Pneumatosis portalis and pneumatosis intestinalis following aortic valve replacement: a case report

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ABSTRACT

Background: Pneumatosis portalis (PP) and pneumatosis intestinalis, when present together, are rare but often ominous radiological findings on imaging of the abdomen, frequently indicating severe intra-abdominal disease such as intestinal ischaemia. We present a case of PP and intestinalis following cardiac surgery without evidence of significant intra-abdominal disease.

Case presentation: A 62-year-old male underwent open bioprosthetic aortic valve replacement for severe aortic stenosis. Four days post-operatively, the patient developed severe abdominal pain and distension. Contrast computed tomography of the abdomen demonstrated a significant degree of gas within the portal venous system and within the intestinal wall. At exploratory laparotomy, there was no evidence of significant bowel ischaemia or other intra-abdominal pathology, a finding confirmed after re-look laparotomy 48 hours later. The patient was eventually discharged with no further sequelae noted.

Conclusion: This case demonstrates the often pre-terminal finding of PP and intestinalis, seen in a patient without evidence of any significant abdominal disease. We report a previously undescribed case of PP and intestinalis following open cardiac surgery. An exploratory laparotomy should be performed if there is any clinical concern, given the potential mortality associated with these findings.

Keywords: Pneumatosis portalis, pneumatosis intestinalis, portal venous gas, aortic valve surgery, case report.

Type of Article: CASE REPORT **Specialty:** General Surgery

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Background

Pneumatosis portalis (PP) – the presence of air within the portal venous system – is often recognised as an ominous radiological finding indicating severe intra-abdominal disease. The mechanisms underlying its development are not fully understood [1], but PP is being diagnosed more frequently in non-life-threatening conditions with the advent of computed tomography (CT) imaging [2,3]. Identifying those patients requiring surgical management is paramount, given the significant associated mortality. PP and pneumatosis intestinalis (PI) have been observed in both benign and pathological contexts, and in some cases without an identifiable cause [4].

Initially described in infants with necrotising enterocolitis, PP (or portal venous gas) has been observed largely in patients with bowel necrosis, with over 40% of a review of 182 patients exhibiting PP in the context of established bowel necrosis [5] and was associated with a 75% mortality in these cases. Interestingly, PP was demonstrated in a number of other intra-abdominal pathologies, but these were not associated with the same increased mortality as bowel necrosis was. Since the initial descriptions associating PP with catastrophic abdominal pathology, its reported incidence has increased with increased availability of CT imaging, and a number of cases are seen in asymptomatic or otherwise well patients [1,2].

Given the observation of PP and PI in both well and unwell patients, discerning benign from pathological cases is paramount but can pose a challenge to the assessing surgeon. There is no consensus decision on when or in whom to perform exploratory laparotomy, and management strategies employed at the time of surgery also differ across the literature. We present a complex case of a patient presenting with PP and PI following cardiothoracic surgery with a favourable post-operative outcome.

Case Report

A 62-year-old male with a background of quiescent Crohn's disease and previous ileocaecal resection underwent an open bioprosthetic aortic valve replacement for

severe aortic stenosis as an elective procedure. This was performed with routine cardiopulmonary bypass and 32°C cardioplegia. An attempt at right superior pulmonary vein vent insertion was unsuccessful, but otherwise, no particular difficulties were noted intra-operatively, and the system was successfully de-aired with an aortic root vent.

While recovering on the ward, the patient was referred to General Surgery for abdominal distension and diffuse tenderness. Serum lactate at initial presentation was 3.0 mmol/l (reference range 0.5-2.2mmol/l). CT imaging was performed, revealing large volumes of gas within the portal venous system and throughout the liver, as well as generalised small bowel dilatation with PI. Areas of the small bowel also demonstrated poor mural enhancement (Figure 1).

The patient was taken to the theatre for laparotomy and abdominal exploration. Pre-operative National Emergency Laparotomy Audit mortality was estimated at 42%. The only concerning finding was a dusky-appearing segment of mid-jejunum approximately 125 cm from the duodenojejunal flexure. Despite this, strong mesenteric vessel pulsations were palpable, and there was no evidence of full-thickness bowel necrosis. Given the substantial morbidity associated with creating a stoma this proximally, the decision was taken to perform laparostomy and a planned re-look 48 hours after initial laparotomy.

At the re-look procedure, the concerning area of bowel was healthy, well perfused, and seen to undergo peristalsis. No evidence of ongoing ischaemia or bowel necrosis was evident, and no resection was deemed necessary. The remainder of the abdominal organs and gastrointestinal (GI) tract all appeared healthy. The abdomen was closed without further complication, and the patient returned to the critical care unit, where he was extubated the following day. Parenteral nutrition was commenced for a period of time, and the patient remained stable.

Echocardiography was performed once the patient returned to the ward, which demonstrated normal ventricular function and an appropriately sited aortic valve prosthesis with normal Doppler values. Importantly, no mural thrombus was seen. A postoperative CT scan demonstrated complete resolution of both portal venous gas and PI (Figure 2). The patient was discharged without event.

Discussion

PI can be observed in one of two patterns, cystoides and linearis, with PI cystoides more likely to be seen in benign aetiology [3]. Theories regarding the pathophysiology of PI include bacterial invasion of the bowel wall and dissection of bowel gas into the bowel wall due to increased intraluminal pressure [6,7]. On the other hand, PP is less likely to be benign and, when observed in conjunction with PI, is associated with the presence of mesenteric infarction and bowel necrosis [8].

To the best of our knowledge, this is the only reported case of concurrent PP and PI following open cardiac surgery. One related study in the literature, a 9-year study observing patients presenting with PI following lung transplant [9]. In this series, 15 patients (2%) of the study population had PI following transplant, in which patients underwent cardiopulmonary bypass. Of these patients, only one had PP, and none of these patients had the combination of PP & PI that we observed in our patient. The authors noted that 3 of the 15 patients in their study underwent bowel resection for confirmed bowel ischaemia at exploratory laparotomy.

Our case is particularly noteworthy given the discordance between the degree of PP & PI observed in this patient, and the lack of any significant ongoing bowel ischaemia at initial laparotomy. The degree of portal venous gas on CT was striking, with widespread gas visualised within the portal system, although the liver appeared normal on both inspection and palpation during the initial laparotomy.

The patient in our case report was a known case of Crohn's disease and had undergone a previous ileocaecal resection many years prior for terminal ileal Crohn's disease. At the time of presentation, however, the patient's Crohn's disease had been in remission for a number of years, and the patient was not on any regular medication. As an inflammatory bowel pathology, Crohn's disease can cause acute abdominal pathology associated with pneumatosis, as was reported in a study of 50 Crohn's patients undergoing CT [10]. However, this study was only able to demonstrate a significant relationship between corticosteroid use and the presence of pneumatosis. No significant relationship was demonstrated between disease severity and the presence of pneumatosis, however. The presence of pneumatosis has been observed in both active, symptomatic Crohn's and also in the asymptomatic patient [11,12]. This highlights the need to assess patients and symptom severity on a case-by-case basis, rather than simply treating the finding of pneumatosis alone.

This case also highlights the complex decision making that must be employed in such a situation, given the stark difference between imaging and intra-operative findings. The combination of PP & PI is associated with completed bowel ischaemia & necrosis in up to 70% of patients [13], which does not fit with our intra-operative findings of largely healthy, viable bowel. The decision on whether or not to resect the suspicious segment of bowel is a complex one, with both patient and technical factors needing consideration. In this case, bowel resection and stoma formation were felt to be unnecessary given the lack of convincing bowel infarction as well as the morbidity associated with the formation of a stoma so proximally in the GI tract.

It is unclear if the portal venous gas was introduced as a result of open cardiac surgery despite the system being vented intra-operatively. Portosystemic anastomoses are

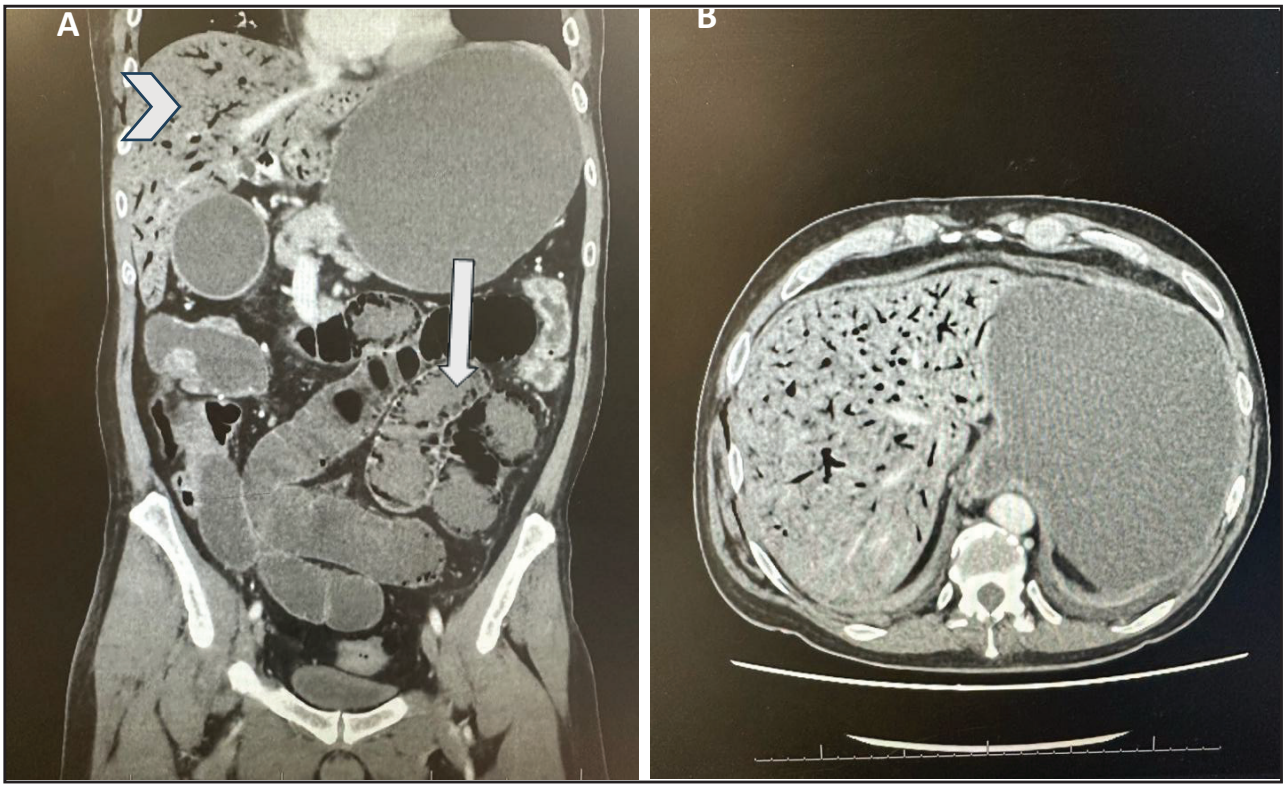

Figure 1. *A: Both pneumatosis portalis (arrowhead) & intestinalis (arrow) visible on coronal pre-operative CT. B: Axial pre-operative CT demonstrating widespread pneumatosis portalis*

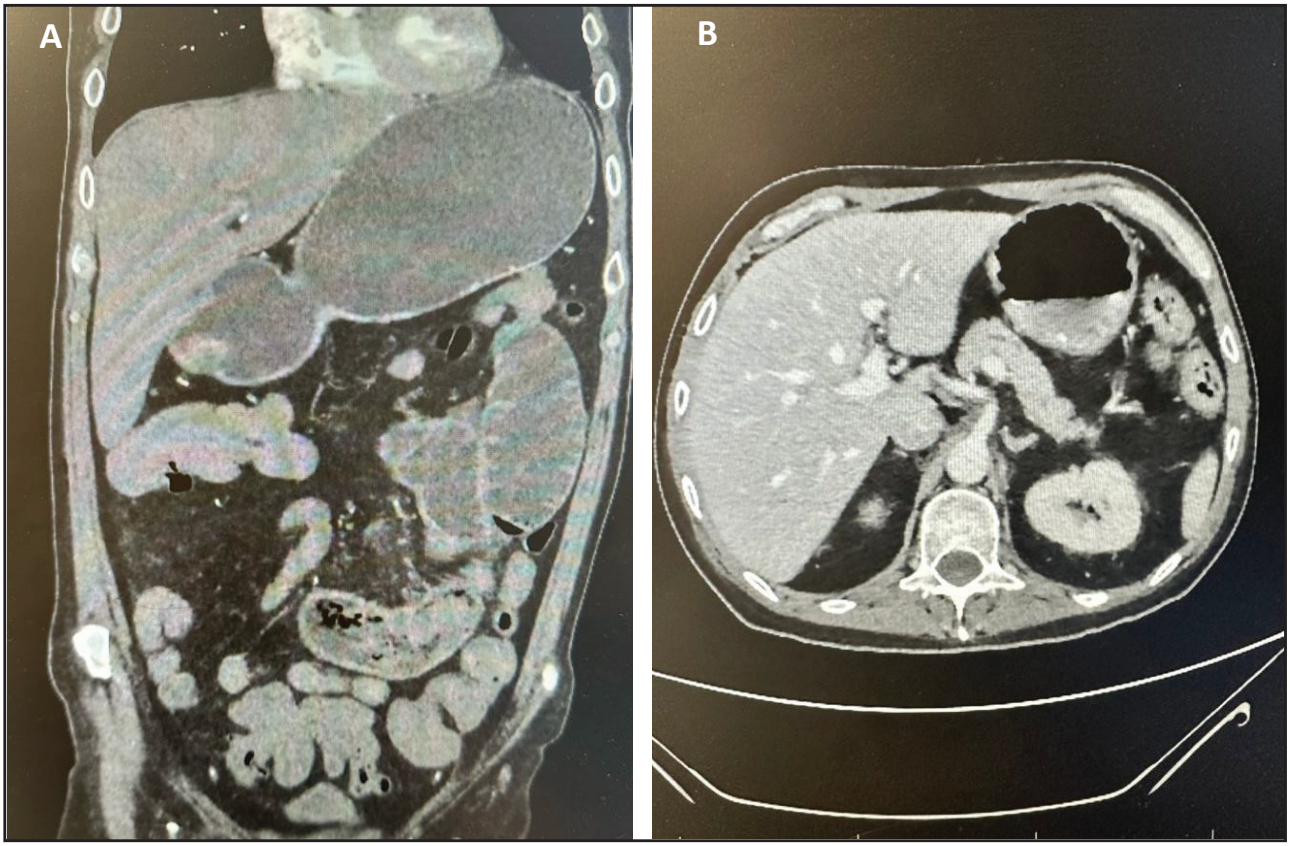

Figure 2. *A: Post-operative resolution of both pneumatosis portalis and intestinalis. B: Axial post-operative CT image demonstrating resolution of pneumatosis portalis.*

well-described, although those in the mediastinum are often overlooked, including anastomoses between the left gastric and paraoesophageal veins. These potentially may provide a route for air to enter the portal circulation if inadvertently introduced intra-operatively, although this is unclear and difficult to prove.

It is possible that the patient had a brief post-operative ileus following initial cardiac surgery, causing a degree of abdominal distension which, combined with the CT findings, may have mimicked a presentation of ischaemic bowel. However, the clinical picture of hypotension, ongoing abdominal pain, and distension in conjunction with the CT findings led to exploratory laparotomy. Exploratory laparotomy has been historically recommended once portal venous gas has been identified, in order to assess the situation and suitability for surgical resection, or whether attempts at this would be futile [14].

In summary, we present a case of PI and PP following cardiac surgery that did not appear to be associated with any significant bowel ischaemia. The recency of open cardiac surgery suggests possible shunting of air into the portal system. Despite the increasing incidence of benign diagnoses of PI and PP, clinical assessment by the surgeon should prevail when considering surgery for such patients. Diagnostic laparoscopy or exploratory laparotomy should be considered if there is any clinical concern regarding the patient, given the potentially high associated mortality with concomitant PP & PI.

Conclusion

Surgeons should be aware of the potential gravity of imaging findings of combined PP and intestinalis, as they are often associated with significant bowel ischaemia and thus with a high mortality. For this reason, surgical intervention is advised if there is any diagnostic uncertainty. We report, to the best of our knowledge, the only described case of PP and intestinalis following open cardiac surgery.

What is new

PP and intestinalis are most commonly seen in conjunction following severe bowel ischaemia and often is a terminal finding on imaging. We present a case of PP and intestinalis following open cardiac surgery. Early laparostomy and re-look laparotomy did not identify any significant bowel ischaemia. The surgeon must be aware of the potentially fatal combination of pneumatoses and laparotomy, considering early to facilitate positive outcomes.

Acknowledgment

We thank the patient for providing consent for the write-up of this case report.

List of Abbreviations

CT	Computed Tomography
NELA	National Emergency Laparotomy Audit
PI	Pneumatosis intestinalis
PP	Pneumatosis portalis

Conflict of interests

The authors declare that there is no conflict of interest regarding the publication of this article.

Funding

None.

Consent for publication

Written informed consent was obtained from the patient.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of the case

1	Patient (gender, age)	62 years, male
2	Final diagnosis	PP & PI following aortic valve surgery
3	Symptoms	Abdominal pain and hyperlactataemia
4	Medications	Symptomatic treatment given
5	Clinical procedure	Emergency exploratory laparostomy followed by re-look laparotomy at 48 hours
6	Specialty	General Surgery