ELSEVIER

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr



Case Report

A ball valving carcinoid tumour as a cause of post bronchoscopy chest pain

Andrew Mowat*, Anna Werpachowska, Ingrid du Rand

Wye Valley NHS Trust, Stonebow Road, Hereford, HR12BN, United Kingdom

ARTICLE INFO

Handling Editor: DR AC Amit Chopra

Keywords: Bronchoscopy Ball-valving Carcinoid tumours LASER

ABSTRACT

The ball valve effect occurs when an obstructing lesion allows inspiration of air, but opposes the egression of exhaled air, causing gas trapping. The phenomenon is commonly described secondary to bronchial foreign body inhalation. However, it is less well reported in other disease processes. We report a unique case of a carcinoid tumour causing ball valving following diagnostic bronchoscopy in a young patient. The procedure caused swelling and oedema around an isolated carcinoid tumour in the left main bronchus. An inspiratory chest X-ray was normal, complicating the diagnosis. At repeat bronchoscopy the tumour was cored with LASER.

1. Background

Diagnostic flexible bronchoscopy is a commonly performed procedure. Complications include minor bleeding, transient hypoxaemia, and pneumothorax. Post bronchoscopy chest pain is well-recognised, occurring after more than 1% of flexible bronchoscopies [1].

We report a rare case of gas trapping secondary to the ball-valve effect due to swelling of a carcinoid tumour following elective bronchoscopy. A literature review is performed.

2. Case report

A twenty nine year old man with a good performance status, presented to respiratory clinic with episodic shortness of breath. An HRCT scan displayed a 2 cm endobronchial lobulated lesion in the left main bronchus (Fig. 1). The lesion was isolated with the lung parenchyma otherwise normal.

He underwent flexible bronchoscopy via a transoral route under moderate sedation with intravenous midazolam and fentanyl. A concentric vascular lesion was identified in the left main bronchus (Fig. 2). No other visible endobronchial lesions or stenosis were seen. Due to a concern over potential haemorrhage, forceps biopsy was avoided. Instead cytology samples were taken by gentle brushings and bronchoalveolar lavage. These were non-diagnostic. The procedure was performed without complication. However, brisk oozing was noted from the lesion during the procedure. The patient was discharged from hospital on the same day.

Ten hours later he presented to the emergency department with a dull pleuritic, non-radiating, central chest pain. This was associated with acute shortness of breath. The pain was worse when supine. Physical examination was reassuring with normal breath sounds throughout both lung fields. Blood tests were within normal limits.

E-mail address: andrewmowat@live.com (A. Mowat).

Abbreviations: CT, computed tomography; HR, high resolution; LASER, light amplification by stimulated emission of radiation.

^{*} Corresponding author.



Fig. 1. High Resolution CT showing a 2 cm endobronchial lesion in the left main bronchus.



Fig. 2. Bronchoscopic image of carcinoid lesion in the left main bronchus.

Inspiratory and expiratory chest X-rays were taken (Fig. 3A and B). The inspiratory film showed no abnormality. However, the expiratory film showed mediastinal shift to the right. The patient was diagnosed with gas trapping secondary to the ball valve effect of the tumour. Distal inflation of the lung was progressively pushing the mediastinum away from the site of the lesion.

The patient was referred to cardiothoracic surgery, and transferred to a tertiary centre. He underwent rigid bronchoscopy under general anaesthetic. Using LASER, the tumour was cored with large areas vaporised. He received a follow up rigid bronchoscope six weeks later. The patient went onto to make a full recovery. Histopathological analysis confirmed a neuroendocrine tumour.

3. Discussion

Diagnostic flexible bronchoscopy is a commonly performed procedure. Complications include minor bleeding, transient hypoxaemia, and pneumothorax.

Infective complications are well described and require exclusion. Abscesses and empyema are reported [2]. In this case our patient was apprexial and had normal inflammatory markers.

Chest pain occurs after more than 1% of bronchoscopies and carries a wide differential [1]. The cause can be pulmonary or systemic. Bronchoscopy represents a significant insult to the cardiovascular system. Myocardial infarction, aortic dissection require exclusion. Air embolism presenting with hypotension, bradycardia, and left hemiplegia is described [3].

Iatrogenic pneumothorax is well known cause of pulmonary chest pain following bronchoscopy. The rate is higher after performing transbronchial lung biopsy or suctioning of a mucus plug [4]. Such patients can represent to the respiratory services, but many remain asymptomatic and hence undiagnosed. One prospective study showed of 1443 subjects undergoing interventional bronchoscopy 3.4% had a pneumothorax on routine chest X-ray. Of the 49 patients with pneumothoraxes 13 were symptomatic. Of these 10 had a



Fig. 3A. Inspiratory film.



Fig. 3B. Expiratory film.

change in management, including chest tube placement, inpatient admission, or observation [5]. In our case, the patient had normal breath sounds and inspiratory chest ray showed good inflation of both lungs exluding the differential.

The patient was correctly diagnosed with gas trapping secondary to oedema following a diagnostic bronchoscopy. The expiratory film showing mediastinal shift support this diagnosis.

The tumour had behaved similarly to an obstructive foreign body, causing over inflation of the lungs, due to gas trapping. The ball valve effect is described most commonly in relation to foreign body inhalation. It occurs when a lesion allows an inspiration of air, however, opposes the egression of exhaled air. This results in air trapping and build up of pressure downstream from the obstruction [6].

The same phenomenon can occur in different disease processes. Intraluminal blood clots have previously been described as a cause of the ball valve effect, and subsequent pneumothorax [7]. Such clots can be managed by rigid bronchoscopy, in the same fashion as foreign bodies. Endobronchial Streptokinase has also been described in the management of tracheobronchial obstruction by blood clots [8]. Similarly, ball valving has been described in an infant secondary to a cheesy granuloma mass in the context of *Mycobacterium Tuberculosis* infection [9].

We report a unique case of a tumour causing ball valving following bronchoscopy. The procedure had caused swelling of the lesion in the postoperative period. This expanded the tumour until it obstructed the lumen of the left main bronchus during expiration, but not inspiration. The case is instructive as it shows the insidious onset of ball valving. The patient was initially asymptomatic and hence discharged. He represented after 10 hours once the gas trapping had caused enough mediastinal shift to induce chest pain, and shortness of breath.

He was treated by repeat rigid bronchoscopy which yielded a histological diagnosis of a neuroendocrine carcinoid tumour. Such tumours account for less than 5% of lung tumours [10], they are the most common type of pulmonary malignancy diagnosed in both children and adolescents. Their incidence is increasing at a rate higher than that explained by increased incidental findings [11]. One quarter of patients remain asymptomatic.

The world health organisation categorises such tumours into four subtypes. Surgery in the form of lobectomy supplemented by dissection is the treatment of choice for localised disease. Minimally invasive endobronchial procedures have been described [12]. The benefit of chemotherapy and radiotherapy remain unclear. There is less agreement on the management of more advanced tumours [13].

The prognosis in this case is unclear. A similar case of a neuroendocrine tumour managed with LASER debulking had a two year remission before localised recurrence [14].

4. Conclusions

Bronchoscopy is a safe diagnostic modality with low complication rates. However, life threatening complications are possible and the clinician should be competent in their diagnosis and management. Ball valving can be seen in the context of swollen neoplastic lesions, not just foreign bodies. Once established a ball valving lesion will cause gas trapping in the distal airway. Such cases require immediate intervention. Comparison of inspiratory and expiratory films is necessary for the diagnosis of subtle gas trapping and early mediastinal shift.

Declaration of competing interest

There is no conflict of interest for any author. This case has not been published elsewhere. Our work is original and approved by all authors.

References

- [1] S. Sinha, R. Guleria, J.N. Pande, R.M. Pandey, Bronchoscopy in adults at a tertiary care centre: indications and complications, J. Indian Med. Assoc. 102 (3) (2004) 152–154.
- [2] C.H.L. Khoong, C.K. Phua, Lung abscess and empyema following bronchoscopy: a case report and review of the literature, Respiratory Medicine Case Reports 30 (2020) 101116.
- [3] T. Tsuji, S. Sonobe, T. Koba, T. Maekura, N. Takeuchi, K. Tachibana, Systemic air embolism following diagnostic bronchoscopy, Intern. Med. 56 (7) (2017) 819–821.
- [4] V. Muthu, I.S. Sehgal, K.T. Prasad, R. Agarwal, Iatrogenic pneumothorax following vigorous suctioning of mucus plug during flexible bronchoscopy, BMJ Case Reports CP 12 (10) (2019) e230943.
- [5] C.P. Centonze, M.S. Davenport, E.S. White, E.A. Kazerooni, Routine chest radiography for the evaluation of pneumothorax following bronchoscopy, Acad. Radiol. 26 (5) (2019) 585–590.
- [6] J. Kenth, C. Ng, Foreign body airway obstruction causing a ball valve effect, JRSM Short Reports 4 (6) (2013) 2042533313482458.
- [7] J.O.H.N. Popovich Jr, R. Babcock, Intraluminal blood clot casts causing obstructive emphysema and recurrent pneumothorax, Crit. Care Med. 10 (7) (1982) 482–483
- [8] S.L. Maxwell, J.L. Stauffer, Endobronchial streptokinase for relief of tracheobronchial obstruction by blood clots, Chest 101 (6) (1992) 1738-1739.
- [9] S. Agarwal, D.K. Hong, J. Soslow, K.W. Chang, Not your routine foreign body: endobronchial tuberculosis in an infant, Pediatrics 116 (1) (2005) 246-248.
- [10] R.R. Paladugu, J.R. Benfield, H.Y. Pak, R.K. Ross, R.L. Teplitz, Bronchopulmonary Kulchitzky cell carcinomas. A new classification scheme for typical and atypical carcinoids, Cancer 55 (6) (1985) 1303–1311.
- [11] A. Petursdottir, J. Sigurdardottir, B.M. Fridriksson, A. Johnsen, H.J. Isaksson, H. Hardardottir, T. Gudbjartsson, Pulmonary carcinoid tumours: incidence, histology, and surgical outcome. A population-based study, General Thoracic and Cardiovascular Surgery 68 (2020) 523–529.
- [12] E.M.B.P. Reuling, C. Dickhoff, P.W. Plaisier, H.J. Bonjer, J.M.A. Daniels, Endobronchial and surgical treatment of pulmonary carcinoid tumors: a systematic literature review, Lung Cancer 134 (2019) 85–95.
- [13] S. Pusceddu, G.L. Russo, M. Macerelli, C. Proto, M. Vitali, D. Signorelli, M.C. Garassino, Diagnosis and management of typical and atypical lung carcinoids, Crit. Rev. Oncol. Hematol. 100 (2016) 167–176.
- [14] K. Venu, A.D. Diggikar, B.K. Rao, Carcinoid tumour: laser therapy, Indian J. Chest Dis. Allied Sci. 39 (2) (1997) 129–132.