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Recurrent spontaneous pneumoperitoneum: A surgical dilemma

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ABSTRACT

INTRODUCTION: Spontaneous pneumoperitoneum describes free air within the peritoneal cavity in the absence of iatrogenic causes or a perforated viscus. This report describes a rare case in which despite raised inflammatory markers, a trial of conservative management proved adequate.
CASE REPORT: AM is a 36-year old woman who presented multiple times with abdominal pain and radiologically proven pneumoperitoneum. Her medical history included self catheterisation and cerebrovascular stenosis (Moyamoya disease), asymptomatic gallstones, livedo reticularis and peptic ulceration. On her index admission she exhibited raised inflammatory markers and fever. Despite these findings, emboldened by similar presentations in the past with no cause found, she declined surgical intervention with no untoward consequences.
DISCUSSION: Most patients presenting with non-surgically induced pneumpoeritoneum display signs of peritonism including pyrexia and raised inflammatory markers. For such patients, surgical intervention is usually required to find and rectify the cause.
CONCLUSION: A trial of conservative management may be appropriate in patients with spontaneous pneumoperitoneum but it must always be borne in mind that clinical deterioration represents an indication for surgical intervention.

1. Introduction

Pneumoperitoneum is defined as an abnormal state characterised by the presence of gas (as air) in the peritoneal cavity [1]. This is a radiological diagnosis, usually made on erect chest radiography or computerized tomography (CT) scan. In 90% of cases it is either caused by perforated viscus or iatrogenic causes such as open abdominal surgery, laparoscopy [2] or following trauma. The remaining 10% of pneumoperitoneum are termed 'spontaneous'. Spontaneous pneumoperitoneum may be due to pneumatosis cystoides intestinalis (the most common abdominal cause) and rupture of lung bullae in patients with patent pleuroperitoneal canal. In females, pneumoperitoneum may result from retrograde migration of air via the genital tract [3].

This report describes an unusual case of a young woman with a known history of spontaneous pneumoperitoneum, who presented with a new episode associated with raised inflammatory markers but no identifiable causes of pneumoperitoneum.

2. Case report

AM, a 36-year old lady presented to the acute surgical team with diffuse abdominal pain, fever and a soft non tender abdomen, accompanied by raised white cell count (WCC) of 19.3×10^9/L, a C-reactive protein (CRP) of 103 mg/L. Her past medical history included intermittent self-catheterisation for idiopathic urinary retention; cerebro-vascular stenosis (Moyamoya disease); asymptomatic gallstones in the gallbladder; livedo reticularis and peptic ulceration demonstrated on oesophagogastroduodenoscopy.

Over an 11-year period, AM had 13 admissions to hospitals in the UK with the same type of abdominal pain. On each of these occasions her inflammatory markers and other blood tests were within normal limits and either an erect chest X-ray or an abdominal CT scan (Fig. 2) showed free air in the intraperitoneal space. Each time, careful monitoring and conservative management resulted in rapid relief of her pain and repeat imaging demonstrated a reduction in the volume of free air. She would usually be discharged after 2–5 days, completely symptom free with follow up imaging arranged to monitor her pneumoperitoneum. The exception to this pattern was one occasion in 2010 when her pain failed to settle with conservative measures. She underwent diagnostic laparoscopy which demonstrated no evidence of visceral perforation, nor any other pathological process that could explain her pneumoperitoneum. Further details of some of her previous hospital admissions are given below.

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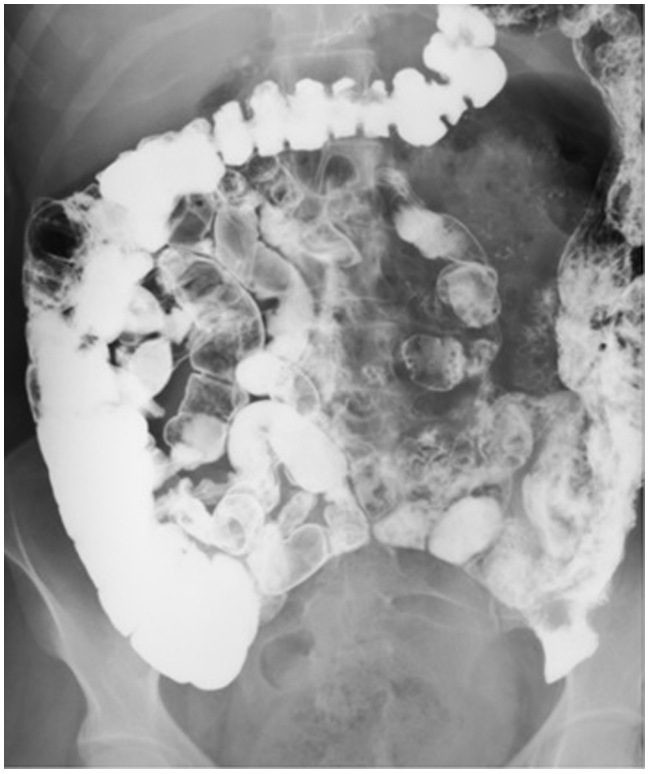

Fig. 1. Barium Small Bowel Meal and Follow Through showing normal small and large bowel.

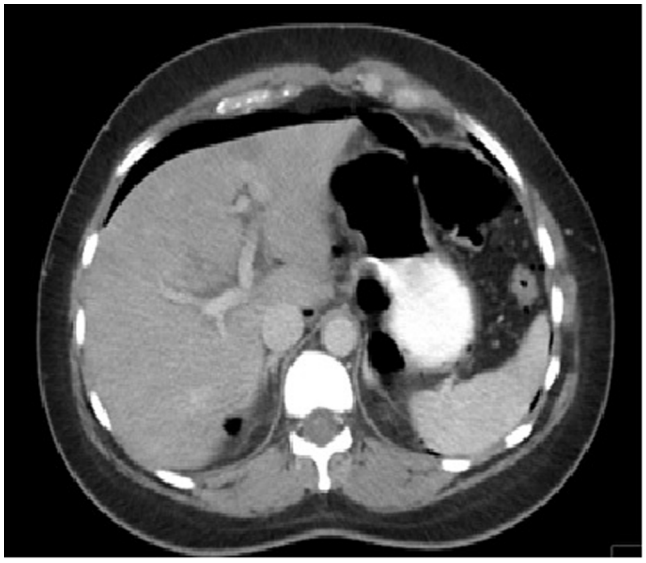

Fig. 2. Axial view of CT scan showing free intraperitoneal gas anterior to the liver and stomach.

AM's first admission with this problem was to a hospital in Wales in May 2003. She complained of epigastric pain and was found to have free air under the diaphragm on an erect chest x-ray. Her symptoms were mild enough to justify a trial of conservative treatment and she made a rapid recovery. A detailed account of this admission is unfortunately not available. However, oesophagoduedenoscopy performed during follow up was reported as showing a sealed duodenal ulcer. She was started on Omeprazole then and has remained on this long term.

She was first admitted to our hospital in 2004 with shoulder tip pain and mild upper abdominal tenderness, with CXR showing free air under the diaphragm. Her blood tests demonstrated a WCC of 12.1×10^9/L but no other abnormalities. She underwent a gastrografin swallow which demonstrated no leak of contrast and she was managed conservatively. She was discharged eight days later with normal blood tests and complete resolution of her symptoms.

In March 2006 she had a similar admission, primarily with shoulder tip pain associated with left sided upper abdominal pain. She had slightly raised WCC on admission and an erect chest x-ray showed free gas under the diaphragm. At this time a nasogastric tube was inserted and antibiotics started. Close monitoring demonstrated rapid clinical improvement and she was discharged seven days later.

In July 2006 she presented with the same symptoms and had an admission lasting 5 days. Again she had normal blood tests and physiology, but suffered mild epigastric pain and had evidence of air under the diaphragm on chest x-ray. Post discharge barium small bowel meal and follow through was performed which showed no abnormality (Fig. 1).

Before the last (index) admission, AM had come into our hospital on eight further occasions, with a similar clinical picture. She had undergone multiple investigations (Table 1) including a diagnostic laparoscopy (March 2010) which found some adhesions around the terminal ileum and mid-small bowel. In spite of such extensive investigation, no specific cause for her pneumoperitoneum has been identified.

During her index admission, an erect chest X-ray demonstrated free intraperitoneal air. Two differential diagnoses were considered; perforated viscus or a recurrence of her previous pneumoperitoneum, for which no cause had been found. After discussion, AM declined an exploratory laparotomy or diagnostic laparoscopy citing her previous multiple similar episodes treated successfully without surgical intervention. Conservative measures (empirical antibiotics and fluids) resulted in a rapid reduction in the level of inflammatory markers and volume of air seen under the diaphragm. She was discharged home with a repeat CT scan organised for follow up. She was reviewed at the outpatient clinic thereafter where a decision was made to intervene in the future, only if a demonstrable pathological entity was found.

3. Discussion

In many cases of spontaneous pneumoperitoneum, associated clinical or laboratory features would provide a lead towards the underlying aetiological factors (Mularksi et al.) [3]. Spontaneous pneumoperitoneum has been recognised following invasive ventilation, particularly in cases requiring high ventilatory pressures, in patients with obstructive pulmonary disease and also in pneumothorax. It has also been recognised following cardiopulmonary rescucitation [4]. In Gynaecological practice, cases of pneumoperitonium have been reported in pelvic inflammatory disease, following sexual or orogenital intercourse, with vaginal douching and after certain exercises in the postpartum period [5].

There is no established link between pneumoperitoneum and Moyamoya disease, a rare progressive cerebrovascular disease caused by blockage of arteries at the base of the brain. Pneumotosis cystoides intestinalis is a benign condition characterised by gas filled cysts in the intestinal wall, most commonly in the terminal ileum [3]. Rupture of these cysts can result in radiologically detectable pneumoperitonium which is usually best treated by watchful waiting [6]. However, in immunosuppressed patients, caution should be exercised with such management approach.

AM has had recurrent episodes which have largely taken the same course. She did not have any relevant gynaecological or intrathoracic history to point towards this type of cause and repeated imaging have shown no signs of peumatosis cystoides

Table 1
Summary of AM's imaging over an 11 year period.

Investigation	Date	Findings	Histology
Oesophago-Gastro- Duodenoscopy (OGD)	Dec 2004	Normal	Mild congestion antral mucosa
Enteroscopy	July 2005	Mild oesophagitis. Duodenum normal.	Chronic inflammation on gastric mucosa
Barium Small Bowel Meal & Follow Through	Aug 2006	Normal	N/A
OGD	Aug 2006	Scarring second part of duodenum.	Chronic inflammation.
Colonoscopy	Aug 2006	Normal	Not applicable (N/A)
Diagnostic Laparoscopy	March 2008	Adhesions around D1 and D2 and also around the terminal ileum	N/A
Capsule endoscopy	July 2008	Normal	N/A
CT Abdomen	March 2010	Free inter-peritoneal gas. No focus of perforation	N/A
OGD	April 2011	Atrophic Duodenum, generalized gastropathy	NAD
CT Abdomen	April 2015	Free inter-peritoneal gas. No focus of perforation	N/A
Ultrasound Abdomen	July 2015	Multiple Gallstones. No active cholecystitis	N/A

intestinalsis. This case raised an unusual management dilemma. Based on the raised inflammatory markers, fever and abdominal pain it was reasonable to consider perforation of viscus as a possible cause, and to advise surgical intervention. However, given her past history of several similar admissions successfully treated with conservative measures and her strong determination to avoid surgery, a trial of conservative management was not unreasonable.

4. Conclusion

Most patients presenting with non-surgically induced pneumpoeritoneum display signs of peritonism including pyrexia and raised inflammatory markers. For such patients, surgical intervention is usually required to find and rectify the cause. In cases where pneumoperitoneum is proven, but the other clinical features are lacking, a trial of conservative management may be appropriate, as in this case, particularly as the patient has had similar presentations in the past with no cause found. It must always be borne in mind that clinical deterioration represents an indication for surgical intervention.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contributions

PE – Drafted the manuscript & made the corrections.
JAA – Conceptualised the report, critically appraised and modified the manuscript.

Conflict of interest statement

None.

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