Pneumorrhachis, Subcutaneous Emphysema, Pneumomediastinum, Pneumopericardium, and Pneumoretroperitoneum After Proctocolectomy for Ulcerative Colitis

Report of a Case

Luther H. Holton, M.D., John Migaly, M.D., Rolando H. Rolandelli, M.D.

From the Department of Surgery, Temple University Hospital, Philadelphia, Pennsylvania

This article presents the first known case of pneumorrhachis (spinal air), pneumomediastinum, pneumopericardium, pneumoretroperitoneum, and subcutaneous emphysema after proctocolectomy for ulcerative colitis. We review the patient's medical history, clinical and laboratory findings, radiographic data, and operative records, as well as the relevant literature. We describe the case of a young male with ulcerative colitis who developed pneumorrhachis, subcutaneous emphysema, pneumoretroperitoneum, pneumomediastinum, and pneumopericardium after a proctocolectomy with ileal pouch-anal anastomosis. Unlike the case we report, previously described episodes of pneumomediastinum and subcutaneous emphysema in patients with ulcerative colitis developed before operative intervention. We offer possible explanations for these unusual complications based on analysis of this case and thorough review of the literature. [Key words: Pneumorrhachis; Subcutaneous emphysema; Restorative proctocolectomy; Ulcerative colitis; Inflammatory bowel disease]

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REPORT OF A CASE

R.W. is a 20-year-old male who was diagnosed with a two-year history of ulcerative colitis in December 1996 by colonoscopy and who required two hospital admissions and multiple blood transfusions for anemia. After maximal medical therapy consisting of azathioprine, cyclosporine, mesalamine, and prednisone failed, he was referred for colectomy. Before his scheduled operative date, he was admitted to an outlying hospital with another flare of colitis. Intravenous hydrocortisone was begun, and he was transferred to our institution. Over the ensuing three days, he achieved remission of symptoms. Serial abdominal films revealed no evidence of megacolon. After adequate bowel preparation, he was taken to the operating room, where he underwent a proctocolectomy with an ileal pouch-anal anastomosis and a diverting ileostomy under general anesthesia without the use of an epidural or spinal catheter.

There were no abscesses or collections found during the procedure. His postoperative course was uncomplicated, and he was discharged to home on postoperative Day 5 on a steroid taper, analgesics, and a proton pump inhibitor.

Six days after discharge, he returned for a scheduled postoperative visit. At that time, he complained of shortness of breath, tinnitus, chills, fever, headache, nausea, neck pain, and low back pain. On physical examination, he was afebrile (98.9°F), tachycardic (114 beats per minute), tachypneic (26 breaths per minute), and normotensive (126/82 mmHg). He was found to have a midline trachea, no jugular venous distention, and mild nuchal rigidity. His lungs were clear to auscultation, and cardiac examination demonstrated no murmurs, rubs, gallops, or crunch. His abdomen was soft, nontender, and nondistended, with active bowel sounds. His ileostomy was noted to

Address reprint requests to Dr. Rolandelli: Department of General Surgery, Temple University Hospital, Broad and Ontario Streets, 4th Floor-Parkinson Pavilion, Philadelphia, Pennsylvania 19140.

be pink, with stool and air in the ostomy bag. His neurologic examination revealed no motor or sensory deficits. Because of his shortness of breath, tachycardia, tachypnea, and recent postoperative status, he was immediately readmitted to the hospital to rule out pulmonary embolism.

An arterial blood gas was obtained (pH 7.41, $PaCO_2$ 34 mmHg, PaO_2 148 mmHg, HCO_3 21 and SaO_2 98 percent, gradient 3). His white blood cell count was 12.2 K/mm^3, and hemoglobin was 12.5 g/dl. Chest film revealed subcutaneous emphysema in the neck (predominantly on the right side) and thorax, as well as pneumomediastinum and pneumopericardium (Fig. 1). No pneumothorax or lung disease was evident on chest x-ray. Ventilation/perfusion scan was interpreted as low probability for pulmonary embolism. Because of the unexpected results of the chest film, a computed tomographic (CT) scan of the chest, abdomen, and pelvis was ordered that revealed subcutaneous emphysema of the right neck, with air tracking through nontissue planes, suggestive of myonecrosis of the right sternocleidomastoid muscle (Fig. 2). Additionally, the CT scan demonstrated pneumomediastinum, air within the spinal canal, and a small pocket of retroperitoneal air (Fig. 3). Given the grossly abnormal radiologic findings and the patient's immunosuppressed status, there was concern about infection caused by a gas-forming organism. The patient was started empirically on vancomycin, meropenem, and ampicillin.

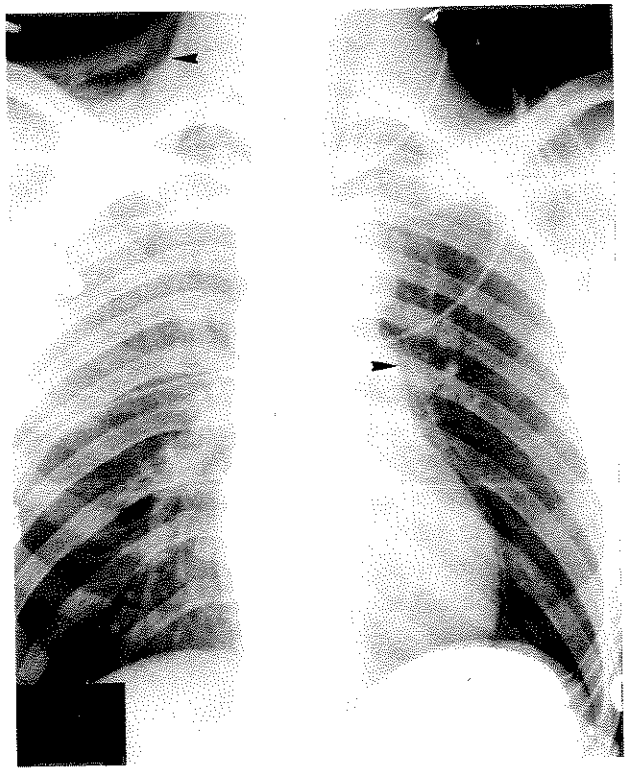

Figure 1. Chest film revealed subcutaneous emphysema in the neck and thorax, as well as pneumomediastinum and pneumopericardium (arrowheads).

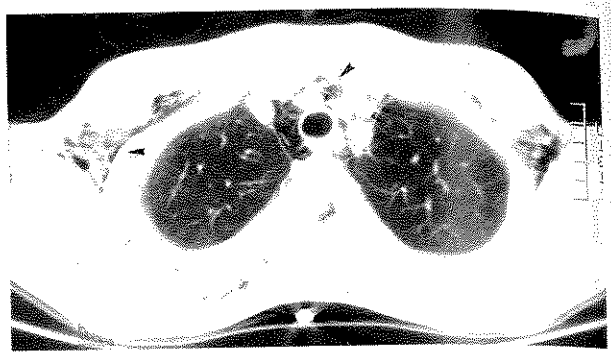

Figure 2. Computed tomographic scan of the chest, abdomen, and pelvis revealed subcutaneous emphysema of the right neck, with air tracking through nontissue planes, suggestive of myonecrosis of the right sternocleidomastoid muscle. Upper arrowhead points to air in the upper mediastinum. Arrowhead at left points to air in the anterior chest wall toward the neck area.

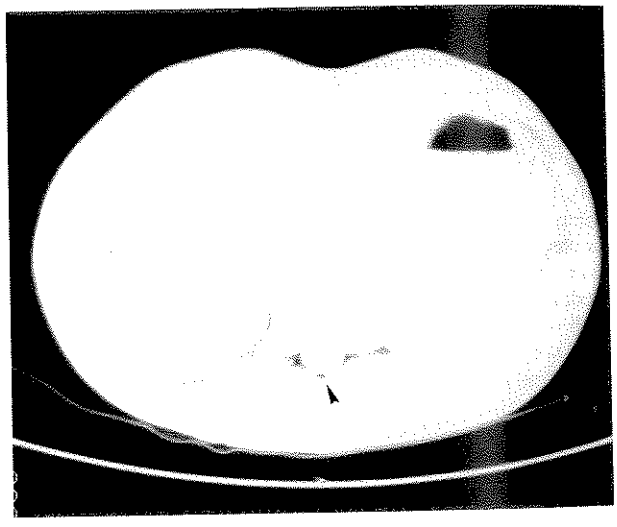

Figure 3. Computed tomographic scan also demonstrated pneumomediastinum, air within the spinal canal, and a small pocket of retroperitoneal air. Arrowhead points to a bubble of air in the spinal canal, indicating pneumorrhachis.

After extensive discussion with consultants from the head-and-neck, cardiothoracic, and infectious disease services, the patient was taken to the operating room for a neck exploration and endoscopic evaluation of the ileal pouch. The concern was that the presumed infection by a gas-forming organism could have been caused by a central line placed in the right

neck or by ischemic necrosis of the ileal pouch. Endoscopy revealed a healthy anastomosis and pouch without evidence of necrosis. Exploration and débridement of the right neck demonstrated no evidence of an infectious or inflammatory process. Irrigation fluid was sent for culture, and a portion of the sternocleidomastoid muscle was also taken for culture and biopsy.

Postoperatively, a barium swallow was obtained to rule out an esophageal perforation; it was found to be negative. By postoperative Day 3, all cultures remained negative, and antibiotics were discontinued. A repeat CT scan of the thorax revealed that the pneumomediastinum, subcutaneous emphysema, and spinal air were resolving. By the fourth postoperative day, the patient was tolerating a regular diet and was asymptomatic. He was discharged to home without a known cause of his presenting findings. Eight weeks later, his ileostomy was reversed, and he did well after that.

DISCUSSION

The unusual development of extraneous air collections within the body has been described in a wide variety of disease states. They have typically been associated with such processes as epidural abscesses, vacuum phenomenon in degenerative disk disease, osteonecrosis, vertebral fractures, epidural anesthesia, and rarely, pneumomediastinum. There have been cases of epidural air attributed to more obscure processes, such as pelvic trauma, and a single case in which a patient with Crohn's disease developed an enterothecal fistula.[1] Four cases of pneumomediastinum in ulcerative colitis have been reported in the literature, but none were associated with pneumorrhachis or occurred in the postoperative period.[2–4]

To best evaluate the cause of our patient's unusual cluster of findings, an understanding of potential pathways of gas travel is essential. It is well established that gas can track between various body compartments, such as the retroperitoneum and the mediastinum.[5–8] Once in the mediastinum, gas can readily communicate with the fascial planes of the neck, the pericardial space, and even the epidural space.

Our patient had no complaints or symptoms related to this unusual process before he underwent proctocolectomy. Nevertheless, in four previously reported cases, pneumomediastinum in patients with ulcerative colitis was attributed to microperforations in the colon. These reports theorized that gas migrated from the colonic lumen into the retroperitoneum and then ascended into the chest. Recently, Fledman and Lustberg,[5] using three-dimensional reconstructed CT scans of the chest and abdomen, demonstrated this phenomenon in a patient with a perforated diverticulum. Although our patient had a significant flare of his disease, his colon was found to be without transmural ulcers, granulomas, or fistulas, all of which were noted in previous case reports.

The operative procedure itself could explain the introduction of air into these various body cavities. Although our patient did not receive epidural or spinal anesthesia, both of which have been associated with pneumorrhachis, he was exposed to positive-pressure ventilation, a process that can cause pneumothorax and pneumomediastinum. Air can also be introduced during dissection of tissues or as the result of an anastomotic leak. Bonardi *et al.*[9] reported a case of subcutaneous emphysema, pneumoretroperitoneum, and pneumomediastinum after rectal surgery. Their group credited the establishment of a communication between the rectum and the pelvirectal space secondary to dissection of the sphincter muscles. Similarly, Mirzayan *et al.*[10] reported a case of subcutaneous emphysema, pneumomediastinum, pneumothorax, pneumopericardium, and pneumoperitoneum in a patient who had rectal barotrauma when a large bolus of air was forcibly insufflated directly into his rectum from a compressor hose. Their group proposed that air entering a rectal mucosal defect could then dissect between the fascia of the internal/external anal sphincter and the conjoined longitudinal muscle to the fascial plane, consisting of the superior fascia of the pelvic diaphragm and the levator ani muscle, before finally entering the supralevator space and the retroperitoneum. In our patient, an anastomotic leak was an unlikely cause of these air collections, because he had a diverting ileostomy that minimized the amount of air in the pouch, he did not have any clinical manifestations of an anastomotic leak, and the CT scan did not show any radiographic signs of a leak, not even any air around the anastomosis. When the clinical presentation is suggestive of a possible anastomotic leak, the study of choice is an absorbable contrast enema of the pouch.

In addition to explanations for the introduction of air into the retroperitoneum, it has been theorized that air can be introduced into the pelvic vasculature di-

rectly, thereby leading to epidural air collections. Chimon and Cantos[11] proposed that spinal epidural air could be secondary to pelvic trauma that breaches the ascending lumbar veins, which anastomose with the intervertebral veins on their course from the common iliacs to the azygos and hemiazygos veins.[11] Although it is possible that these vessels were dissected or traumatized in the course of our patient's procedure, it is unlikely that such a mechanism could explain the other large collections of air throughout his body.

The idea of infection mediated by a gas-forming organism was entertained but discounted in light of negative cultures and the noninfected appearance of his tissues on surgical exploration. Additionally, a postoperative pneumomediastinum would have been possible, especially given our patient's tall, thin body habitus, his exposure to steroids, positive-pressure ventilation, and the possible existence of subclinical lung pathology related to inflammatory bowel disease. However, had the genesis of these extraneous air collections been from the lung, one would have expected an exacerbation while he was ventilated for his exploratory procedures. Clinical improvement continued despite reintubation and ventilation.

CONCLUSIONS

The findings of pneumorrhachis, pneumomediastinum, and pneumopericardium, as described above, may represent a serious postoperative complication, such as introduction of air during dissection, or other such diverse processes, such as microperforations and bleb disease, which cannot be dismissed without an in-depth investigation. This case represents another unexpected development in a patient with ulcerative colitis, a disease for which both the medical and surgical therapies are still in evolution.

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