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Case report

A case of broncho-cutaneous fistula secondary to tuberculosis successfully managed with awake veno-venous extracorporeal membrane oxygenation

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ABSTRACT

A broncho-cutaneous fistula (BCF) is a communicating tract between the bronchus and the cutaneous surface of the thoracic wall and can be the primary presenting sign of several disease processes. It has been associated with positive pressure ventilation (PPV), post pneumonectomy, thoracostomy tubes, perforating chest trauma, neoplasia and chronic empyema. We report a case of a 45-year-old immunocompetent man presenting with severe hypercapnic respiratory failure secondary to a BCF as a result of tuberculosis (TB)-related empyema necessitans. Veno-venous extracorporeal membrane oxygenation (VV ECMO) was employed during spontaneous breathing to mitigate the risks of PPV, to facilitate diagnostics and enable targeted treatment. Awake VV ECMO is an effective supportive therapy for complex, destructive lung pathologies with a known reversible aetiology in which PPV would be potentially detrimental.

1. Introduction

Disseminated tuberculosis (TB) is life threatening, affecting 2 or more non-contiguous sites owing to hematogenous or lymphogenous spread either resulting from progressive untreated primary infection or reactivation of latent infection [1,2]. Rarely, untreated tuberculous empyema results in empyema necessitans with multisystem involvement and infrequently broncho-cutaneous fistulae. The empyema corrodes the pleura, tissues and even ribs to form a communication with the cutaneous structures [3]. Here we present an unusual case of previously undiagnosed disseminated TB with complex lung involvement and an anterior chest wall broncho-cutaneous fistula (BCF) which was successfully managed with veno-venous extracorporeal membrane oxygenation (VV ECMO).

2. Case presentation

A 45-year-old previously healthy man with an extensive history of backpacking through Far East Asia and Africa presented to a district hospital with a 6-week prodrome of shortness of breath associated with fevers, night sweats, anorexia, 20-kg weight loss and an indolent right anterior chest wall mass. There was no clear recent infective exposure. He had been self-isolating at home with anosmia and ageusia during the Coronavirus pandemic.

On presentation he was extremely cachectic with a body mass index of 18, diaphoretic, and deteriorating with type-2 respiratory failure. He was transferred to the intensive care unit (ICU) where he was commenced on non-invasive ventilation (NIV). It was noted that the right anterior chest wall mass was waxing and waning with the respiratory cycle. He underwent a computerised tomography (CT) scan of the thorax which showed an abnormal communication between his pleura and the right anterior chest wall. He also had a complex right-sided, loculated effusion and diffuse apically predominant cystic changes in both lungs.

Despite NIV he continued to deteriorate with hypoxia, hypercarbia and high work of breathing. He was referred to an ECMO centre due to the risks associated with PPV in the context of a BCF.

At the referring hospital the percutaneous cannulation was performed under local anaesthesia and the patient was initiated on VV ECMO. To ensure patient safety and cooperation a low dose of 25mg intravenous ketamine was administered peri procedure. The patient was conscious throughout. Once hypoxia and hypercarbia were corrected with VV ECMO, his work of breathing settled and NIV could be safely discontinued. He did not require further sedation or analgesia. Endotracheal intubation and further risks of PPV were therefore avoided, and he was transferred to the tertiary centre for further management.

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On arrival, and according to local protocol, he underwent CT imaging of his brain, thorax, abdomen, pelvis and a CT pulmonary angiogram. In addition to the known previous findings, the striking abnormalities in the lungs were two adjacent defects in the right anteromedial chest wall with gas extending from the pleural space, through the defects into a large gas-filled cavity in the overlying subcutaneous tissues [Figs. 1–4]. Additional findings were paraaortic fluid, a right pneumothorax, multiple hypoattenuated liver lesions, complete destruction of the right 5th to 8th ribs and lytic lesions on thoracic vertebrae 1 and 6. A transthoracic echocardiogram (TTE) demonstrated a pericardial effusion of 2.0 cm anterior to the right ventricle with no tamponade physiology. He received 24% oxygen via a nasal cannula and required a low level of ECMO support which alleviated his work of breathing, reducing the risk of patient self-inflicted lung injury (P-SILI) with exacerbation of the underlying lung injury.

The clinical picture and multisystem involvement suggested a unifying diagnosis of disseminated TB. Empiric *anti*-TB therapy in the form of rifampicin, isoniazid, pyrazinamide and ethambutol were commenced promptly in addition to an empiric broad spectrum cover with a 7-day course of co-amoxiclav plus anidulafungin for community acquired pneumonia (CAP). Atypical pathogens were covered with doxycycline for 2 days which was discontinued upon negative microbiology results. Methylprednisolone 1 mg/kg/day was also added in view of the pericarditis presumed to be tuberculous in origin [4]. The patient declined a bronchoscopy and induced sputum for analysis, however agreed to have an intercostal chest drain (ICD) inserted. Two litres of empyema were drained under ultrasound control. Rifampicin sensitive *Mycobacterium tuberculosis complex* was confirmed from the drain fluid using a nucleic acid amplification test (NAAT) (GeneXpert® MTB/RIF assay). No acid-fast bacilli were seen on Ziehl-Neelsen staining. Gram stain showed some pus cells and scanty growth of bacillus simplex from the culture.

All diagnostic sampling for bacterial, viral and fungal species were negative for legionella, pneumococcus, cryptococcus, histoplasma, syphilis, human immune deficiency virus, hepatitis B & C, cytomegalovirus, herpes simplex virus and SARS CoV-2; serum beta-D-glucan and galactomannan were also negative.

After 7 days he was weaned from VV ECMO and successfully decannulated. The right lung had re-expanded [Figs. 5–7] and the air leak spontaneously resolved. No complications related to the ECMO support were noted during his ICU stay. A repeat TTE demonstrated a mild reduction in the size of the pericardial effusion.

3. Discussion

To the best of our knowledge, we are reporting the first case of BCF secondary to empyema necessitans from TB supported with awake VV ECMO.

There have been reports of BCF associated with chronic suppurative lung infections such as aspergillus [5], actinomyces [6], necrotic neoplasia and interestingly an isolated case report of bronchopleurcutaneous fistula in the absence of any empyema in pulmonary TB [7]

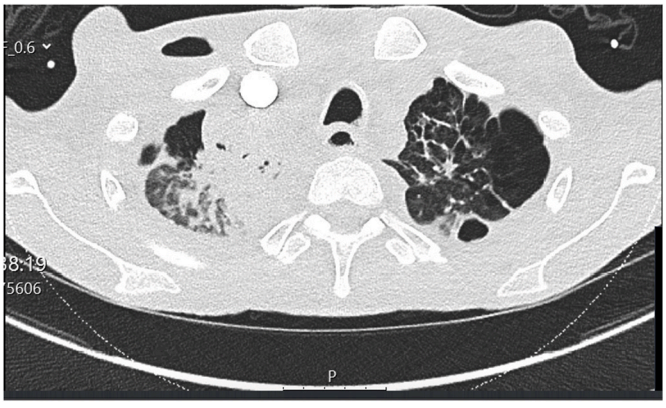

Fig. 1. CT scan at tertiary centre. Cystic chances at apices and air noted in the right anterior chest wall.

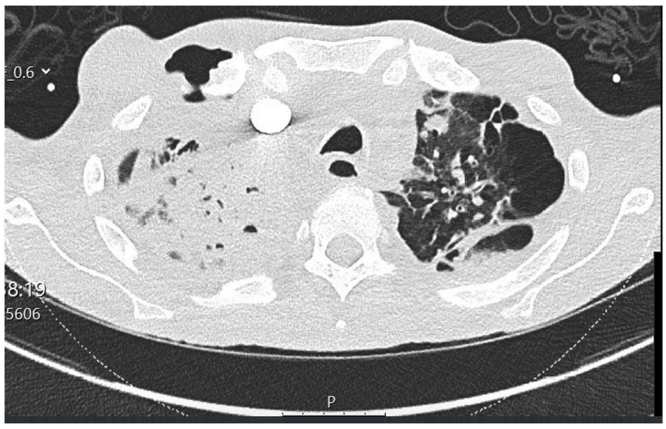

Fig. 2. CT scan showing the defect in the right anteromedial chest wall.

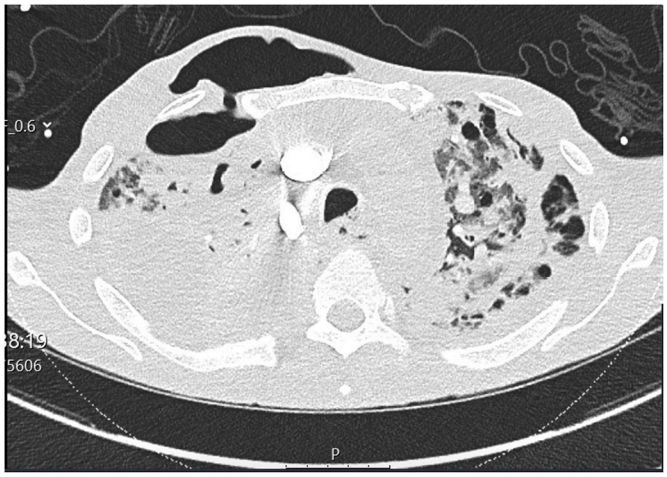

Fig. 3. CT scan showing gas extending from pleural space to the right anteromedial chest wall.

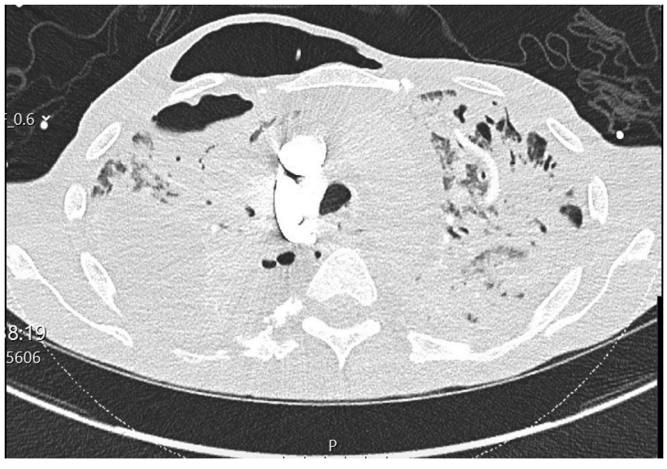

Fig. 4. CT scan showing large gas-filled cavity overlying the subcutaneous tissues.

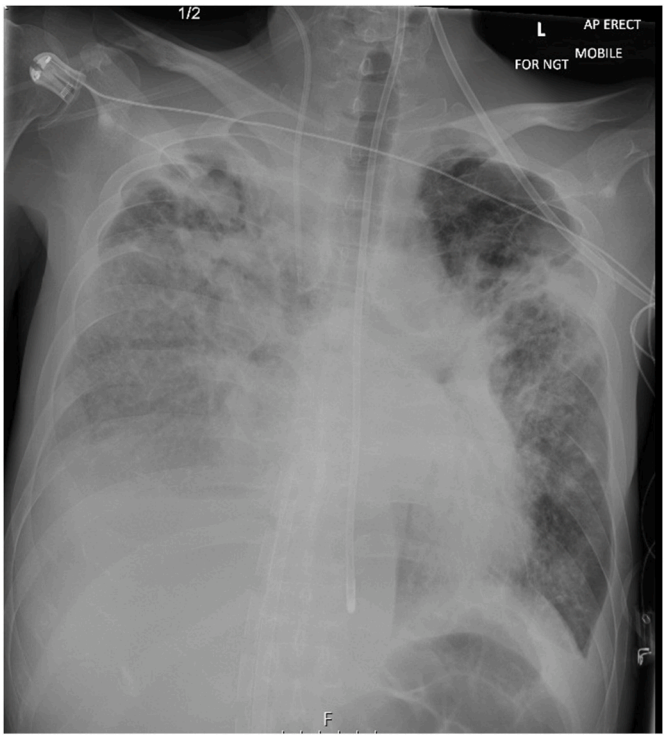

Fig. 5. Chest X-ray before intercostal chest drain insertion.

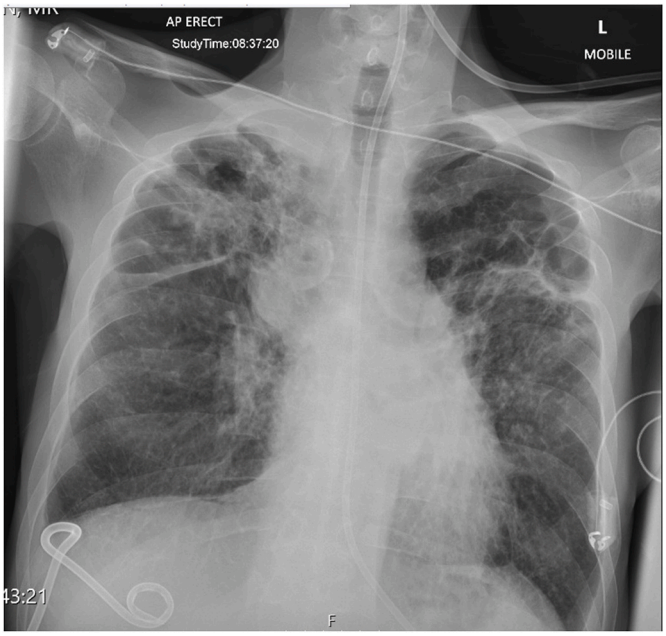

Fig. 6. Chest X-ray after intercostal chest drain insertion and drainage of empyema.

but few reports of TB causing severe respiratory failure requiring ECMO. Barotrauma is a well described complication of mechanical ventilation (particularly in those with chronic lung disease) [8], as well as self-inflicted lung injury [9] due to excessive work of breathing. Mechanical ventilation in TB cases has been associated with a very high mortality of 60–90% [10], and high-pressure ventilation in air leaks resulting in unfavourable outcomes due to delay in fistula healing [11]. In this case with VV ECMO, PPV was completely avoided escaping the cycle of worsening of the BCF and exacerbation of pre-existing lung

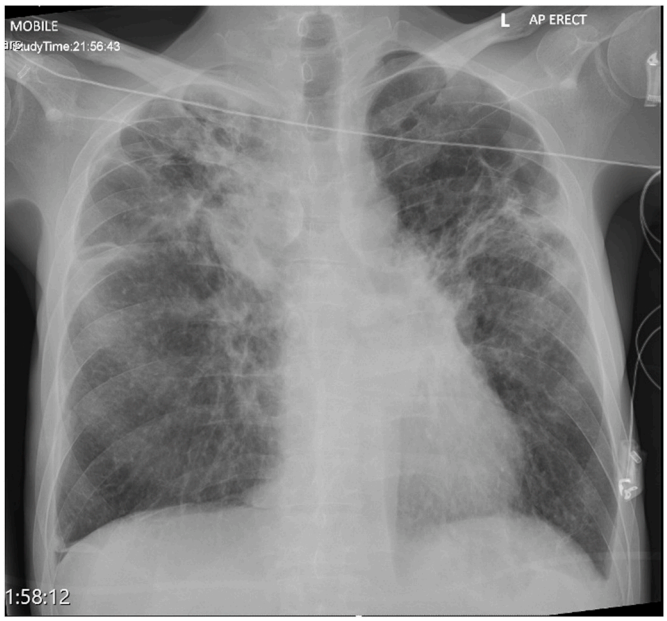

Fig. 7. Chest X-ray after removal of intercostal chest drain and decannulation from VV ECMO.

injury which could have proven catastrophic. Trans-pleural fistulae owing to tuberculous empyema require prompt *anti*-TB medications and sometimes surgery as the mainstay of treatment [12]. Despite advances in diagnostics and medical therapies, delayed presentations with thoracic fistulous disease and air-leaks are not unheard of with reports of medical management being the most appropriate course of treatment in such cases [13].

Lung protective invasive ventilation in combination with ECMO, promoting lung rest and healing or closure of fistulae have been used in cases of air leaks [14–17], alternatively awake VV ECMO can provide a bridge to diagnosis, treatment and recovery whilst avoiding potentially injurious PPV altogether.

There has been an increase in use of awake ECMO in recent years as a bridge to lung transplant in patients with end-stage respiratory disease [18]. Furthermore, it is associated with a lower mortality compared to patients on ECMO who are mechanically ventilated [19]. Risks associated with mechanical ventilation such as prolonged sedation, ventilator-induced lung injury and ventilator associated pneumonia can be bypassed [20]. Awake ECMO can be challenging as patient compliance is crucial to avoid inadvertent complications [21] and also requires experienced multi-disciplinary staff to appreciate the dynamic changes in an unpredictable awake patient. Extracorporeal support has to be used in a safe and effective manner in an awake patient to ensure therapeutic success. Careful titration of sedation and the ECMO support requires a skilled approach in order to control the patient's respiratory drive and avoiding hypoventilation.

Physical therapy and early mobilisation can also be performed safely for patients on ECMO [22]. Awake patients on ECMO have the added advantage of engaging in prompt rehabilitation. This reduces deconditioning frequently seen in patients with severe respiratory failure and leads to a multitude of benefits including improved muscle strength, physical function, reduced incidence of delirium and improved quality of life. The advantages are not limited to the patient but also results in reduced healthcare costs and length of stay.

4. Conclusions

We report a case of BCF secondary to a delayed presentation of disseminated TB successfully managed with a short duration of awake VV ECMO. The use of awake VV ECMO enabled our patient to safely

breathe spontaneously, receive optimal medical therapies, eat and drink, engage in physical therapy and receive psychological support.

Amongst lung injury amenable to surgical therapy, BCF is a recognised indication for ECMO. For those patients who are unsuited for surgical intervention, awake VV ECMO can be a viable alternative strategy for supportive management. This approach should be investigated and explored further.

Consent for publication

Informed consent was provided by the patient for the publication of this case report.

Availability of data and material

All data supporting the findings is contained within the manuscript.

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Author's contributions

Conceived and designed the report: GC and DW. Wrote the first draft of the manuscript: GC. All authors read, contributed towards and approved the final manuscript.

Declaration of competing interest

The authors know of no conflicts of interest associated with this publication.

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References

[1] F.Y. Khan, Review of literature on disseminated tuberculosis with emphasis on the focused diagnostic work up, J. Family Community Med. (2) (2019) 83–91. May-Aug 26.
[2] S. Ribeiro, D. Trabulo, C. Cardoso, et al., Disseminated Tuberculosis in an Immunocompetent Patient: the Answer Is the Liver- GE Port of Gastroenterol, 2016, pp. 208–213. Jul-Aug 23 (4).
[3] S.A. Kono, T.D. Trausser, Contemporary empyema necessitates, Am. J. Med. 120 (2007) 303–305.
[4] C. Schutz, A.G. Davies, B. Sossen, et al., Corticosteroids as an adjunct to tuberculous therapy, Expet Rev. Respir. Med. 10 (12) (2018).
[5] E.J. Soto-Hurtado, E. Marín-Gámez, N. Segura-Domínguez, et al., Pleural aspergillosis with bronchopleurocutaneous fistula and costal bone destruction, Lung 183 (2005) 417–423.
[6] A. Gupta, R.F. Lodato, Empyema necessitatis due to actinomyces israelii, Am. J. Respir. Crit. Care Med. 12 (2011) 185.
[7] M.A. Siddiqui, M. Shameem, J. Akhtar, et al., Bronchopleuralcutaneous fistula in the absence of empyema. A rare presentation of pulmonary tuberculosis, Jan-Mar 28 (1) (2011) 72–73. Lung India.
[8] A. Anzueto, F. Frutos, V.A. Esteban, et al., Incidence, risk factors and outcome of barotrauma in mechanically ventilated patients, Intensive Care Med. 30 (2004) 612–619.
[9] D.L. Grieco, L.S. Menga, D. Eleuteri, et al., Patient self-inflicted lung injury: implications for acute hypoxemic respiratory failure and ARDS patients on non-invasive support, Minerva Anestesiol. 85 (9) (2019) 1014–1023. September.
[10] M. Anderson, P Tapia M Mercado, et al., Catastrophic respiratory failure from tuberculosis pneumonia: survival after prolonged extra corporeal membrane oxygenation support, Respir Med Case Rep 10 (2013) 19–22.
[11] O. Daoud, P. Augustin, P. Mordant, Extracorporeal membrane oxygenation in 5 patients with bronchial fistula with severe lung injury, Ann. Thorac. Surg. 92 (1) (2011) 327–330. Jul.
[12] N. Navani, S. Punwani, P.D. Humphries, et al., Pleuro-cutaneous fistula complicating chest drain insertion for tuberculous effusion, QJM 103 (2010 2010) 799–800, 10.
[13] V. Muthu, S. Dhooria, R. Agarwal, et al., Rare case of spontaneous subcutaneous emphysema, Lung India 33 (6) (2016) 688–689.
[14] S. Marek, S. Martin, Z. Ondrej, et al., Extracorporeal membrane oxygenation in the management of post-pneumonectomy air leak and adult respiratory distress syndrome of the non-operated lung, Perfusion 32 (5) (2017).
[15] H.S. Ali, I.F. Hassan, S. George, Extra corporeal membrane oxygenation to facilitate lung protective ventilation and prevent ventilator-induced lung injury in severe Pneumocystis pneumonia with pneumomediastinum: a case report and short literature review, BMC Pulm. Med. 16 (2016) 52.
[16] HWan H He, X. Li, et al., Successful rescue combination of extracorporeal membrane oxygenation, high-frequency oscillatory ventilation and prone positioning for the management of severe methicillin-resistant Staphylococcus aureus pneumonia complicated by pneumothorax: a case report and literature review, BMC Pulm. Med. 17 (2017) 103.
[17] A.A. Grant, E.B. Lineen, A. Klima, et al., Refractory traumatic bronchopleural fistula: is extra corporeal membrane oxygenation the new gold standard? J. Card. Surg. 35 (1) (2020) 242–245. Jan.
[18] K.M. Olsson, A. Simon, M. Stueberet al, Extracorporeal membrane oxygenation in non-intubated patients as bridge to lung transplantation, Am. J. Transplant. 10 (9) (2010) 2173–2178. Sep.
[19] O. Wiesner, J. Hadem, W. Sommer, et al., Extracoporeal membrane oxygenation in a nonintubated patient with acute respiratory distress syndrome, Eur. Respir. J. 40 (2012) 1296–1298.
[20] T. Langer, A. Santini, N. Bottino, et al., "Awake" extracorporeal membrane oxygenation (ECMO): pathophysiology, technical considerations, and clinical pioneering, Crit. Care 20 (1) (2016) 150.
[21] F. Haneke, T.A. Schildhauer, J. Strauch, et al., Use of extracorporeal membrane oxygenation in an awake patient after a major trauma with an incidental finding of tuberculosis, Perfusion 31 (4) (2016) 347–348.
[22] D. Abram, J. Javidfa, E. Farrand, et al., Early mobilization of patients receiving extracorporeal membrane oxygenation: a retrospective cohort study, Crit. Care 18 (2014) R38.