CASE REPORT

Endotracheal metastasis from basaloid squamous carcinoma of the esophagus – A case report

Running Head: Endotracheal metastasis of esophageal cancer

Kazuto Harada[1], MD, Masayuki Watanabe[1], MD, PhD, FACS, Yohei Nagai[1], MD, Yoshifumi Baba[1], MD, PhD, Masaaki Iwatsuki[1], MD, PhD, Yuji Miyamoto[1], MD, PhD, Ken-ichi Iyama[2], MD, PhD, Hideo Baba[1], MD, PhD, FACS

[1]Department of Gastroenterological Surgery, and [2]Department of Pathology, Graduate School of Medical Sciences, Kumamoto University, Kumamoto, Japan.

Corresponding to: Masayuki Watanabe, MD, PhD, FACS

Department of Gastroenterological Surgery, Graduate School of Medical Sciences, Kumamoto University, 1-1-1, Honjo Kumamoto-shi, Kumamoto 860-8556, Japan

Tel.: +81-96-373-5212; Fax: +81-96-371-4378

E-mail address: masanabe@fc.kuh.kumamoto-u.ac.jp

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ABSTRACT

Endobronchial metastases from extrapulmonary neoplasms are rare events and there is no report on the metastasis originating from esophageal cancer. Basaloid squamous carcinoma of the esophagus is a rare histologic type, which is known as biologically aggressive phenotype. We describe a rare case of metachronous endotracheal metastasis originating from esophageal basaloid squamous carcinoma. A 72-year-old Japanese man underwent esophagectomy for stage I esophageal cancer. Pathological diagnosis of the resected specimen was basaloid squamous carcinoma. One year later, a follow-up computed tomography displayed a tumorshadow in the tracheal wall. Bronchoscopy revealed a protruding tumor in the tracheal wall and pathologic diagnosis of the biopsy specimen was also basaloid squamous carcinoma. Under the diagnosis of metachronous endobronchial metastasis from esophageal basaloid squamous carcinoma, we treated the patients with chemotherapy containing Docetaxel, cisplatin and 5-fluorouracil followed by chemoradiotherapy and then complete response was achieved. The patient has been alive without disease for 23 months since the diagnosis of endobronchial tumor. A careful follow-up is needed not to miss the rare type of metastasis in cases with biologically aggressive tumors.

INTRODUCTION

An endobronchial metastasis of extrapulmonary solid neoplasms is rare [1, 2]. Although various tumors have been associated with endobronchial metastasis [2], there is no report of endobronchial metastasis from esophageal origin. Basaloid squamous carcinoma of the esophagus is a rare histologic type, which is known as biologically aggressive phenotype [3]. We herein report a rare case with metachronous endotracheal metastasis from basaloid squamous carcinoma of the esophagus, successfully treated with chemotherapy followed by chemoradiotherapy.

CASE REPORT

A 72-year-old Japanese man underwent esophagectomy for clinical stage I esophageal cancer. Gross appearance of the esophageal tumor was 0-Ip+IIc type (Figure 1A). Microscopically, the polypoid tumor consisted of nests of tumor cells with round to ovoid nucleus and scanty cytoplasm, and basal membrane-like structures were observed among the tumor nests (Figure 1B, 1C). A histopathologic diagnosis was basaloid squamous carcinoma limited within the submucosal layer. Neither lymph node metastasis nor lymphatic invasion was observed while venous invasion was obvious. One year later, a follow-up computed tomography displayed a tumor shadow inside the tracheal wall (Figure 2). A bronchoscopy revealed a protruding tumor in the trachea (Figure 3). Biopsy under bronchoscopy presented solid nests of

tumor cells in the submucosal layer of the bronchial wall (Figure 4A). The biopsy specimen contained the bronchial epithelium which was free from neoplastic change (Figure 4B). The histologic feature of the bronchial tumor (Figure 4C) resembled that of the esophageal cancer (Figure 4D). Both of the specimens consisted of nests of basaloid cells with round to ovoid nucleus and scanty cytoplasm. Based on the pathologic features and the fact that primary basaloid squamous carcinoma of the lung or bronchi is also rare tumor [5], we diagnosed the tumor as endobronchial metastasis from the esophageal cancer rather than primary bronchial basaloid carcinoma. We treated the patient with systemic chemotherapy because the reported prognosis of endobronchial metastasis was poor [1, 2]. Two courses of chemotherapy with docetaxel, cisplatin and 5-fluorouracil (DCF) were performed, and significant tumor shrinkage was observed. As new lesion did not appear during the chemotherapy, a definitive chemoradiotherapy was performed. After 60 Gy irradiation combined with 2 courses of DCF, a complete response was achieved and the patient has been still alive without disease for 23 months since the detection of the endotracheal metastasis.

DISCUSSION

Metastatic tracheobronchial tumors from extrapulmonary neoplasms are rare. An autopsy study has revealed that metastatic involvement of a major airway was present in only 2% of 244 patients who died with extrapulmonary solid tumors among 1359 consecutive autopsies [1]. Sørensen et al. have collected 204 cases of endobronchial metastases originating from 20 different extrapulmonary primary tumors from 1962 through 2002 [2]. In this review, among 136 patients whose metastatic endobronchial tumors were diagnosed by bronchoscopy with biopsy, tumors located in the trachea was observed in only 8 cases (5%). These data suggest that endotracheal metastasis is an extremely rare metastasis. The common sites of primary cancers included breast, kidney and colorectal cancers [1, 2]. We have retrieved all literatures from PubMed using ‘endobronchial metastasis’ or ‘metastatic bronchial tumor’ as key words, and there was no report of endobronchial metastasis originating from esophageal cancer.

Basaloid squamous carcinoma is a rare variant of poorly differentiated squamous cell carcinoma and is commonly observed in the upper aerodigestive tract [3]. The true incidence of basaloid squamous carcinoma of the esophagus is still uncertain because of delayed recognition of this neoplasm at this location, and the reported incidences ranged from 1.9% to 11.3% [3]. Feature of this histologic type is characterized by poor differentiation and high proliferation [4]. The biologic aggressiveness may affect the occurrence of the rare type of metastasis in this case.

Differential diagnosis between endobronchial metastasis and primary bronchogenic tumor is difficult. The incidence of primary bronchogenic malignancy is very low and Bramen

et al. described that it is important to realize that the bronchial malignancy may be either a metastasis or a second primary tumor originating in the lung [1]. The demonstration of carcinoma in situ in adjacent bronchial epithelium clearly indicates primary bronchogenic tumor [1], but neoplastic change in the normal bronchial epithelium was not observed on the biopsy specimen. Sørensen et al. reported that revision of the endobronchial histologic specimen together with the histologic specimens from the extrathoracic primary tumor is of paramount importance in order to reach the correct diagnosis [2]. In the present case, very similar microscopic appearances were observed between the specimens of esophageal carcinoma and that of endobronchial tumor. As primary basaloid carcinoma of the lung or bronchi is also a rare neoplasm [5] and multiple occurrence of basaloid carcinoma within the aerodigestive tract has never been reported, we diagnosed the tracheal tumor as endobronchial metastasis from esophageal basaloid squamous carcinoma.

Metastatic pathway to the major bronchi has not been understood yet, because the incidence is very low. There are two possible metastatic pathways to the major bronchi, including the lymphatics and blood stream. Rosenblatt et al. reported that the earliest changes observed in the endobronchial metastases were permeation of the mucosal lymphatics by malignant cells and distension of the lymph channels [6]. The finding supports the possibility of lymphatic pathway. On the other hand, pulmonary metastases were observed in approximately 50% of the cases, suggesting the possibility of hematogenious metastasis [6]. In this case, neither lymph node metastasis nor lymphatic permeation was observed in the specimens of the esophageal cancer, whereas venous invasion was obvious. In addition, mediastinal lymphadenopathy was not observed in the computed tomography at the time of diagnosis of the endobronchial tumor. These findings suggest that hematogenic metastasis rather than lymphatic spread might be possible in this case.

Treatment for the endobronchial metastases in a review article included surgery, radiotherapy, chemotherapy and brachytherapy [2]. Many of cases with endobronchial metastases aimed to relieve the distressing airway symptoms instead of treatment with curative intent, because half of these cases had metastases other than the bronchus. Therefore, the mean survival time after the diagnosis of endobronchial metastasis from extrapulmonary tumors was as poor as 15 months. In the present case, the endotracheal tumor was the only malignant disease manifestation and he had no symptom of respiratory distress. Therefore, surgical resection was considered as a choice of treatment. However, because of the estimated poor prognosis, we chose induction chemotherapy with DCF, and performed CRT after confirmation of the response to chemotherapy. As a result, the tumor was successfully treated and he is still alive without disease for 18 months after the detection of the metastasis. An early detection of the recurrent tumor may contribute the better survival.

In conclusion, a careful follow-up is needed not to miss the rare type of metastasis in cases with biologically aggressive tumors. Combination of systemic chemotherapy with local treatment may achieve long-term survival in patients with solitary endobronchial metastasis.

DISCLOSURES

The authors declare that no financial or conflict of interest exists in relation to the content of the article.

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FIGURE LEGENDS

Figure 1 Pathology of the esophageal cancer. A: Gross appearance of the tumor was 0-Ip+IIc. B: The polypoid lesion consisted of basaliod squamous carcinoma and intraepithelial spread of squamous cell carcinoma was observed (X40). C: The polypoid tumor consisted of nests of tumor cells with round to ovoid nucleus and scanty cytoplasm, and basal membrane-like structures were observed among the tumor nests (X100).

Figure 2 Chest computed tomography on 1 year after esophagectomy. A tumor shadow is observed inside the tracheal wall.

Figure 3 Bronchfiber revealed a protruding tumor in the tracheal wall.

Figure 4 Pathology of biopsy from the tracheal tumor. A: Nests of tumor cells were located in the submucosal layer of the tracheal wall (X40). B: There was no neoplastic change in the bronchial epithelium (arrows, X200). C: The tracheal tumor consisted of solid nests of basaloid cells with round to ovoid nucleus and scanty cytoplasm (X200). D: Basaliod squamous carcinoma of the primary esophageal tumor (X200). Similar pathologic features were observed between C and D.