a brief discussion of the literature. The premature neonate conveyed an interesting series of rare X-ray findings which were secondary to extensive gastric, duodenal and proximal jejunal infarction. She was managed palliatively and died.

BACKGROUND

Neonatal gastric pneumatosis is an extremely rare radiological finding and may be indicative of a severe gastrointestinal tract (GIT) ischaemia or necrosis. We report a neonate with an unusual presentation of a stomach infarction and an interesting abdominal X-ray series including markedly thickened stomach wall and gastric pneumatosis.

CASE PRESENTATION

A female infant was born to a 39 years old primigravida after an in vitro fertilization conception and an unremarkable family history. Pregnancy was complicated with early in utero twin demise but otherwise normal fetal anatomy scans, protective serology and a negative antibody screen. She presented at 24 weeks of gestation with per vaginal bleeding and premature rupture of membranes, received adequate antibiotics and β -methasone before having a spontaneous vaginal delivery at 24⁺⁵ weeks of gestation.

A female infant, birth weight 640 g (25th centile) head circumference 21 cm (25th centile) and length 32.5 cm (50th centile), was born with Apgar score of 5, 8 and 9 at 1, 5 and 10 min, respectively. She received delayed cord clamping and positive pressure ventilation followed by intubation. Umbilical venous (UVC) and umbilical arterial (UAC) catheters were placed in appropriate position. The UAC was removed 4 days later and the UVC was left in situ until the time of death.

She had a stable clinical course during her first week of life including early extubation into biphasic continuous positive airway pressure, a normal head ultrasound and was tolerating daily feeds increase of 16 mL/kg/day of exclusive mother's own milk with milk fortifier added at 9 days of age. At day 9, she had an acute deterioration with profound apnoea, marked respiratory acidosis, abdominal distension and hypotonia, requiring intubation, antibiotics and antifungal treatment in view of a suspicious cutaneous fungal rash.

INVESTIGATIONS

Blood cultures were subsequently negative and her coagulation profile was normal. Abdominal X-rays initially showed gaseous bowel distension with no evidence of pneumatosis or free air, but follow-up X-rays 24 h later showed a markedly thickened stomach wall (figure 1) followed by gastric and intestinal pneumatosis with evidence of free air in the abdomen tracking upwards to the mediastinum 48 h later (figure 2).

OUTCOME AND FOLLOW-UP

The patient was taken to the operating theatre on an urgent basis where a senior paediatric surgeon found frank necrosis of the entire stomach, duodenum and proximal jejunum with jejunal perforation.

After consultation with an additional senior surgeon, it was agreed that there was no surgical manoeuvre, in a premature infant <750 g that could salvage such extensive necrosis of the stomach and small bowel. A palliative plan was pursued and she died within 1 h in her parent's arms. An autopsy was declined.

Figure 1 Anterolateral view of an abdominal X-ray showing a thickened stomach wall and distended bowel loops.

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Figure 2 An anterolateral X-ray view of the chest and abdomen showing evidence of free intraperitoneal air and pneumomediastinum with dilated bowel loops and gastric intestinalis pneumatosis.

DISCUSSION

The X-ray finding of gastric wall thickening with progression to pneumatosis and then frank necrosis of the stomach found in the operating theatre are rare and unexpected findings. The underlying pathology of severe stomach necrosis may be related to either a variant of necrotising enterocolitis (NEC) or a vascular insult that can happen perinatally or postnatally.

Isolated gastric wall thickening by itself in a plain radiographic finding is not a commonly reported finding in neonates. Alternatively, gastric pneumatosis intestinalis has been described as a feature of fulminant NEC or a sign of other form of severe and widespread GIT involvement that may lead to intestinal perforation.¹ In 2003, Travadi *et al* reported that gastric pneumatosis is an extremely rare finding presenting in only 32 infants since 1966. He described it as either gastric emphysema or emphysematous gastritis. In gastric emphysema, there is an increased intragastric pressure that causes a mucosal breach and escape of gas such as in proximal intestinal obstructions. This is seen as linear streaks of intramural gas around the stomach in the abdominal X-ray. In emphysematous gastritis, there is ischaemia or inflammation and infectious agents that form intramural gas. This is seen as a cystic or bubbly appearance of gas in the abdominal X-ray such as seen in fulminant NEC, erosive gastritis or following cardiac surgery secondary to hypoperfusion.²

Interestingly, Johnson *et al* described a case in neonate with extensive infarction of the stomach wall and spleen which necessitated a gastrectomy with a Bilroth II repair and splenectomy. Yeast was present in the gastric vessels, the gastric wall and in the splenic sinusoids, and it was hypothesised that fungi gained access to the gastric vessels and intravascular hyphae caused enough obstruction, vessel wall damage, thrombosis and extensive gastric infarction.³ Similarly in our case, although we

cannot comment on evidence of a fungal invasion in the absence of a histological sample and a negative blood culture, a temporal association may be suggestive in view of the cutaneous yeast rash at the time of her medical deterioration for which intravenous fluconazole was given.

Alternatively, a perinatal event causing a vascular insult such as fetal thrombotic vasculopathy can be postulated. Interestingly, an association has been shown between acute chorioamnionitis, umbilical cord arteritis and fetal sepsis. This may result in a thrombogenic focus with thrombi that may embolise via the umbilical vein back to the fetus leading to brain or other visceral infarcts.⁴ Our patient's placenta was reviewed by a pathologist. The placenta showed evidence of acute chorioamnionitis with acute funisitis and chorionic vasculitis but no thrombi were found in the fetal or maternal vessels, although the possibility exists that areas with true thrombi formation were not sampled.

Postnatal vascular injury such as neonatal thromboembolism (TE) is another possibility. It has a strong association with the presence of central venous catheters as well as NEC, prematurity and other rare genetic thrombotic mutations. Around 30% of neonatal TE may be asymptomatic and symptoms depend on site of occlusion and end organ ischaemia.⁵ The calculated probability of developing aortic thrombosis with an UAC in situ is 16% for day 1, increasing to 32% by day 7 and 57% by 2 weeks. Our patient had the UAC for 4 days, a negative family history for thrombosis and a normal coagulation profile. However, as an autopsy was declined, we cannot exclude a TE that may have occluded the coeliac axis.

Learning points

- ▶ This is a case of a premature infant with gastric, duodenal and proximal jejunal infarction.
- ▶ Neonatal X-ray findings such as gastric pneumatosis and markedly thickened gastric wall are very rare.
- ▶ These X-ray signs may be indicative of a severe gastrointestinal tract ischaemia that may prompt an early surgical exploration.

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