Rare disease

Intractable positional borborygmi – an unusual cause diagnosed by barium contrast study

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Summary

The authors report the case of a 48-year-old woman, with a 2-year history of prominent borborygmi, nausea, abdominal discomfort after large meals and weight loss. Continuous, prominent, audible borborygmi were evident while the patient remained standing. However, these noises abated when she held her breath or when pressure was applied over the left hypochondrium. When lying flat, abdominal examination was normal. Gastroscopy, colonoscopy, small bowel follow-through, abdominal CT scan, small bowel transit study and laparoscopy were all normal. A barium meal showed that her stomach was normal when lying flat, but adopted an hourglass deformity in the sitting position due to compression from her left anterior ribs. Compression from the diaphragm, on inspiration, then resulted in audible borborygmi.

BACKGROUND

This unusual case highlights a dynamic, positional, gastric deformity, causing intractable and refractory borborygmi. This case is important, as it is the first report to describe such an anatomical anomaly. It also highlights the importance of considering physical abnormalities in patients with refractory borborygmi, where an incorrect diagnosis of dysmotility or functional bowel disorder may be made.

We are also keen to ascertain whether any other clinicians have encountered such a case. If so, we would welcome their advice because, thus far, no therapeutic measure has afforded any relief to our patient, who is experiencing considerable disability and distress.

CASE PRESENTATION

A 48-year-old woman presented with a 2-year history of borborygmi of gradual onset. These became overtly audible, leading to social embarrassment. Initially, she had nausea with occasional vomiting, especially after meals. She ate small meals, as she developed abdominal cramps after larger ones. She had lost 10 kg over the 2 years. She denied abdominal pain and was completely asymptomatic from borborygmi at night. On average, she opened her bowels twice per week. She had a history of recurrent venous thromboemboli, for which she was on life-long warfarin.

She had a past history of anorexia and depression, and had been under the care of clinical psychologists. There was no other significant past medical history. Her medications included warfarin, for recurrent venous thromboembolism; sertraline; carbamazepine; and zopiclone. She did not smoke and gave no significant alcohol history. There was a family history of diabetes mellitus. She previously worked as a drycleaner.

When lying flat, abdominal examination was normal. However, on standing up, easily audible borborygmi became

evident. These noises abated when she held her breath or when there was pressure over the left hypochondrium.

INVESTIGATIONS

Barium enema, abdominal CT scan, gastroscopy with duodenal biopsies, colonoscopy with colonic biopsies and small bowel follow-through were all normal. A small bowel transit study was normal, with a mouth to cecum time of 43 min. Laparoscopy was also normal.

A barium meal showed that her stomach was normal on lying down; however, in the sitting position it adopted an hourglass deformity due to compression from her left anterior ribs (figures 1–3). Compression from the diaphragm, on inspiration, then resulted in audible borborygmi.

DIFFERENTIAL DIAGNOSIS

She was initially thought to have intestinal dysmotility or a functional bowel disorder.

TREATMENT

As the borborygmi could be abolished by pressure over the left hypochondrium, a tight-fitting corset was suggested. Unfortunately, this was not successful in controlling her symptoms. A percutaneous endoscopic gastrostomy (PEG tube) was also discussed, as it was thought that this might decompress her stomach, by redirecting the flow of air and abolish the borborygmi. However, it was later felt that a PEG tube would not provide significant decompression. In addition, due to the patient's anticoagulation for recurrent thromboembolic disease, and significant psychological issues, surgical intervention has been deemed high risk.

OUTCOME AND FOLLOW-UP

Our patient continues to have troublesome, audible borborygmi as described, which in turn continues to cause social

Figure 1 (Patient erect): Note how her 'hourglass' stomach relates to her left anterior ribs (arrows), indenting her gas-filled stomach in the standing position.

embarrassment. At present, she also has symptoms suggestive of recurrent anorexia nervosa, with food avoidance and a perception of being overweight despite being markedly underweight. She remains under outpatient follow-up.

DISCUSSION

We have presented an unusual case of a patient with intractable and positional borborygmi, nausea and early satiety due to a gastric deformity in the upright position causing an hourglass stomach. On inspiration, air was forced through the deformity, causing audible borborygmi. These sounds stopped completely on breath-holding when the patient was in the upright position or when she was lying down. The prominence of the borborygmi, combined with the patient's distress and associated symptoms, resulted in numerous investigations that aimed to find an underlying cause. A number of initial investigations, including gastroscopy, abdominal CT scan and laparoscopy, failed to identify a cause. This may have been the case as the patient would have been supine for these tests and the gastric abnormality would not have been present.

Gastric deformities may be associated with audible borborygmi. However, case reports of this are sparse. In 1944, Lawrence reported a patient, with a symptomatic paraoesophageal

hernia, who had audible inspiratory borborygmi.¹ Borborygmi were synchronous with inspiration and were loudest on auscultation over the left lower chest and epigastrium. They ceased when the patient held her breath or sat up.

The main cause of an hourglass deformity is scarring of the mid-stomach from chronic peptic ulcer disease. Other rarer causes are gastric syphilis,² caustic ingestion,³ tuberculosis⁴ and failed anti-reflux surgery.⁵ These defects, however, tend to be fixed and not reversible by changing position.

This case also poses a number of management difficulties. Although surgery was initially considered, this was felt to be potentially hazardous, given the patient's recurrent thromboemboli and long-term anticoagulation and psychological issues. In addition, because no abnormality had been seen on laparoscopy, surgery may not have been successful. As it was noted that the sounds were reduced by pressure in the left hypochondrium, a tight-fitting corset was suggested to the patient so as to apply pressure in that area. This suggestion was not effective, although there were questions about compliance with this treatment option.

It is unclear why our patient's borborygmi only began 2 years ago, given that the problem would seem to be

Figure 2 (Patient erect): Note how slim the gastric body is (double arrow) in the erect symptomatic position. The two single arrows depict the position of her left anterior ribs.

anatomical. An interesting hypothesis is that significant weight loss due to anorexia nervosa, resulting in loss of abdominal wall and visceral fat, in her case, resulted in close apposition of the gastric wall and ribcage in the standing position. Weight gain, with increased 'cushioning' between the stomach and ribcage due to abdominal wall and visceral fat, might therefore alleviate the problem. Clearly this will require vigorous management of her psychological issues through counselling and support and continuing dietetic input.

This case would seem to be the first report of a positional gastric hourglass deformity causing intractable borborygmi. The cause was eventually determined, by barium-contrast

study, to be due to compression of the stomach by the left anterior ribs in the upright position.

Learning points

- Consider gastric anatomical abnormalities in patients with recurrent borborygmi, especially if repeated investigations fail to identify a cause.
- Borborygmi that are related to respiration are likely to be caused by diaphragmatic compression.

Figure 3 (Patient lying supine): note the gastric indentation is now absent; the metal clip marking the left anterior ribs has shifted in position vis-à-vis the stomach. The width of the gastric body in the lying position is markedly increased (double arrow), accounting for the lack of borborygmi.

Competing interests None.

Patient consent Obtained.

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Sharma A, Moriarty K, Burnett H, Paraoan M, Thompson D. Intractable positional borborygmi – an unusual cause diagnosed by barium contrast study. *BMJ Case Reports* 2010;10.1136/bcr.01.2010.2637, date of publication

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