

CASE REPORT

Emergency Laser Treatment of a Tracheobronchial Carcinoid during ECMO

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Abstract

Early diagnosis of endobronchial carcinoids is challenging, as they often mimic other common acute respiratory conditions at first presentation. Increasing consensus favours surgical resection over endoscopic management of bronchial carcinoids whenever possible.

ECMO has been reported to be an effective supportive strategy in many cases of elective and urgent surgical or endoscopic airway procedures. However, it has never been described as a supportive technique for the emergency endoscopic management of an endobronchial carcinoid.

Herein we report the case of a 17 year-old girl presenting with spontaneous pneumomediastinum and an almost complete endotracheal obstruction at the level of the main carina, due to a typical carcinoid that was treated successfully by endoscopic laser disruption under veno-venous extracorporeal circulation in an emergency scenario.

Introduction

Bronchial carcinoids are malignant neoplasms with neuroendocrine differentiation, representing 1-5% of all lung tumors.¹ Carcinoids may be defined according to the 2015 WHO classification on the basis of the number of mitoses; low-grade (typical) carcinoids and intermediate-grade (atypical). Atypical carcinoids have a higher likelihood of metastases, a higher recurrence rate and a worse prognosis.²

Treatment recommendations suggest endobronchial laser resection, rather than surgical resection, only for low- and intermediate-grade neoplasms in patients with compromised general conditions³. However, to the best of our knowledge, there is no evidence of emergency extracorporeal membrane oxygenation (ECMO) application for the endoscopic management of bronchial carcinoids.

Case-report

A 17-year-old, female non-smoker, was diagnosed with allergic asthma six months before coming to our observation due to progressive dyspnea and diffuse bilateral wheezing, which worsened when supine. During the night, acute dyspnea with orthopnea, violent coughing and severe bronchospasm occurred

suddenly while fresh-snow crackles were clearly detectable at neck palpation. Arterial blood gas test, performed under oxygen therapy, demonstrated a very severe respiratory failure (P/F = 56). Chest CT scan confirmed the presence subcutaneous emphysema with pneumomediastinum and evidenced a solid, dense, mucoid lesion, almost completely occluding the trachea at the level of the main carina (figure 1). The patient was taken to the operating theatre, where a flexible bronchoscopic inspection was performed. It revealed a spontaneously bleeding formation, originating from the main carina, which almost totally occluded both main bronchi. As there is no thoracic surgeon on night duty in our hospital and given the life-threatening condition of the patient, surgery was excluded. Furthermore, orotracheal intubation or tracheostomy would have been ineffective due to the low airway obstruction. Therefore, we decided to set up a veno-venous extracorporeal circulation (ECMO) to attempt an endoscopic laser disruption of the tumor. The active lesion bleeding necessitated a minimizing anticoagulation for the extracorporeal circulation (on a risk-benefit basis). Rigid bronchoscopy allowed us to control the bleeding and to carry out progressive debulking using a diode laser (Figure 2).

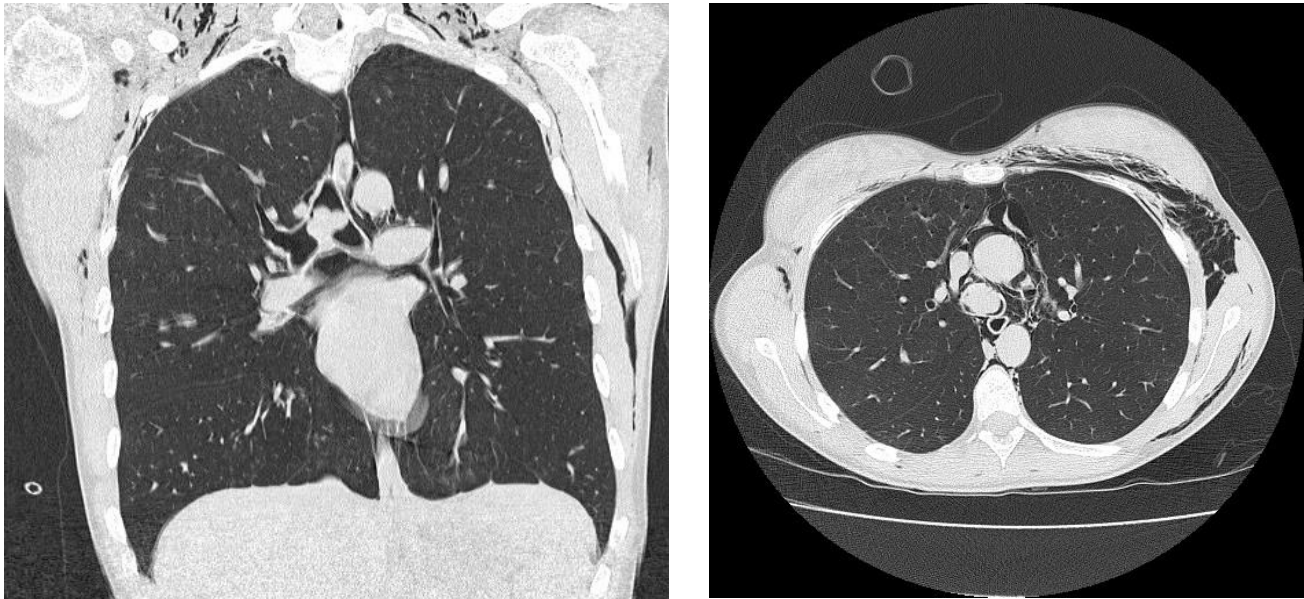


Figure 1. Chest CT showing the endobronchial mass occluding the trachea and both main bronchi (coronal section, left panel; transverse section, right panel). Diffuse subcutaneous emphysema is also evident.



Figure 2. Endoscopic view of the main carina after laser disobstruction.

Extemporaneous and histological examination provided a diagnosis of a well-differentiated (grade I) neuroendocrine neoplasm, also known as a typical carcinoid. Extracorporeal circulation was opted out twelve hours later, thanks to a

notable improvement in gas exchanges and the absence of further bleeding. The Octreoscan test, performed two months after discharge, found no tissue receptor overexpression at either a thoracic or abdominal level. After three years of

regular 6-monthly endoscopic follow-ups, subcarinal lymph node sampling (EBUS-TBNA) evidenced a carcinoid relapse (figure 3). Therefore, thoracic surgery was performed carrying out mediastinal lymphadenectomy, excision of the distal part of the trachea and the last three tracheal rings, removal of the proximal

tract of the right main bronchus and airway reconstruction by means of end-to-end anastomoses. A close follow-up with flexible bronchoscopy and chest CT scans was then re-initiated, the last available ones were negative for disease relapse after two years from surgery (figure 4).

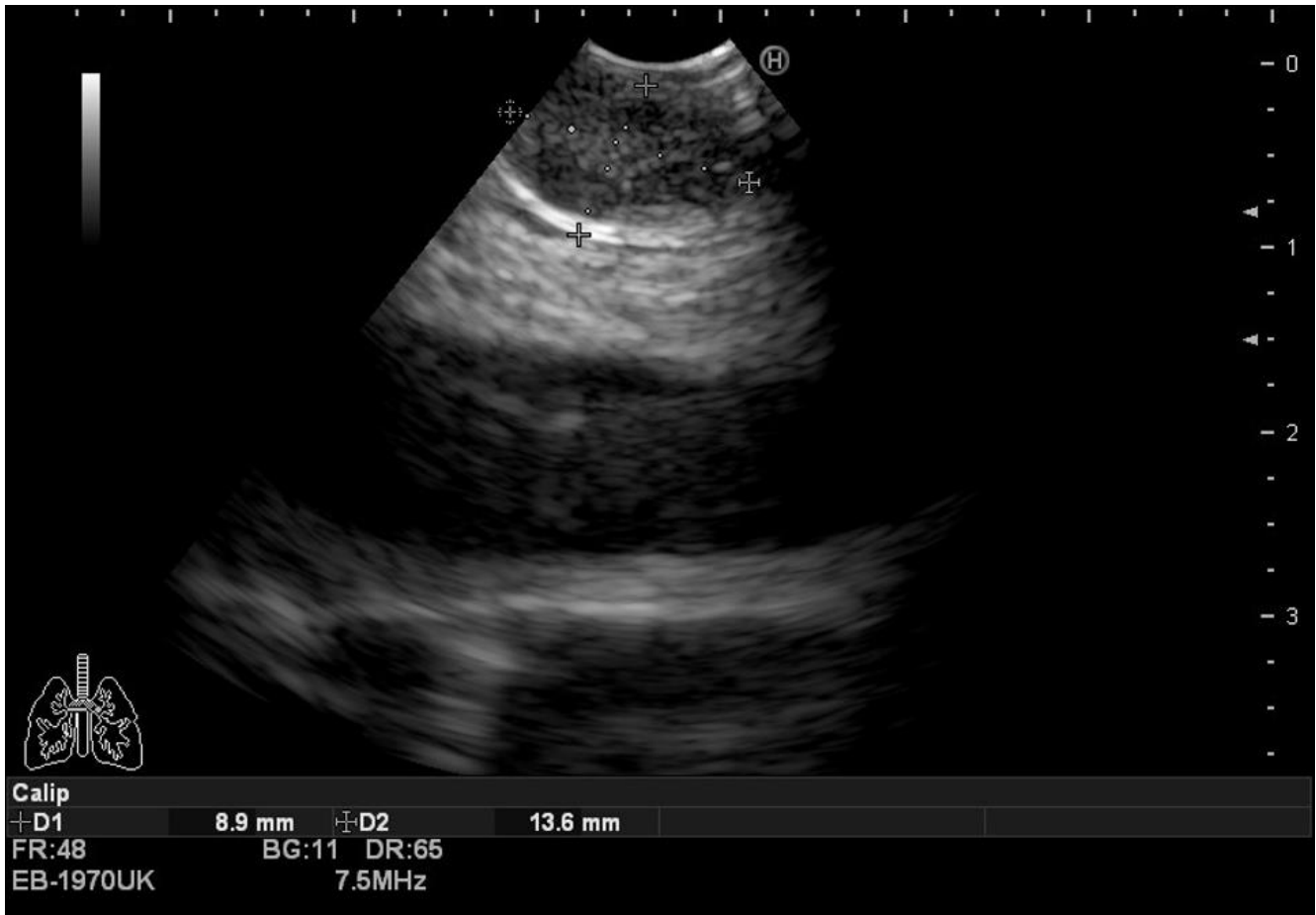


Figure 3. Endobronchial ultrasound (EBUS) imaging of enlarged subcarinal lymph node. Needle aspiration sampling (TBNA) showed evidence of disease relapse.



Figure 4. Chest CT showing no macroscopic evidence of disease relapse after two years from surgery, as confirmed by bronchoscopy. The anastomosis between the distal part of the trachea and the proximal tract of the right main bronchus is also visible in this section.

Discussion

Bronchial carcinoids are usually low-grade malignant neoplasms with neuroendocrine differentiation and an indolent clinical behaviour representing 1-5% of all lung tumors.¹ There is no association with carcinoids and cigarette smoking and they are usually sporadic, although there are some familial cases of Multiple Endocrine Neoplasia type 1.^{4,5} Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia (DIPNECH) is a distinct condition, where generalized proliferation of pulmonary neuroendocrine cells may develop into carcinoid tumours or present with symptoms mainly related to hormone production.² According to the 2015 WHO classification, low-grade (typical) carcinoids are characterized by less than 2 mitoses per mm² in 10 high-power fields and usually have an excellent prognosis. Conversely, intermediate-grade (atypical) carcinoids are

characterized by more than 2 mitoses per mm² in 10 high-power fields and the presence of coagulative necrosis; and are more prone to metastases, have a higher recurrence rate and a worse prognosis, especially if mediastinal nodes are involved.²

Most patients have a centrally located tumor and are asymptomatic, except for a long history of coughing, breathlessness and wheezing, often misdiagnosed as asthma⁶. Indeed, there are few reports of typical bronchial carcinoid tumors presenting with pneumomediastinum.^{7,8} Early recognition of lung carcinoids is also made difficult due to the poor sensitivity of chest X-rays. However, flattening of both limbs of the flow-volume loop with a FEF50/FIF50 ratio close to 1 may be of help in earlier recognition of a fixed intrathoracic obstruction compared to chest imaging.⁹

The most recent treatment recommendations suggest surgical resection (including mediastinal lymph node sampling or dissection) in every patient with either low- or intermediate-grade tumour as long as they would be able to tolerate the procedure.³ However, in our case, surgery was rapidly ruled out given the hyperacute presentation, the emergency setting and no thoracic surgeon in active service during the night.

Extracorporeal membrane oxygenation (ECMO) has been described as safe support in patients with severe tracheal obstruction undergoing elective or urgent surgery for severe tracheal obstruction or complex tracheo-bronchial procedures.¹⁰⁻¹⁷ ECMO has also been set-up before the induction of general anesthesia, due to the risk of ventilation failure during bronchoscopic resection of endobronchial lesions¹⁸ and there are some reports on urgent ECMO support for the bronchoscopic management of tracheal obstruction due to exogenous aspiration, pulmonary hemorrhage, tracheal papillomatosis or sarcoma.^{19,20}

However, to the best of our knowledge, there is no evidence of ECMO being used for the emergency endoscopic management of a bronchial carcinoid.

Conclusion

Herein we report the case of a 17 year-old girl presenting with pneumomediastinum and an almost complete endotracheal obstruction at the

level of the main carina due to a typical carcinoid, successfully treated by endoscopic laser disruption under veno-venous extracorporeal circulation in an emergency scenario. Our patient had a three-year disease-free period before relapse and definitive surgical excision. Further studies on larger populations are needed to confirm that endoscopic resection might be considered a first-line approach in the rescue management of typical endobronchial carcinoids, allowing for the planning of elective, less risky and more effective surgical strategies.

Conflicts of interest

The authors have no conflicts of interest to disclose.

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