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Getting the wind knocked out of him – Report of a broncho-cutaneous fistula caused by staphylococcal pneumonia, and review of literature

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Abstract

A broncho-cutaneous fistula (BCF) refers to the formation of an abnormal fistulous connection between the tracheobronchial tree and the cutaneous surface of skin. A rare occurrence in and of itself, the disease entity may have varied etiologies, and may or may not be associated with a broncho-pleural fistula. We describe a case of a young patient who developed a BCF as a complication of a necrotizing pneumonic process, and his subsequent clinical course. In so doing, we review the clinical features of this peculiar disease entity, analyzing the available medical literature similarities in etiology and variations in management strategies described in the literature thus far.

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Introduction

An aberrant connection (fistula) between bronchi and subcutaneous tissue planes is termed a broncho-cutaneous fistula (BCF), and maybe created as a result of a variety of causes or circumstances. When this connection also includes the pleura, it is termed "broncho-pleural fistula". In the medical specialties, some causes that have been implicated in its formation are - ionizing radiation to the area, necrotizing pneumonia, placement of cardiac pacemaker wires, mechanical ventilation and as a consequence of chest tube placement. Thoracic surgeons and other surgical specialties may encounter the entity more frequently, through patient with trauma or as a complication of surgical procedures such as lobectomy. Herein, we describe a young immunocompromised patient who contracted a necrotizing form of staphylococcal pneumonia that resulted in the formation of a broncho-cutaneous fistula, and his subsequent clinical course. The formation of a BCF is rare occurrence, especially as a complication of staphylococcal pneumonia, and consequently, the index of suspicion for it among practicing clinicians is likely low. Prompt diagnosis and management may lead to considerably less morbidity and patient discomfort.

Case Report

A 43-year-old male presented with fever, malaise and generalized weakness. His history was notable for active intravenous drug use (IVDU), uncontrolled Human Immuno-Deficiency Virus (HIV) infection, diabetes mellitus type 2 and multiple abscesses at prior injection sites. Physical examination was notable for tachycardia 114/min and fever of 101.7 Fahrenheit (38.7 degrees Celsius), with an erythematous left arm, and a normal systemic examination of the cardiac, respiratory and other systems. Laboratory investigations yielded the presence of methicillin sensitive *Staphylococcus aureus* (MSSA) in blood, on two separate blood cultures. An antibiogram of the isolated organism demonstrated sensitivity to oxacillin, cefazolin and most other antibi-

otics, except penicillin. The patient was admitted for intravenous antibiotic therapy with cefazolin 2 grams every 8 hours, for treatment of MSSA bacteremia, with the source likely being cellulitis of the left arm. Over the next week, the patient's symptoms responded to therapy, and bacteremia resolved. An echocardiogram did not demonstrate signs of endocarditis, but a new symptom of left sided chest pain, swelling and crepitus emerged. Imaging of the area (Figure 1) revealed a new 3cm gas and fluid containing collection in the left pectoralis major muscle. This collection was seen in continuity with a 5 cm thick walled cavitary mass of the anterior segment of the left upper lobe, which was a new finding, in comparison to his chest film from a few days ago. The two collections were seen in continuity across the first left costochondral articulation. A phlegmon in the base of the neck and the anterior mediastinum with fluid and gas extending to these areas was also suspected. Due to the suspicion of necrotizing soft tissue infection in this high-risk patient, local surgical wound exploration was performed. The muscle planes and tissues were found to be viable, and scant purulent fluid was evacuated, which grew the same organism with similar antibiogram sensitivities to antibiotics. Unfortunately, in the immediate post-operative period, the patient developed worsening subcutaneous emphysema (Figure 2) extending superiorly through the neck into his face, ending in his eyelids and inferiorly, into his abdomen, ending in the scrotal area.

Due to the extent and rapidity of spread, an intercostal percutaneous pig tail catheter was placed into the cavitary lung lesion (Figure 2C), as it was felt to be the source of the worsening subcutaneous emphysema. The pig tail was attached to low intermittent suction of -20 cm H_2O and a wound vacuum (VAC) dressing was applied to the surgical incision in order to facilitate resolution of subcutaneous emphysema and pneumomediastinum. The patient also received oxygen supplementation to facilitate nitrogen washout and resorption of extraneous air. Over the next 10 days, the patient experienced clinical and radiological resolution of his symptoms (Figure 3). The chest tube and wound VAC dressing was removed, and the patient was discharged to complete a course of long-term antibiotic therapy, with good result (Figure 3).

Discussion

A BCF is an abnormal fistulous connection between the tracheobronchial tree and the subcutaneous tissue planes and may or may not be associated with a broncho-pleural fistula. Infrequent as it may be, many precipitating causes of BCF have been reported in the literature, such as necrotizing bacterial pneumonia, fungal pneumonia, ionizing Radiation therapy, trauma, mechanical ventilation, as a complication of chest tube placement, pacemaker lead placement and post- surgical lobectomy. The spontaneous contigu-

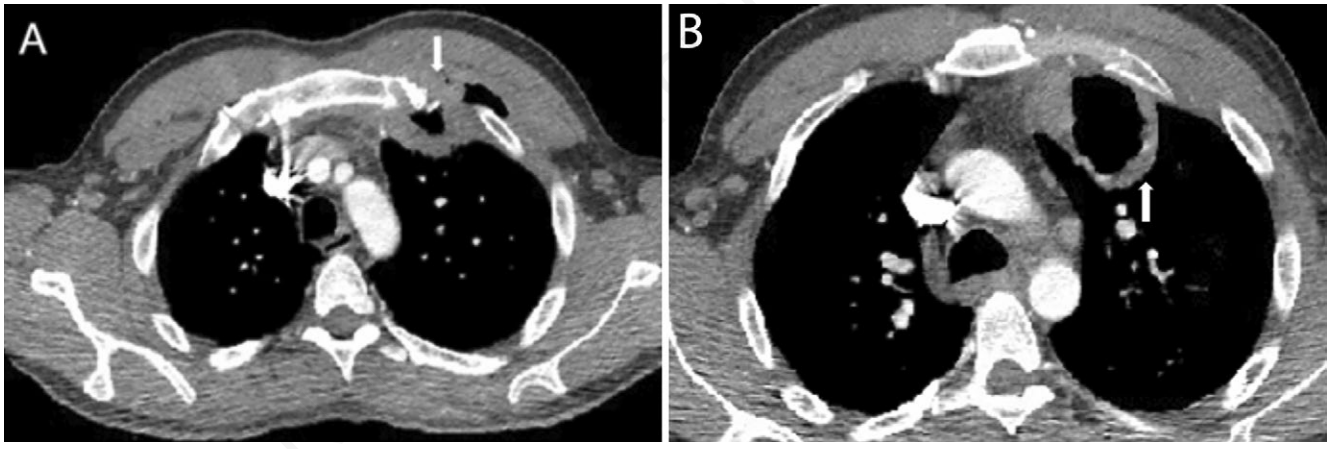

Figure 1. A) Axial cross-sectional images of Computed Tomography of chest, demonstrating air (arrow) tracking across the pectoralis muscle into the subcutaneous tissue planes. B) Axial cross section of CT chest demonstrating left upper lobe cavity (arrow).

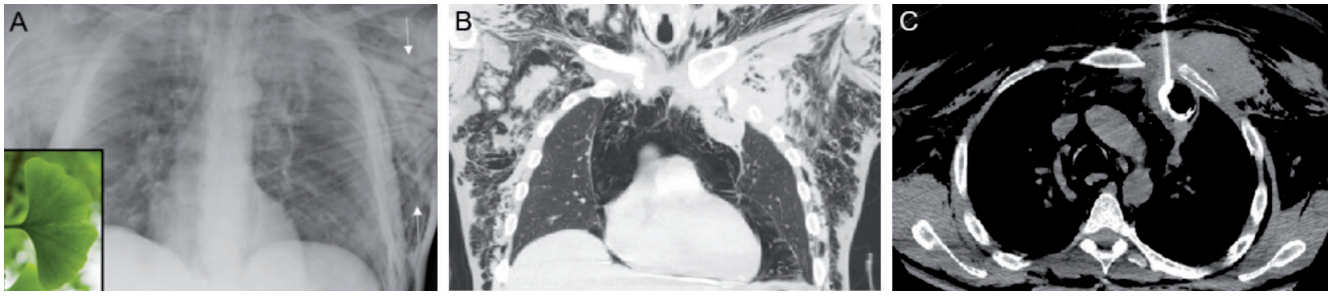

Figure 2. A) Chest X ray post-surgical wound exploration, demonstrating air(arrows) tracking along muscle fibers of pectoralis major muscle, akin to the "gingko leaf" (inset). B) Coronal reconstruction of Chest CT scan demonstrating extensive subcutaneous emphysema (arrow) and pneumo-mediastinum (star). C) Insertion of intercostal chest tube into cavity.

ous extension of intrathoracic infection to the skin surface remains a rare cause of a BCF.

In order to form a broncho-cutaneous fistula, at least two prerequisites must be met. First, a direct communication between the broncho-alveolar tissue and the subcutaneous tissue must occur, and second, the pressure within the bronchus must at least transiently exceed the pressure in the subcutaneous tissues, leading to a pressure gradient, along which air can flow out into the subcutaneous tissues [1]. In order to form a BCF without a BPF (and by extension, a pneumothorax), the parietal and visceral pleural surfaces must be adherent to each other, so that the entry of bronchial air into the pleural space is averted.

The diagnosis of a BCF is usually suggested by physical examination findings of swelling and crepitus overlying the affected skin. Interestingly, Krumpe and colleagues [2], in 1981, described a diagnostic sign known as the "bronchial leak squeak" . When a patient with a BCF performs a Valsalva maneuver, bronchial air is forced through the fistula with turbulence, causing an audible "squeak" at the bedside. In so doing, they suggest that variations in the pitch and intensity of the squeak maybe inversely related to the inner diameter of the fistula. Aside from physical diagnostic signs, a common finding on a chest radiograph is the radiolucent striations overlying the area of the pectoralis major muscle, as air under positive pressure tracks along the pectoralis muscle fibers. The radiological sign has been named the "gingko leaf sign", referring to the similarities of the gaseous outlines of muscle fibers of the pectoralis major with the radiating and branching veins of the gingko leaf (Figure 2A). Chest radiology literature has described this as a typical radiological finding of the thoracic polytrauma patient [3]. A pneumomediastinum is suggested by the presence of the "continuous diaphragm sign", where on a Chest radiograph, the central portion of both diaphragms is seen in continuity across the midline.

We performed a PubMed search using the string of terms "broncho-cutaneous fistula" and reviewed the results to identify reports describing the etiologies and management strategies employed to treat these cases. We similarly searched Google Scholar with the same string to capture any publications not indexed in PubMed. The reference lists of pertinent articles identified through this strategy were also screened for possible additional matches. Only English-language publications were extracted. Table 1 summarizes the most salient features of the 25 cases, including the current one, described in available English-language publications.

From the data available (Table 1), it would appear that the disease is commonly associated with ionizing radiation to the chest, as a complication of thoracic surgery and more infrequently, associated with contiguous spread from necrotizing pneumonia. Management strategies have varied widely, with conservative minimally invasive options being favored initially, and more aggressive surgical interventions favored late in the disease course, when symptom burden tends to be heavy. The infrequent nature of this complication precludes any form of organized study of interventions mapped to their respective success rates, resulting in a paucity of evidence-based guidelines and recommendations for the management of this condition. Interventions range from Endobronchial valve placement, chemical pleurodesis, Fibrin glue sealant, Intercostal chest tube placement into the bronchus, to surgical pedicled muscle flap creation and an infra-scapular "blowhole skin incisions" that allows release of air from the subcutaneous tissues.

The role of bronchoscopy in managing a BCF has historically been limited to localization of the culprit bronchus feeding the air leak. Recently, with the advent of novel therapeutic bronchoscopy techniques, pulmonologists are assuming a central role in the management of patients with the condition, especially once conservative measures have failed. The cornerstone of management remains the localization of the bronchial lobe/ segments that contribute to the continued leak of air through the fistula, thus preventing its healing and closure [4]. The technique involves the successive balloon occlusion of the ipsilateral lobe / segmental bronchus, while observing for a decrease in the air leak through the chest tube over the next few breaths. Culprit bronchial segments are not always predictable anatomically (despite the assistance of CT imaging), due to the phenomenon of distal collateral ventilation with neighboring anatomically distinct lobes [5]. On average, 3-4 bronchial segments maybe contributing to the air leak [4,6], and require a measured and deliberate approach in their localization. A digital airflow system connected to the chest tube is a useful adjunct to this process, as relying purely on a decrease in "bubbling" through the chest tube maybe insensitive in determining which bronchial segments contribute significantly to the air leak, aside from having large inter-observer variability.

Once isolated, a variety of bronchoscopic techniques have been utilized to close off the feeding bronchi, including fibrin glue, scle-

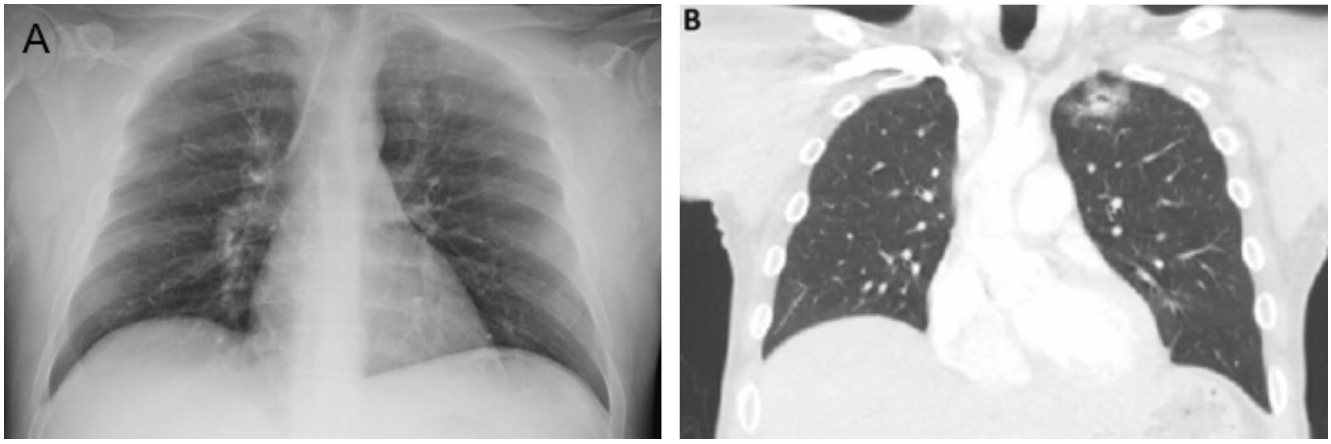

Figure 3. A) Chest X ray 6 weeks post chest tube and long term intravenous antibiotic therapy. B) Coronal Chest CT set to lung window showing a healing cavitary lesion in the right upper lobe, with resolution of the subcutaneous emphysema and pneumomediastinum.

Table 1. The most salient features of the 25 cases described in the available English-language publications.

#	Source	Location	Age/sex	PMHx	Etiology	BPF Y/N	Management strategy	Outcome
1	Haubrich [8]	USA	71/F	Squamous cell cancer lung, local radiotherapy.	Crepitant pneumococcal cellulitis	N	Failed IV antibiotics →Surgical thoracoplasty successful	Cure
2	Roberts [9]	UK	78/M	CAD, s/p CABG	Large bore intercostal chest tube	N	VATS repair	Cure
3	Patris [10]	Iran	79/M	CAD, s/p CABG	Epicardial pacemaker leads	N/A	Conservative (IV antibiotics) due to patient wishes	Frequent LRTI and URTI
4	Omori [11]	Japan	72/M	Esophageal cancer, surgical lobectomy, endobronchial valve placement	Surgical lobectomy	Y	Surgical thoracoplasty	No data
5	Vakil [12]	USA	52/M	Breast cancer, ternal mesh implantation	Sternal mesh infection	N	Endobronchial valve placement after methylene blue instillation	Cure
6	Bui [13]	USA	63/F	Adenocarcinoma lung	Microwave lung ablation	N	Conservative management	Cure
7	Daib [14]	USA	61/M	Pulmonary tuberculosis, BPF, surgical repair	Tuberculosis	Y	Endobronchial valve placement	Cure
8	Thivolet [15]	France	63/F	Sarcomatoid breast cancer	Microwave ablation of nodule	N	Percutaneous chest tube in cavity, fibrin glue	Persistent pulmonary cavity, BCF
9	John [16]	USA	53/F	Pneumonia, parapneumonic effusion	Chest tube insertion	N	Thoracotomy with fibrin glue placement	Cure
10	Koh [17]	Singapore	47/M	Pulmonary tuberculosis, lobectomy	Thoracoplasty, Aspergillosis	Y	Right mainstem stenting with omental path repair	Cure
11	Baildam [18]	UK	25 weeks /M	Surfactant deficient lung disease	Mechanical ventilation, chest compressions	Y	Conservative	Cure
12	Snell [19]	Australia	53/M	Aspergillosis	Lobectomy	N	Endobronchial valve placement	Cure
13	Reich [20]	USA	33/M	Diabetes	Necrotizing Fungal infection (phycomycetes)	N	Percutaneous chest tube placement →Thoracoplasty	Passed away
14	Abu-hijileh [21]	USA	63/M	NSCLC, s/p wedge resection	Radiofrequency ablation	Y	Endobronchial valve placement	Cure
15	Azuma [22]	Japan	64/F	Breast cancer, radiation therapy	Radiation therapy	N	Surgical thoracoplasty, omental flap	Cure
16	Fraser [23]	Canada	63/M	Squamous cell Cancer Lung	Radiotherapy	N	N/A	N/A
17	Radvany [24]	USA	82/F	Adenocarcinoma lung	Radiofrequency ablation	N	Percutaneous chest tube into cavity, fibrin sealant	Cure
18	O'Neil [25]	Ireland	60/M	NSCLC Lobectomy	Lobectomy	Y	N/A	N/A
19	Biswas [26]	UK	45/M	None	Trauma	N	Surgical repair	Passed away
20	Kaur [27]	India	16/M	None	iatrogenic (during laparotomy for amoebic liver abscess)	N	Surgical repair	Cure
21	Marwah28	India	25/M	Tuberculosis	Lobectomy	N	Transcutaneous closure by Duct occlude device	Cure
22	Isomura29	Japan	34/M	Large cell lung cancer	Contiguous spread of malignancy	N	Conservative	N/A
23	saad30	Saudi Arabia	60/F	Diabetes, CKD	Necrotizing fungal infection	N	Conservative management (poor general condition)	Persistent fistula
24	Virgilio31	Italy	55/F	Hydatid cyst removal by laparotomy	Contiguous extension	N	Conservative, anthelminthic agents	N/A
25	Current case	USA	47/M	Diabetes, IV drug abuse	Contiguous spread of necrotizing pneumonia	N	Percutaneous chest tube placed into cavity	Cure

PMHx, past medical history; IV, intravenous; CAD, coronary artery disease; CABG, coronary artery bypass grafting; VATS, video assisted thoracoscopic surgery; BPF, broncho-pleural fistula; N/A, not available.

rosant agents and implants, with mixed results [4]. Endobronchial valves have recently benefited from a wider adoption in the management of BPF and BCF [7], as well as their original indication of use, for non- operative lung volume reduction. After patients have responded to the intervention, close follow up in 4-6 weeks is recommended, to assess for the possibility of valve removal [4]. Positive pressure ventilation may exacerbate and/or contribute to the formation of a broncho-cutaneous fistula. When a BCF is suspected, every effort must be taken to avoid positive pressure ventilation during elective operative procedures. Possible options include selective intubation of the contralateral main-stem bronchus for the procedure, or the employment of spontaneous breathing with pressure support ventilation, which may decrease the exposure of positive pressure to the area. Although both conditions are generally benign, cases of airway compromise and hemodynamic compromise have been reported. Airway compromise is usually preceded by dysphonia and a change in voice. Hemodynamic compromise occurs when the pressure outside the major venous vessels in the mediastinum exceeds the relatively lower pressures within them, causing a decrease in venous return, and fall in cardiac output by the Frank-starling mechanism.

In our case, we opted for a conservative approach, with supplemental Oxygen and placement of an intercostal chest tube into the cavity. We initially placed the apparatus on low intermittent suction of -20 cm of water, in an effort to evacuate air from the area, allowing antibiotics time to clear the site of infection, allowing wound healing and closure of the fistula. Supplemental oxygen was used to wash out Nitrogen from the proximal airways, to facilitate a gradient along which air in the subcutaneous tissues and mediastinum maybe absorbed. The distal end of the BCF at the exit site from the skin was treated with a wound vacuum dressing to assist in evacuation of air from the subcutaneous tissues. The patient eventually improved, his fistula healed, and was discharged to complete a course of long-term antibiotics.

Conclusions

A broncho-cutaneous fistula is a rare complication of necrotizing pneumonia, caused by contiguous spread of infection into the subcutaneous planes. Increased awareness of the entity will allow clinicians to anticipate complications early, avoiding diagnostic delays. Techniques to avoid further worsening of the clinical status during episodes that require mechanical ventilation include selective intubation of the contralateral main-stem bronchus, or the use of spontaneous breathing modes on the mechanical ventilator. Clinicians may favor conservative options such as pig-tail insertion and oxygen supplementation initially, with more invasive surgical options indicated with increased symptom burden.

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