Subcutaneous emphysema of the neck after shunt surgery for hydrocephalus in a case of metastatic ovarian cancer: illustrative case

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BACKGROUND A 46-year-old female, a patient with a relapsed carcinoma in her ovary, had undergone ventriculoperitoneal (VP) shunt surgery for obstructive hydrocephalus due to vermian metastasis. Two weeks after the shunt surgery, she complained of discomfort in the neck. There was subcutaneous emphysema along the shunt track without tenderness or signs of inflammation. She was afebrile, and her vital parameters were stable.

OBSERVATIONS The authors ruled out pneumothorax and airway trauma as potential sources of emphysema. They tapped the shunt chamber and detected gram-negative bacilli. Ascitic fluid culture grew gas-forming *Escherichia coli*.

LESSONS Although some amount of air can get trapped in the subcutaneous plane during the tunneling procedure of a VP shunt tube insertion, the reappearance of a new, large column of air along the shunt track can be an ominous sign of shunt infection. The shunt became contaminated by bacteria of gut origin, which seeded the ascitic fluid, and a florid bacterial growth ascended up the shunt track, producing gas along the subcutaneous plane. Physicians should consider this rare etiology in their differential diagnoses of subcutaneous emphysema following VP shunt surgery.

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KEYWORDS neck emphysema; shunt surgery; gas-producing organisms

Subcutaneous emphysema is the pathological presence of air in the skin. It usually results from surgical, traumatic, infectious, and spontaneous etiologies. The small amount of air that gets trapped during a tunneling procedure usually is absorbed within a few days. When emphysema is seen after the 1st week of surgery, or if it increases dramatically, physicians should consider other serious causes. We report the case of a patient with metastatic ovarian cancer who developed delayed shunt track emphysema and had a downhill clinical course.

Illustrative Case

A 46-year-old female with a known case of relapsed carcinoma in her ovary presented with features of raised intracranial tension. She had headaches and vomiting for a duration of 1 week. On examination, she was well oriented, and her central nervous system examination findings were within normal limits. She was started on medical decompression with mannitol and dexamethasone. Magnetic resonance imaging of the brain revealed a well-defined enhancing lesion in the roof of the 4th ventricle causing obstructive hydrocephalus. Although the

lesion was solitary and large, we could not excise the lesion due to the coronavirus disease 2019 (COVID-19)-related disruption of elective operation schedules. Surgical removal of the posterior fossa tumor would have required an operation in our usual elective operating room. During the intense transmission phase of COVID-19, all our elective operating room slots were cancelled, intensive care unit beds were limited, and the usual neurosurgical wards were closed. This affected our ability to do any surgery more complex than a simple shunt surgery. Instead, we performed an emergency right ventriculoperitoneal (VP) shunt surgery.

We placed the abdominal end of the shunt intraperitoneally using an open technique, under vision, through a small subcostal incision. The abdominal part of the surgery was done after tapping the ventricle at the parietooccipital burr hole and connecting it to the rest of the shunt in the tunnel from the scalp incision to the abdominal end. Although the surgeon encountered dense adhesions, she did not notice any overt injury to the bowel at the time of surgery.

The immediate postoperative period was uneventful. We started the patient on palliative whole-brain radiotherapy (30 Gy/10 fractions).

ABBREVIATIONS COVID-19 = coronavirus disease 2019; CT = computed tomography; VP = ventriculoperitoneal.

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FIG. 1. Plain radiograph posteroanterior view of the neck and thorax, showing subcutaneous emphysema on the right side of neck (arrows) along the track of the VP shunt.

FIG. 2. Non-contrast CT thorax coronal section, showing air in the subcutaneous plane along the VP shunt in the neck. No airway or lung injury was noted.

After 8 fractions of radiation, in the 2nd postoperative week, she complained of discomfort on the right side of the neck. There was an ill-defined swelling and crepitus on the neck and chest. She had no signs of meningeal irritation. She was afebrile, her pulse rate was 106/min, her respiratory rate was 16/min, her peripheral capillary oxygen saturation was 98% in room air, and her blood pressure was 106/72 mm Hg. Abdominal and systemic examination findings were unremarkable.

Radiograph and ultrasound examinations of the neck showed subcutaneous emphysema in the track of the shunt tube (Fig. 1). A computed tomography (CT) scan of the neck, thorax, and abdomen ruled out pneumothorax, pneumomediastinum, and trauma to the lung (Figs. 2 and 3). She had no tracheobronchial or laryngeal injury. The air track was confined to the neck, chest, and abdomen along the track of the VP shunt (Fig. 4). Progression of the malignancy was evident in the form of multiple peritoneal, liver, and spleen deposits, and there were metastatic lymph nodes in the abdomen and in the left supraclavicular station. There was mild ascites, and a mass was seen infiltrating the rectum. The next day, she had one episode of high-grade fever. We tapped the shunt chamber, and the mildly turbid cerebrospinal fluid showed gram-negative bacilli. We started her on empirical broad-spectrum antibiotics (vancomycin, amikacin, and ceftazidime) and removed the shunt.

On day 3, she had worsening abdominal distension and constipation. There was localized guarding and tenderness over the abdominal shunt incision. Bowel sounds were sluggish.

An ultrasound of the abdomen revealed dilated bowel loops with sluggish peristalsis, and there was mild ascites. A plain radiograph of the abdomen showed multiple air-fluid levels suggestive of intestinal

obstruction (Fig. 4). The radiograph did not show gas under the diaphragm. We inserted a nasogastric tube for decompression of the stomach and advised the patient to have no oral intake and kept her on intravenous maintenance fluids in view of suspected intestinal obstruction. The serum procalcitonin level (16.81 ng/mL) was suggestive of sepsis. The serum lactate level was 1.53 mmol/L. The blood culture was sterile. Ascitic fluid cultures were sent to the microbiology laboratory. We substituted ceftazidime with meropenem for broader coverage.

On day 4, she developed headache and neck rigidity and became drowsy and disoriented the next day. She also developed urinary incontinence. A CT scan of the brain showed pneumoventricle, but there was no ventriculomegaly. She had a high respiratory rate of 30 breaths per minute. Arterial blood gas analysis revealed respiratory alkalosis with high anion gap metabolic acidosis, and the lactate level was 1.53 mmol/L, which is indicative of sepsis. Abdominal girth, tenderness, and guarding increased.

Ascitic fluid culture sensitivity revealed *Escherichia coli* sensitive to amikacin and resistant to ceftazidime, cefoperazone-sulbactam, ceftriaxone, meropenem, and piperacillin/tazobactam. Vancomycin and amikacin were continued, and colistin was added at a loading dose of 9 million units and a maintenance dose of 4 million units twice a day. It was concluded that bacterial peritonitis by gas-producing *E. coli* resulted in ascending VP shunt meningitis and subcutaneous emphysema along the shunt track.

On day 7, there was discharge of fecal matter through a dehiscence in the abdominal incision site. She continued to deteriorate, and her relatives took her home by request. She died the next day.

FIG. 3. Non-contrast CT neck axial section, showing air in the subcutaneous plane along the VP shunt in the right side of the neck.

FIG. 4. Plain radiograph of the abdomen erect shows multiple air fluid levels and dilated bowel loops, suggestive of intestinal obstruction, and air along the VP shunt track in the abdomen (as indicated by arrows), indicative of an ascending infection along the shunt to the neck, presenting as subcutaneous emphysema and further upwards to the brain, resulting in meningitis.

Discussion

Observations

Although some amount of air can get trapped in the subcutaneous plane during the tunneling procedure of VP shunt tube insertion, the reappearance of a new, large column of air along the shunt track proved to be an ominous sign of shunt infection. We believe that the shunt hardware became contaminated by bacteria of gut origin, probably from a microperforation, and it seeded the ascitic fluid, and a florid bacterial growth ascended up the shunt track, producing gas on the way.

The total absence of free gas in the abdomen on the CT scan early in the disease process gave the impression that the shunt surgery was the source of the bacterial infection of the shunt track. The large doses of dexamethasone that she was receiving probably blunted the inflammatory response that usually follows shunt track infection, and hence, there was little tenderness or redness on the neck, chest, and the abdomen. The preterminal event of fecal material extruding from the abdominal incision site indicated that a perforation had indeed occurred, probably at the site of the mass growing into the rectal wall. It is very unlikely that the soft silicone tube could penetrate the intact gut, although that has been described in the pediatric population in which there are a few reports of shunt tubes spontaneously extruding through the anus due to slow erosion after many months to years.

Lessons

Although some amount of air could get trapped while the tunneling of the shunt is done during surgery, its presence 2 weeks later is ominous. If CT scans of the neck and thorax do not reveal an airway injury or lung injury as the source of this air, then the possibility of infection by a gas-producing organism should be suspected. Lactate-

fermenting organisms capable of producing gas include *E. coli*, *Klebsiella pneumoniae*, *Proteus*, *Candida*, and *Clostridium*.¹ High-dose steroids mask the usual signs of inflammation of the shunt track as well as the systemic symptoms of infection.

Prompt identification and treatment of the site of infection and initiating appropriate antibiotics early in the course of the disease are of paramount importance in the treatment of gram-negative bacteremia and sepsis.² Ideally, antibiotic therapy should be started within 1 hour of diagnosis because each hour delay in the administration of antimicrobials is associated with a measurable increase in mortality.³ We faced a diagnostic dilemma in ascertaining the cause of emphysema. Starting antimicrobial therapy on the 1st day may have favorably altered the course of the disease.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Kumari, Sathish, Prem. Acquisition of data: Kumari, Sathish, Prem. Analysis and interpretation of data: all authors.

Drafting the article: Kumari, Sathish, Prem. Critically revising the article: all authors. Reviewed submitted version of manuscript: Kumari, Sathish, Prem. Approved the final version of the manuscript on behalf of all authors: Kumari. Administrative/technical/material support: Prem. Study supervision: Kumari.

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