CASE REPORT

Typical carcinoid involving the main carina managed with arterial embolisation, endobronchial resection and ablation, ultimately followed by carinal resection

Michael W Y Tan,¹ Paul Leong,^{1,2} Phillip Antippa,² Barton R Jennings¹

¹Department of Monash Lung and Sleep, Monash Health, Clayton, Victoria, Australia
²Department of Cardiothoracic Surgery, Royal Melbourne Hospital, Parkville, Victoria, Australia

Correspondence to
 Dr Michael W Y Tan,
 mwytan@live.com

Accepted 18 September 2017

SUMMARY

Bronchial carcinoids are uncommon pulmonary tumours, and the gold standard management is surgical resection. Their management is often complicated by their proximal location and propensity to bleed when manipulated. A 22-year-old man was found to have typical carcinoid tumour involving the carina and surgical resection was considered not feasible. We report our experience with a multimodality approach involving arterial embolisation, with subsequent endobronchial resection and ablation. Residual disease was found and managed with definitive carinal resection.

BACKGROUND

Bronchial carcinoid tumours are uncommon and proximal carcinoids can pose technical challenges at resection; furthermore, not all patients are surgical candidates. An emerging management modality is endobronchial resection. Experience with endobronchial resection for carcinoid is relatively limited. Our case adds to the literature in this growing field, and highlights the importance of a multidisciplinary approach to lung cancer.

CASE PRESENTATION/INVESTIGATION/TREATMENT

We report our experience with a patient whose typical carcinoid tumour involved the carina, with initial bronchial artery embolisation followed by endobronchial resection and ablation, ultimately followed by definitive carinal resection.

Carcinoid tumours often present in proximal locations. Definitive surgical resection, as extensive as pneumonectomies,¹ can therefore involve resection of large portions of healthy lung tissue. However, endobronchial manipulation of these tumours often results in bleeding.²

A 22-year-old male smoker presented with 12 months of haemoptysis, dyspnoea and wheeze. On CT an endobronchial lesion arose from the carina, nearly obstructing the right main bronchus (figure 1). Arterial phase scans demonstrated a feeding bronchial artery.

Flexible bronchoscopy revealed a broad-based lesion arising from the medial right main bronchial wall, involving the main carina (figure 2). The bronchoscope could just pass the lesion. Endobronchial biopsies demonstrated features consistent with typical carcinoid tumour with a mitotic figure count

of 1 mitosis/2 mm² with no evidence of necrosis (figure 3).

At multidisciplinary discussion in a metropolitan specialist lung cancer service, cardiothoracic surgeons, interventional pulmonologists and radiologists were consulted. No definite extrabronchial or distant disease was seen. Surgical resection was considered technically challenging with risk of pneumonectomy and substantial complications. The goal of therapy was to alleviate symptoms and prevent right main bronchus obstruction. Given surgical risk, an endobronchial approach was initially undertaken.

Bronchial arterial embolisation of the feeding vessel with polyvinyl alcohol and coils under radiological guidance was performed. The following day, the tumour was resected with an electrocautery snare via rigid bronchoscopy with no bleeding. The residual tumour bed was treated with argon plasma coagulation with an applied power of 30 W at a flow rate of 0.5 L/min in 1–2 s bursts. This was followed by thirty 10 s freeze-thaw cycles of cryotherapy via a flexible 2.4 mm Erbecryo cryoprobe.

Repeat bronchoscopy 3 weeks later demonstrated a necrotic tumour bed. Multiple endobronchial biopsies were taken and 30 additional 10 s freeze-thaw cycles were similarly applied via flexible bronchoscopy to ablate residual tumour. No tumour was identified on the biopsies.

A non-contrast enhanced CT scan at 1 month demonstrated very mild irregularity at the endobronchial site; however, 68-Gallium DOTATATE positron emission tomography (PET)/CT 3 months later showed activity in the carina without nodal or extrathoracic spread (figure 4).

The patient was referred to a quaternary multidisciplinary lung cancer service with dedicated general thoracic surgeons experienced in complex bronchoplastic procedures. Selective intubation of the left main bronchus was followed by a muscle sparing right thoracotomy and carinal resection. Initial frozen section demonstrated positive margins, therefore a subsequent right mainstem bronchus resection was undertaken to the maximal possible extent. A neocarina was formed from the remaining left and right mainstem bronchi with a running 3-0 polydioxanone suture. This was anastomosed to the trachea, and was watertight. Previous arterial embolisation did not cause any technical difficulty for resection as the coils were

CrossMark

To cite: Tan MWY, Leong P, Antippa P, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-221800

Figure 1 Contrast enhanced CT scan.

retained within feeding vessels rather than within the tumour. The patient was extubated in the recovery room and discharged 3 days later.

OUTCOME AND FOLLOW-UP

Final histology in accordance with WHO 2015³ demonstrated typical carcinoid infiltrating carina with Ki67 index of 2%, and positive neuroendocrine markers (synaptophysin, chromogranin). The tumour at the right main bronchus measured 24×5×3 mm and at the carina dimensions were 22×11×8 mm. The resection margins were free from tumour. Station 7 demonstrated 2 mm direct involvement and 10R had a 2 mm focus of disease. Follow-up multidisciplinary discussion concluded that no additional benefit would be gained from postoperative chemotherapy or radiotherapy. At present, the patient remains asymptomatic with planned surveillance CT and DOTATATE PET scans. Written consent was obtained from the patient for publication.

DISCUSSION

The best established treatment modality for carcinoid tumours is surgery,⁴ although carinal tumours may be technically demanding.⁴ Recently, multimodal intervention with sequenced

Figure 3 Forceps biopsy of carina.

embolisation and endobronchial resection has been suggested as an alternative in patients where surgery is not possible.⁵

In our case, structural intervention was necessary to prevent right main bronchus obstruction and control haemoptysis. Given the difficulty of resection due to the tumour's position, a non-surgical curative-intent approach was initially undertaken. While successful for symptom management, these interventions were not curative. Persistence was presumably due to residual tumour deep in the bronchial wall which remained untreated by diathermy and two cryotherapy sessions.

Endobronchial treatment is a second-line option to resection, as there is substantively less experience with this approach and uncertain cure rate. In Salamonsen's median 26-month follow-up of four typical and one atypical carcinoid tumour cases who underwent embolisation and endobronchial resection, there was one case of local recurrence. This was in one patient with typical carcinoid and the recurrence was successfully treated with cryotherapy.⁶ This contrasts with the gold standard of resection: during a median follow-up of 3.5 years after surgery, only 21 out of 337 (6%) patients had recurrence.⁶

We observed negligible endobronchial bleeding owing to arterial embolisation. Without embolisation, endoscopic manipulation of carcinoid tumours is complicated by bleeding in 1%–4%.⁷ Arterial embolisation is generally safe: of 88 patients in a large tertiary hospital, bronchial artery dissection occurred in 3 and

Figure 2 Endoluminal views at fibre optic bronchoscopy.

Figure 4 68-Gallium DOTATATE positron emission tomography scan.

bronchial artery spasm in 2. There were no cases of spinal cord ischaemia.⁸

Prior to planned definitive endobronchial intervention, confirmation that disease is entirely endoluminal is suggested.⁹ In our patient, it is possible that endobronchial ultrasound transbronchial needle aspirate would have missed the small tumour focus, and would not have alleviated the need to perform endobronchial resection to relieve symptoms.

Our follow-up recommendations are in line with the European Neuroendocrine Tumour Society recommendations.¹⁰ Following intervention, we propose repeat CT chest imaging at 3, 6 and 12 months for typical carcinoid tumours. Surveillance imaging should be paired with chromogranin A measurements for the first 2 years. Thereafter, we suggest annual chest X-rays and biochemistry testing with 3-year CT chest. We would increase the surveillance frequency for patients who were node positive or for resection margin-positive patients. We would perform brain imaging for metastases for cases with high tumour burden or localising symptoms. Routine bronchoscopies should occur every 5–10 years and sooner if there are symptoms suggesting local progression.

Carinal resections are infrequently performed. They are technically demanding even with careful patient selection.¹¹ Anatomically, challenges faced include dehiscence of the anastomosis site, necrosis, fistulas and granulation tissue.⁴ Postoperative mortality rates range from 3% to 20%, while morbidity rates range from 11% to 50% following carina resection.⁴ The major complication is acute respiratory distress syndrome, reported in up to 20% of cases.¹²

Learning points

- ▶ The best established treatment modality for carcinoid tumours is surgical resection.
- ▶ Embolisation, endobronchial resection and ablation are an option for patients who are not surgical candidates but require structural intervention.
- ▶ Arterial embolisation leads to negligible bleeding risk with endobronchial resection.
- ▶ Following intervention, recommendations for surveillance are for CT chest imaging at 3, 6 and 12 months for typical carcinoid tumours. Routine bronchoscopies at 5 and 10 years are recommended.

For patients who are not surgical candidates and in whom structural intervention is necessary, embolisation, endobronchial resection and ablation remain an option, although the cure rate falls below that of surgical resection. Careful attention to residual tumour is critical during ablation. Rescue systemic therapies remain viable options if the above strategy is unsuccessful.

Contributors MWYT, PL and BRJ were responsible for study concept, design and drafting of the manuscript. MWYT, PL, PA and BRJ contributed to critical revision.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

© BMJ Publishing Group Ltd (unless otherwise stated in the text of the article) 2017. All rights reserved. No commercial use is permitted unless otherwise expressly granted.

REFERENCES

- 1 El Jamal M, Nicholson AG, Goldstraw P. The feasibility of conservative resection for carcinoid tumours: is pneumonectomy ever necessary for uncomplicated cases? *Eur J Cardiothorac Surg* 2000;18:301–6.
- 2 Kurul IC, Topçu S, Taştepe I, et al. Surgery in bronchial carcinoids: experience with 83 patients. *Eur J Cardiothorac Surg* 2002;21:883–7.
- 3 Travis WD, Brambilla E, Nicholson AG, et al. The 2015 world health organization classification of lung tumors: impact of genetic, clinical and radiologic advances since the 2004 classification. *J Thorac Oncol* 2015;10:1243–60.
- 4 Weder W, Inci I. Carinal resection and sleeve pneumonectomy. *Thorac Surg Clin* 2014;24:77–83.
- 5 Salamonsen M, Tazi-Mezalek R, López-Lisbona R, et al. Combined bronchial artery embolization and endobronchial resection for bronchial carcinoid: a safety and feasibility pilot study. *Respiration* 2016;91:63–8.
- 6 Lou F, Sarkaria I, Pietanza C, et al. Recurrence of pulmonary carcinoid tumors after resection: implications for postoperative surveillance. *Ann Thorac Surg* 2013;96:1156–62.
- 7 Jüttner FM, Pinter H, Friehs G, et al. Management of severe hemorrhage after biopsy in bronchial carcinoids. *Panminerva Med* 1986;28:287–8.
- 8 Dabó H, Gomes R, Marinho A, et al. Bronchial artery embolisation in management of hemoptysis-A retrospective analysis in a tertiary university hospital. *Rev Port Pneumol* 2016;22:34–8.
- 9 Colt HG, Murgu SD, Korst RJ, et al. Follow-up and surveillance of the patient with lung cancer after curative-intent therapy: diagnosis and management of lung cancer, 3rd ed: American college of chest physicians evidence-based clinical practice guidelines. *Chest* 2013;143: e437S–54.
- 10 Caplin ME, Baudin E, Ferolla P, et al. Pulmonary neuroendocrine (carcinoid) tumors: European Neuroendocrine Tumor Society expert consensus and recommendations for best practice for typical and atypical pulmonary carcinoids. *Ann Oncol* 2015;26:1604–20.
- 11 Blasberg JD, Wright CD. Surgical considerations in tracheal and carinal resection. *Semin Cardiothorac Vasc Anesth* 2012;16:190–5.
- 12 Mitchell JD, Mathisen DJ, Wright CD, et al. Resection for bronchogenic carcinoma involving the carina: long-term results and effect of nodal status on outcome. *J Thorac Cardiovasc Surg* 2001;121:465–71.

Copyright 2017 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow