Two cases of the bronchial carcinoid tumors successfully treated with the parenchymal-sparing bronchoplastic resections

Katarzyna Drożdż^{1,2}, Mariusz Chabowski^{3,4}, Angelika Chachaj^{1,2}, Paweł Gać⁵, Małgorzata Szołkowska⁶, Dariusz Janczak^{3,4}, Andrzej Szuba^{1,2}

¹Division of Angiology, Faculty of Health Science, Wroclaw Medical University, Wroclaw, Poland; ²Department of Internal Medicine, 4th Military Teaching Hospital, Wroclaw, Poland; ³Division of Surgical Procedures, Department of Clinical Nursing, Faculty of Health Science, Wroclaw Medical University, Wroclaw, Poland; ⁴Department of Surgery, 4th Military Teaching Hospital, Wroclaw, Poland; ⁵Department of Radiology and Diagnostic Imaging, 4th Military Hospital, Wroclaw, Poland; ⁶Department of Pathology, National Research Institute of Tuberculosis and Lung Diseases, Warsaw, Poland

Correspondence to: Andrzej Szuba, MD, PhD, Professor of Medicine. Division of Angiology, Wroclaw Medical University, 4th Military Hospital in Wroclaw, Weigla 5 street; 50-981 Wroclaw, Poland. Email: szubaa@yahoo.com.

Abstract: Carcinoid tumors account for 2% of primary lung tumors. We report two cases of the relatively young patients with typical and atypical carcinoid (AC) tumors that were managed successfully with a parenchymal-sparing bronchoplastic procedure.

Keywords: Surgery; bronchial tumor; carcinoid; bronchoplasty; lung sparing procedure

Submitted Mar 31, 2017. Accepted for publication May 28, 2017. doi: 10.21037/jtd.2017.06.141

View this article at: http://dx.doi.org/10.21037/jtd.2017.06.141

Introduction

Neuroendocrine tumours (NETs) typically arise in the gastrointestinal tract, pancreas, lung and thymus. Pulmonary NETs are a distinct subset of tumors accounting for approximately 20% of all primary lung cancers (1). The 2015 World Health Organization (WHO) Lung Tumors Classification combined architectural patterns (cell size, organoid palisading rosettes, distinct nuclear/ cytoplasm ratio) with other parameters (different mitotic index, presence of necrosis), aiming at classifying pure pulmonary NETs in four different types: typical carcinoid (TC), atypical carcinoid (AC), large-cell neuroendocrine carcinoma (LCNC) and small-cell lung carcinoma (SCLC) (2). Carcinoid tumors account for 2% of primary lung tumors (2,3). Definitely more frequent occurs TC tumors (2,3). There are found mostly in the 5th and 6th decade of life, more often in women (4). AC tumors are more common in the older age group than TC (average age about 57 years), more often in men (3,4). Carcinoid tumors differ in location, size, presence of necrosis and mitotic activity (3,5,6). About 30% of patients with lung

carcinoid initially do not show any clinical signs (7). Symptoms occur sooner in patients with the central tumor bronchial location, e.g. cough (32%), hemoptysis (26%), pneumonia (24%) (3,8-10). Carcinoid syndrome is found in about 2–3% of patients (3,11). Cushing's syndrome, acromegaly, hypercalcaemia or hypoglycemia affect a small group of patients (9-11). In the diagnosis of carcinoid IHC is very important (12). The most effective, radical treatment carcinoid tumors is surgery (11,13). We present the cases of two patients with bronchial carcinoid efficiently treated with the parenchymal-sparing bronchoplastic procedures.

Case presentation

Case 1

A 28-year-old woman was admitted to the Department of the Internal Medicine of the 4th Military Hospital in Wroclaw, Poland, because of dyspnoea. Since two years she has been under the Pulmonary Clinic care with suspected bronchial asthma. There was no significant clinical improvement with applied treatment (combination



Figure 1 The CT scans of the chest have shown the left main bronchus tumor size $1.5 \text{ cm (AP)} \times 1.3 \text{ cm (TR)} \times 1.5 \text{ cm (CC)}$. AP, anteroposterior; TR, transverse; CC, cephalocaudal.

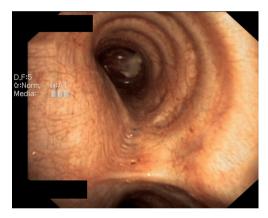


Figure 2 The smoothly contoured tumor with a vessels net, surrounded by necrotic and purulent masses located in the left main bronchus about 2.5–3 cm below the carina.

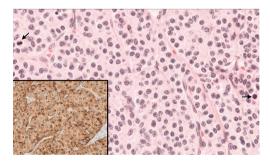


Figure 3 Atypical carcinoid. The tumor revealed similar morphology as in second case, however single mitotic figures were found (arrows). The tumor cells expressed neuroendocrine markers (insert) (H&E staining, magnification ×400, insert: chromogranin, magnification ×200).

of inhaled corticosteroids and long-acting beta-2 agonists). Because of worsening shortness of breath, including rest, the patient was referred to the hospital diagnosis. At the time of the current admission to the hospital the physical examination revealed: tachycardia 110/min and wheezing over the lungs. The patient underwent diagnostics. The routine blood tests showed no abnormalities. The arterial blood gas showed hypoxaemia. The respiratory functional study revealed the forced expiratory volume in one second (FEV₁) of 2.03 L (actual); 3.46 L (predicted normal); 58.75% (% of predicted). The moderate-to-severe degree of the obstruction with lung hyperinflation, the increased airway resistance and mild decrease in DLCO (the carbon monoxide diffusion capacity) were diagnosed. The CT scans of the chest have shown the left main bronchus tumor size 1.5 cm (AP) × 1.3 cm (TR) × 1.5 cm (CC) with one bronchopulmonary lymph node (Figure 1). The bronchoscopy has confirmed in the left main bronchus, below the carina of about 2.5-3 cm, smoothly contoured tumor with a vessels net, surrounded by necrotic and purulent masses (Figure 2). Additionally, the patient underwent octreotide scan, gastroscopy, colonoscopy and the abdominal and pelvis MRI to exclude any metastases. The levels of laboratory studies such as chromogranin A, 24-hour 5 hydroxyindoleacetic acid (5-HIAA) and tumor markers were not elevated.

The patient was qualified for surgical treatment. The sleeve bronchiectomy, i.e., the partial resection of the distal left main bronchus, with end-to-end anastomosis was performed. The intraoperative histopathological examination of the bronchial ring showed no neoplastic tissue or in the proximal or the distal margin. The final histopathology confirmed AC tumor with negative lymph nodes, G2, pT1aN0M0R0L0V0. The lymph nodes 7 (0/1), 11 (0/2) and 5 (0/2) were without tumor metastases. The IHC: NCAM (+), synaptophysin (+), chromogranin (+), index Ki-67 7%, 4/10 HPFs (*Figure 3*). The follow-up (chest CT and bronchoscopy) in the year after surgery revealed no local recurrence. The patient remains in the further observation for at least five consecutive years.

Case 2

A 44-year-old man was admitted to the Department of the Internal Medicine of the 4th Military Hospital in Wroclaw, Poland because of cough, fever and hemoptysis. At the time of the current admission to the hospital the physical examination revealed only wheezing over the lungs. The

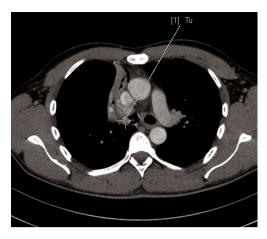


Figure 4 The CT scans of the chest have shown the right main bronchus tumor of size $1.0 \text{ cm} \times 1.1 \text{ cm}$ with nodal reaction.



Figure 5 The smoothly contoured tumor with a vessel net located in the right main bronchus protruding from the opening of the right upper bronchus.

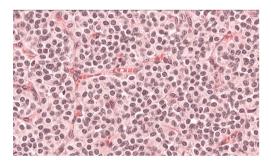


Figure 6 Typical carcinoid. The tumor was composed of uniform cells with round or polygonal nuclei and inconspicuous nucleoli. The cells were arranged in small nests separated with fine, thinwalled vessels. Neither mitotic figures nor foci of necrosis were found (H&E staining, magnification ×400).

patient underwent diagnostics. The routine blood tests showed increased markers of inflammation. The arterial blood gas showed hypoxaemia. The respiratory functional study revealed FEV1 (the forced expiratory volume in one second) of 3.00 L (actual); 3.48 L (predicted normal); 86.11% (% of predicted). The ventilatory impairments of the moderate obstruction type and the mild decrease in DLCO (the carbon monoxide diffusion capacity) were diagnosed. The total lungs capacity (TLC) and vital capacity (VC) were within normal limits. The CT scans of the chest have shown the right main bronchus tumor of size 1.0 cm × 1.1 cm with nodal reaction (Figure 4). The bronchoscopy has confirmed in the smoothly contoured tumor with a vessels net in the right main bronchus (Figure 5). The patient was qualified for sleeve right upper lobectomy with lymphadenectomy. The histopathological examination of intraoperative showed no neoplastic tissue or in the proximal or the distal margin. The final histopathology confirmed TC tumor with negative lymph nodes, G1, pT1N0M0R0L0V0, 1/10 HPFs (Figure 6). The lymph nodes 4R (0/4), 7 (0/3) and 10R (0/1) were without tumor metastases. The patient remains in the further observation for at least five consecutive years.

Discussion

In 1947 Price-Thomas performed the first sleeve resection of the bronchus to remove an adenoma originating in the right main bronchus. In 1952 Allison performed the first sleeve resection for lung cancer (14).

Bronchial carcinoids occur rarely. They comprise fewer than 3% of all lung tumors and are of low-grade malignancy (15). Chabowski *et al.* reported only 19 (5%) cases of carcinoid in two years period [1998–1999] with an excellent five-year survival rate of 94.7% (16). Kasprzyk *et al.* reported only three cases of carcinoid in five-year period [2001–2005] treated with sleeve procedures (14). Therefore, any prospective trials on bronchial carcinoids are difficult to perform.

The carcinoids are most often located centrally within the tracheobronchial tree. So, the presenting symptoms are usually related to mechanical obstruction of the bronchus by central tumors. The presence of the symptoms allows for early detection of the tumor. In our cases the symptoms were as follows: dyspnea, cough, hemoptysis and fever.

The lung saving resection, i.e., the avoiding pneumonectomy, remains the main treatment option

for carcinoids as they are resistant to chemotherapy or radiotherapy on principle (17). The complete resection with disease-free margins is mandatory. Terzi et al. claimed that the bronchoplastic procedures are successful if a frozen section of the bronchial margins is negative, which is very important to reduce the risk of local recurrence and the risk of anastomotic dehiscence (17). Sternau et al. pointed out that the bronchoplastic resections are technically more difficult and more precise to perform in comparison with traditional lobectomy (18). TCs as less aggressive can be completely resected, sparing as much unaffected lung parenchyma as possible. ACs should be treated by radical resection, as other non-small cell lung cancer, because of possible lymph node involvement (15). Approximately 20% of carcinoids present as endobronchial polyp-like lesion without involvement of lung parenchyma (19). Main bronchial resection is regarded as an appropriate technique for selected malignant lesions that are limited to the main bronchus without lymphatic involvement. In the presented cases the second patient had TC without parenchyma involvement, so only the tumor with the ring of left main bronchus was resected. The first patient had AC with parenchyma infiltration, so the sleeve right upper lobectomy with lymphadenectomy was performed. In both cases the frozen sections of the bronchial margins were negative.

The postoperative follow-up visits are very important in order to diagnose potential stenosis in the anastomosis site.

Conclusions

Conservative lung resection with bronchoplasty for bronchial carcinoid seems to be a safe procedure with low morbidity and good long-term survival. The surgeon's experience and appropriate bronchoplastic technique decrease the number of postoperative complications.

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patients for publication of this manuscript and any

accompanying images.

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Cite this article as: Drożdż K, Chabowski M, Chachaj A, Gać P, Szołkowska M, Janczak D, Szuba A. Two cases of the bronchial carcinoid tumors successfully treated with the parenchymal-sparing bronchoplastic resections. J Thorac Dis 2017;9(8):E669-E673. doi: 10.21037/jtd.2017.06.141

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