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CASE REPORT

Surgical outcomes in 13 patients with bronchopulmonary carcinoid tumors including one recurrent oncocytic carcinoid tumor

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Abstract

Introduction Bronchopulmonary carcinoids are low-grade tumors for which the standard treatment is surgical resection. We retrospectively evaluated the surgical outcomes.

Methods Thirteen patients underwent surgical resection for them at our institution between January 2005 and December 2016. We collected their clinicopathologic data to evaluate surgical outcomes.

Results The 13 patients comprised seven men and six women. Complete resection was performed in all cases. All the tumors were typical carcinoids, including one oncocytic carcinoid which showed highest fluorodeoxyglucose (FDG) uptake (SUVmax 45.7). The 5-year overall survival rates were 100%. The only patient with oncocytic carcinoid developed recurrence of liver metastasis 49 months after the primary lung resection. The metastasis showed low FDG uptake (SUVmax 2.8) and its histology was typical carcinoid and not oncocytic carcinoid.

Conclusion Surgical outcomes in our patients were favorable. In oncocytic carcinoid, metastatic site may have a radiologic and histologic appearance different from the primary tumor.

Keywords Lung · Carcinoid · Surgery · Oncocyte

Introduction

Bronchopulmonary carcinoid tumors account for approximately 1% of all primary lung tumors [1]. They are derived from Kulchitsky cells and generally have an indolent growth pattern. Carcinoid tumors are categorized into typical and atypical, based on the number of mitoses per high-power field and the presence of necrosis [2]. Surgical resection is the standard treatment for these tumors and the prognosis after complete resection is favorable [1, 3]. We retrospectively evaluated the surgical outcomes in patients with bronchopulmonary carcinoid tumors at our institution.

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Patients and methods

Thirteen patients underwent surgery for bronchopulmonary carcinoid tumors at the Shikoku Cancer Center between January 2005 and December 2016. The clinicopathologic characteristics of these patients, including age, sex, smoking status, previous malignancy, performance status, imaging, pathologic findings and TMN stage according to the eighth edition of the Lung Cancer Stage Classification, and prognostic data were collected. Pathologic diagnosis was confirmed by two pathologists in all cases. Surgical outcomes were evaluated by 5-year overall survival and disease-free survival rates using the Kaplan–Meier method.

Results

Patient characteristics

Demographic and clinical characteristics of the 13 patients are shown in Table 1. Patients comprised seven men and six women [mean age 60.3 (range 40–72) years]. Three patients

Table 1 Patient characteristics

Characteristics	$n = 13$	Range
Age, years (mean)	60.3	(48–72)
Gender, man/woman	7/6	
Smoking history, yes/no	5/8	
Previous malignancy	3	
PS, 0/1	12/1	
Imaging		
Location, right/left	7/6	
Tumor size, cm (median)	1.1	(0.5–4.5)
SUV max (median)	2.1	(0.8–45.7)
cTNM, IA/IB	11/2	
Operation		
Lobectomy	8	
Segmentectomy	2	
Wedge resection	3	
Complication	2	
Pathology		
Typical/oncocytic carcinoid	12/1	
Tumor size, cm (median)	1.1	(0.4–6)
Lymph node metastasis	0	
pTNM, IA/IB/IIB	11/1/1	
Adjuvant therapy	0	
Observation period, months (median)	67	(9–104)
Recurrence	1	

Demographic and clinical characteristics of 13 patients are shown in Table 1. All the tumors were typical carcinoids, including one oncocytic carcinoid, which had the highest maximum standardized uptake value. The patient with oncocytic carcinoid developed recurrence of liver metastasis

PS performance status, *SUV max* maximum standardized uptake value

had had previous malignancies, namely, cervical cancer, leiomyosarcoma and breast cancer (one patient each), all without recurrence. Median tumor size on computed tomography was 1.1 (range 0.5–4.5) cm and the most frequent tumor location was the right upper lobe (38.5%). Median maximum standardized uptake value (SUV max) on fluorine-18 fluorodeoxyglucose (FDG) positron emission tomography (PET) in the primary tumors was 2.05 (range 0.8–45.7). We performed eight lobectomies, two segmentectomies, and three wedge resections using a minimally invasive video-assisted approach. Complete resection was performed in all patients and there was no perioperative mortality. Prolonged air leak and seroma were documented as postoperative complications in two patients. Twelve tumors were typical carcinoids and one was an oncocytic carcinoid tumor containing a mixture of typical component. The patient with the oncocytic carcinoid was previously reported by Tanabe et al. at our institution [4]. The median pathologic tumor size was 1.1 (range 0.4–6) cm. There was no lymph node or distant metastasis. No patient received adjuvant treatment.

Prognosis

Median follow-up duration was 67 (range 9–104) months; all the patients were alive at the final follow-up. The 5-year overall survival and disease-free survival rates were 100 and 85.7%, respectively. The only recurrence was in the patient with the oncocytic carcinoid and this occurred in the liver 49 months after the primary lung resection. The FDG uptake had been high in the primary lung tumor (SUVmax 45.7), while it was low in the metastatic liver tumor (SUVmax 2.8) (Fig. 1). The patient underwent a partial hepatic resection for the metastatic tumor and pathology revealed the specimen

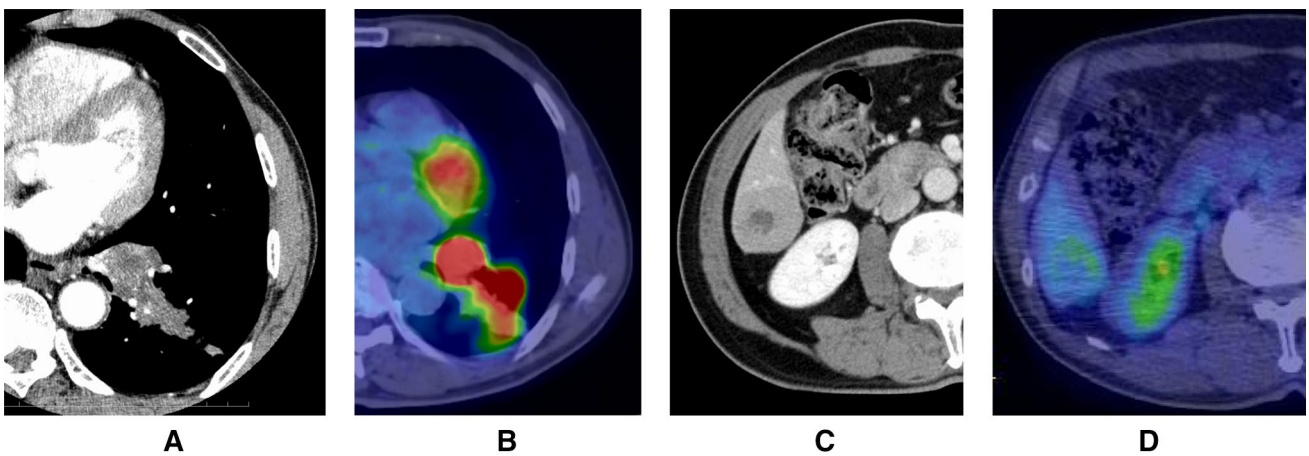

Fig. 1 Primary lung tumor and liver metastasis on positron emission tomography. Primary lung tumor (**a, b**) and liver metastasis (**c, d**) were shown on positron emission tomography. The fluorodeoxyglucose uptake was high in the primary lung tumor (SUVmax 45.7), while it was low in the metastatic liver tumor (SUVmax 2.8)

to be typical carcinoid, not oncocytic carcinoid (Fig. 2). The patient has had no further recurrences in the 16 months since. One patient developed a second primary lung cancer and another developed uterine cancer.

Discussion

The 5-year overall survival rates after the complete resection of typical and atypical bronchopulmonary carcinoid tumors have been reported to be 93.7–96 and 79–87%, respectively [1, 3]. Factors associated with an unfavorable prognosis include atypical histology, mediastinal lymph node involvement, and distant metastasis [5]. The prognosis of the patients in our study was good because all tumors had typical carcinoids without lymph node involvement or distant metastasis. A large study of 1109 patients with typical carcinoid tumors reported associations of mortality with older age, male sex, a history of malignancy, higher pathologic TNM stage and poor performance status [6]. In this study, the prediction model for mortality contains six categories, age (< 55 years: 0 point, 55–64 years: 1 point, 65–74 years: 2 points and > 75 years: 3 points), gender (male: 1 point), previous malignancies (1 point), peripheral tumor (1 point), TNM stage (I: 0 point, II: 1 point and III: 2 points) and ECOG PS (1–2: 1 point, ≥ 3: 2 points). According to these total points, the patients were stratified into four risk groups, namely, A (≤ 1 point), B (2–3 points), C (4–5 points), and D (≥ 6 points), and the respective 5-year survival rates were 99.7, 96.3, 84.2 and 53.9%. In our study, four patients were in risk group A, eight were in B, and one was in C. The patient who had recurrence scored 4 points, and so was in risk group C, which has a poorer prognosis.

Oncocytic carcinoid is a rare subtype of typical carcinoid and consists of tumor cells with eosinophilic cytoplasm resulting from the accumulation of mitochondria [7]. Oncocytic carcinoids show high FDG uptake on PET, which is believed to be related to the high glucose transmembrane protein (GLUT-1) content in these tumors [4, 8]. Expression of GLUT-1 and FDG uptake increase in pulmonary neuroendocrine tumors in proportion to their malignant behavior [8]. In our patient with oncocytic carcinoid, the primary tumor contained both oncocytic and typical components and GLUT-1 in the oncocytic component was more overexpressed than in the non-oncocytic component [4]. The recurrent metastatic liver tumor showed low FDG uptake because it contained only the typical component. This finding suggests that PET might not be useful for follow-up after surgical resection of an oncocytic carcinoid.

This study has some limitations, the main one being its retrospective design.

Conclusion

Surgical outcomes were favorable in patients with bronchopulmonary carcinoid tumors. In oncocytic carcinoid, metastatic site may have a radiologic and histologic appearance different from the primary tumor.

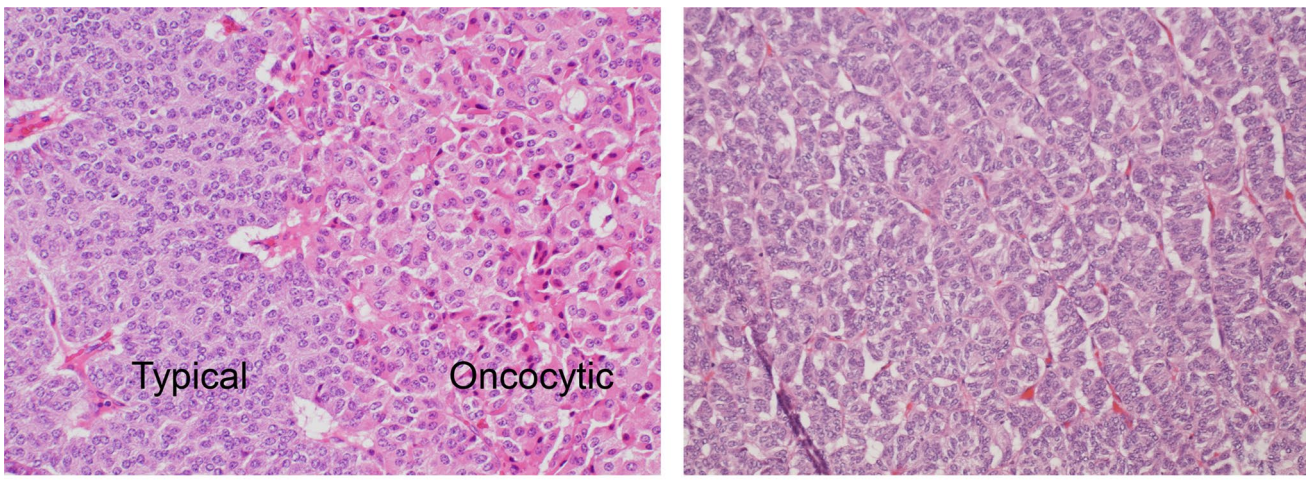

Fig. 2 Pathology of primary lung tumor and liver metastasis. The pathology of primary lung tumor (**a**) and liver metastasis (**b**) was shown. The primary lung tumor contained both oncocytic (in right side) and typical components (in left side). The liver metastasis revealed that the specimen was typical, not oncocytic carcinoid

Compliance with ethical standards

Conflict of interest The authors have declared that no conflict of interest exists.

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