

CASE REPORT

A rare collision tumor of squamous carcinoma and small cell carcinoma in esophagus involved with separate lymph nodes: a case report

Jingpei Li^{1*}, Xiaoke Chen^{1*}, Yaxing Shen¹, Yingyong Hou², Shumin Zhang³, Hao Wang¹, Mingxiang Feng¹, Lijie Tan¹, Qun Wang¹, Zhaochong Zeng³

¹Department of Thoracic Surgery, ²Department of Pathology, ³Department of Radiotherapy, Zhongshan Hospital of Fudan University, Shanghai 200032, China

ABSTRACT

We report a case of an esophageal collision tumor composed of squamous cell carcinoma and small cell carcinoma (SmCC). A 66-year-old man complained of chest pain after oral intake for nearly one month. The patient received two cycles of neoadjuvant platinum-based combination chemotherapy and enhanced computed tomography showed a partial response of the tumor. He then underwent a thoracoscopic esophagectomy with extensive mediastinal lymphadenectomy. Two cycles of chemotherapy and prophylactic irradiation of the lymphatic drainage region were sequentially achieved after surgery. The patient has survived for more than 18 months with no evidence of recurrent disease since surgical resection.

KEYWORDS

Esophageal cancer; collision tumor; small cell carcinoma; squamous cell carcinoma

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Introduction

Esophageal squamous cell carcinoma (ESCC) accounts for 90% of the cases among Asian countries (1), while esophageal small cell carcinoma (SmCC) is a rare pathologic entity which only accounts for about 1.6% of all esophageal malignancies (2). The definition for a collision tumor is the concrescence of two neighboring independent neoplasms (3). An esophageal collision tumor of both squamous carcinoma and SmCC is extremely rare. On review of the literature, we only found one reported case of similar collision tumor which was found in the middle third of the esophagus (4). We present here with a more advanced tumor with separate lymph node involvements.

Case presentation

A 66-year-old male complained of chest pain after oral intake for about one month. Physical examination on admission revealed no pertinent abnormal findings. Serum concentrations

of carcinoembryonic antigen, squamous cell carcinoma antigen, CA19-9 and CA125 were all normal, though the neuron specific enolase (NSE) was 18.3 ng/mL (normal <15.2 ng/mL). Endoscopic findings showed an irregular hyperplastic lesion, which was accompanied with ulcer and located at the posterior wall of the distal esophagus, 37 to 41 cm from the incisor teeth. Pathological examination and immunohistochemistry of a biopsy specimen confirmed a SmCC of the esophagus with positive staining for CKpan, CD56, and negative staining for CgA and Syn. Enhanced computed tomography demonstrated a thickening and enhanced lesion of the lower esophageal wall, and no typical swelling mediastinal lymph node was found (Figure 1A). Abdominal and cervical region ultrasound and computed tomography showed no metastasis in distant organs or cervical and abdominal lymph nodes.

The patient received two courses of neoadjuvant chemotherapy which included docetaxel 120 mg/dL and oxaliplatin 200 mg/dL. His chest pain was apparently eased then. Revealed by enhanced computed tomography, the lesion decreased obviously (Figure 1B). He underwent a minimally invasive McKeown esophagectomy (thoracoscopic surgery, laparoscopic surgery and neck anastomosis) together with dissection of extensive mediastinal and abdominal lymph nodes (5) three weeks after neoadjuvant chemotherapy.

Gross examination of the resected specimen showed 1.5-cm diameter greyish red erosion (Figure 2A), and 0.4 cm diameter greyish red nodule (Figure 2B), which was located 6 cm away from the proximal edge. Histologic findings indicated

*These authors contributed equally to this paper.

Corresponding to: Lijie Tan. Department of Thoracic Surgery, Zhong Shan Hospital, Fu Dan University, 180 Feng Lin Rd, Shanghai 200032, China. Email: tan.lijie@zs-hospital.sh.cn.

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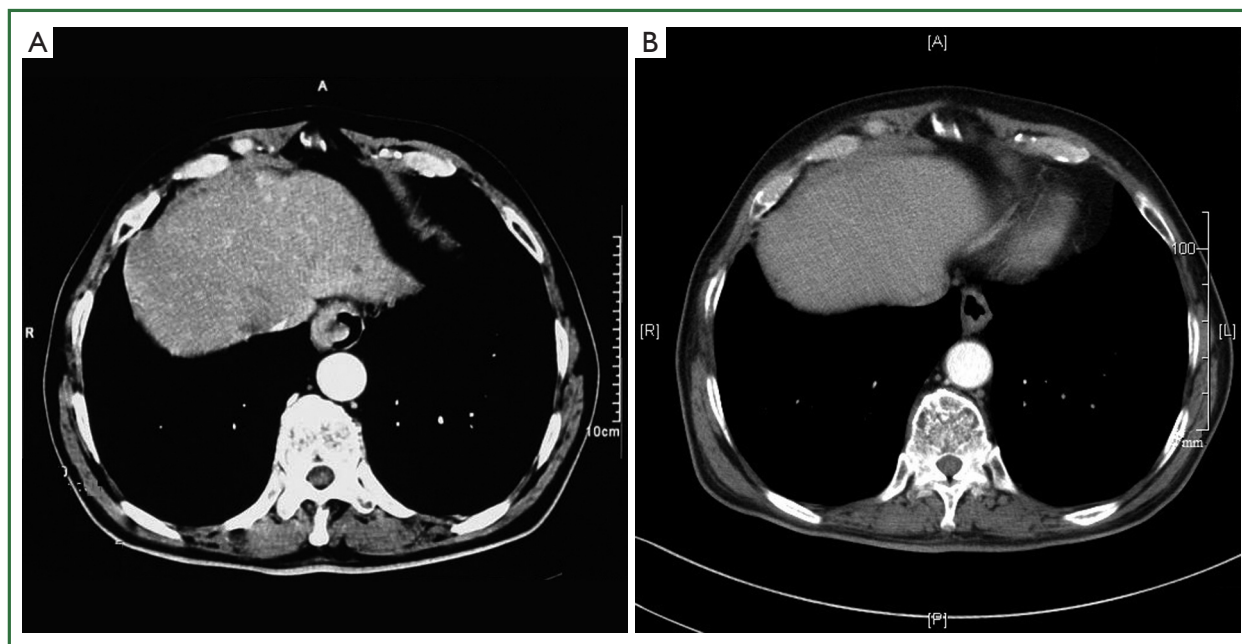


Figure 1. (A) Enhanced computed tomographic scan showing the tumor of the lower esophagus; (B) Enhanced computed tomographic scan showing a partial response of the tumor after two cycles of neoadjuvant chemotherapy.

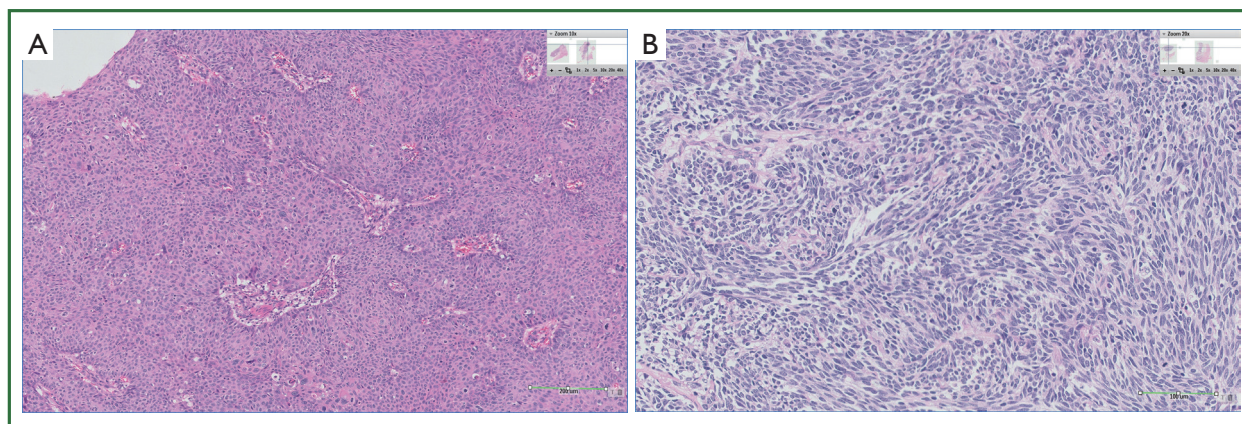


Figure 2. (A) Squamous cell carcinoma (HE; original magnification $\times 20$); (B) Small cell carcinoma (HE; original magnification $\times 20$).

that the larger erosion was squamous cancer (Figure 2A). The smaller nodule was SmCC (Figure 2B) according to the immunohistochemistry, which showed positive staining for Syn, CD56, CgA and CKpan. Depth of both tumor cell invasions was limited to the submucosa. There were as many as 38 lymph nodes harvested during the surgery which include 2 paraesophageal lymph nodes, 3 right recurrent laryngeal nerve lymph nodes, 18 surcarinal lymph nodes, and 15 paracardial lymph nodes. One of the paraesophageal lymph nodes was involved with squamous cancer (Figure 3A), while one of the paracardial lymph nodes was involved with small-cell cancer (Figure 3B). Both surgical margins were clear.

Respecting that the pathological diagnose was collision tumor as well as advanced stage, a multidiscipline discussion was

conducted. As a result, the patient continuously received two courses of chemotherapy which include docetaxel 120 mg/dL and oxaliplatin 200 mg/dL after definitive resection. And he then underwent prophylactic irradiation with no significant radiotherapeutic reaction. The total radiation dose was 40 Gy and was focused on lymphatic drainage region of the mediastinum, upper abdomen and posterior peritoneum (Figure 4). The patient led a normal life and reported no evidence of recurrence or metastasis in position emission tomography/computed tomography 18 months after surgery.

Discussion

The majority of reported esophageal collision tumors were primary

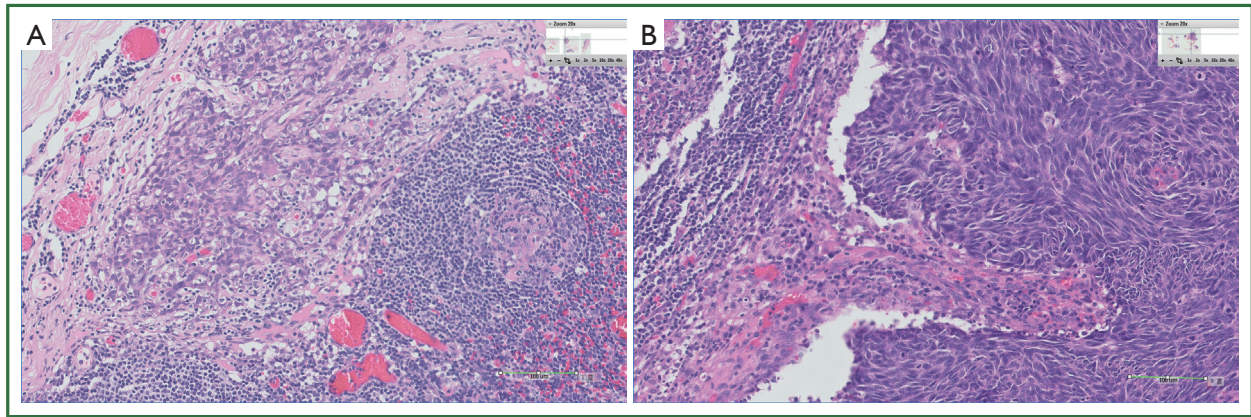


Figure 3. (A) One of the paraesophageal lymph nodes was involved with squamous cancer; (B) One of the paracardial lymph nodes was involved with small-cell cancer.

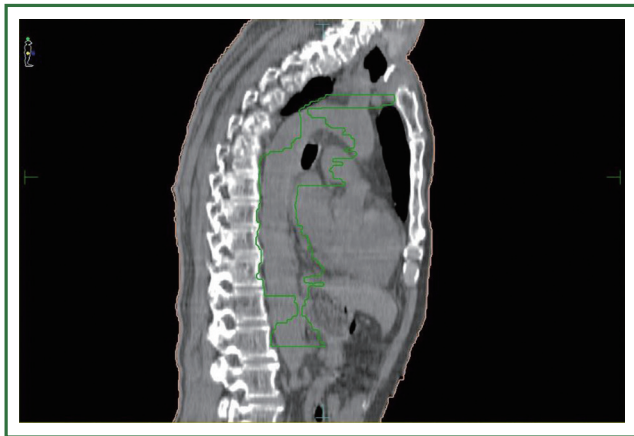


Figure 4. Prophylactic irradiation of lymphatic drainage region (within the green curve) of the mediastinum, upper abdomen and posterior peritoneum.

tumor located at the cardioesophageal junction. Examples may include adenocarcinoma and lymphoma (6), adenocarcinoma and SmCC (7), or adenocarcinoma and squamous cell (8,9), while squamous cell carcinoma plus SmCC have only been reported in one case (4). The collision tumor of our patient was a homochromous tumor of SmCC and squamous cell carcinoma at one site which metastasized to separate regions of lymph node. A similar case could not be found in the literature, suggesting that this is an extremely rare collision tumor.

The patient was primarily diagnosed as esophageal SmCC, which is a rare but highly malignant disease characterized by early dissemination and poor prognosis. Although the standard treatment protocol for such aggressive disease has not been established, most patients were treated by surgical resection, chemotherapy, or radiotherapy, whether alone or in combination (2,10-12). Recently, several cases and literature review suggested that using platinum-based polychemotherapy as initial treatment may result in survival benefits (4,13). Since

2005, more than two thousands patients from our department of thoracic surgery have generally received chemotherapy which included docetaxel and oxaliplatin, and complete response was also recorded among those patients. As for this case, after two cycles of neoadjuvant chemotherapy, the patient claimed an eased chest pain, so was shown in computed tomography with tumor regression (Figure 1B).

Many if not most studies indicated that patients with esophageal SmCC treated with surgery following chemotherapy and/or radiotherapy had been reported to survive longer than those treated with chemotherapy and/or radiotherapy (11,12,14). Therefore, a radical surgery together with dissection of extensive mediastinal and abdominal lymph nodes was applied which led to the upstaging of TNM (5). The surgical procedure was, however, difficult due to interfusion of esophagus and membranous tracheae, which was mainly caused by neoadjuvant chemotherapy. Hence, we dissected the esophagus far from the normal part and excised the abscess along with the esophagus and the tracheal membranous part was carefully preserved.

The postoperative recovery was uneventful, however, interesting things happened, that the pathological findings of the resected specimen revealed a collision carcinoma. Depending on the positive effect of neoadjuvant chemotherapy and well recovery after surgery, the patient received two more cycles of chemotherapy. Based on the survival benefits of radiotherapy for SmCC (11), postoperative radiotherapy was also applied to improve the local control rate in considering of lymph node involvements in both lesions.

In conclusion, we reported a rarely seen case of collision tumor, for which we treated with neoadjuvant chemotherapy, radical esophagectomy, adjuvant chemoradiotherapy sequentially. The patient led a normal life in 18 months follow-up. With our limited experience, we recommend multidisciplinary therapy depending on the patient's general condition in treating with esophageal collision tumor.

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