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Unusual Late Life-Threatening Arterial Bleeding after Salvage Total Laryngectomy Following Chemotherapy and Radiotherapy

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> 항암 방사선 치료 후 시행한 구제 후두 전적출술 후에 발생한 생명을 위협하는 특이한 지연 동맥 출혈

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= 국 문 초 록 =

본방사선 치료 후에 후두 절제술을 시행할 경우 인두피부누공, 감염, 출혈 등 상처부위 합병증의 가능성이 높다. 저자들은 최근에 항암치료와 방사선 치료 후에 재발한 성문상부암에 대해 후두 전적출술을 시행하였고, 수술 66일 째 생명을 위협하는 인두의 동맥 출혈을 경험하였기에 문헌고찰과 함께 보고하는 바이다.

중심 단어 : 후두절제술 · 방사선치료 · 출혈.

Introduction

Concurrent chemoradiotherapy(CCRT) has become more widely used for organ preservation in cases of advanced laryngeal cancer.¹⁾ However, total laryngectomy is still required as salvage treatment when residual or recurrent disease is present.²⁾ Preoperative radiation affects wound healing and increases the risk of developing wound complications.³⁾ Here we report a case of unusual life-threatening arterial bleeding from the pharynx on the 66th day after salvage total laryngectomy following induction chemotherapy and CCRT.

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Case Report

A 66-year-old male presented with hoarseness that had occurred for over 1 month. The patient had been diagnosed with well-controlled hypertension six months previously. The patient had a history of smoking(13 pack-years), and reported moderate alcohol consumption. Flexible laryngoscopic examination and computed tomography(CT) revealed an irregular exophytic mass on the left false vocal fold that extended to the left true vocal fold, left vocal process, paraglottic space, epiglottis, and preepiglottic space. No neck mass was found. The initial diagnosis was supraglottic carcinoma(graded as T3N0M0). Induction chemotherapy including three cycles of docetaxel, cisplatin, and 5-FU was administered to the patient. The response to induction chemotherapy was partial remission with a 90% decrease in the primary tumor. The patient underwent CCRT(radiotherapy of 6840cGy delivered to the larynx, bilateral neck, and supraclavicular area and intravenous administration of cisplatin). After completion of CCRT, there was no

evidence of disease. The patient remained healthy with no signs of cancer recurrence during the 4-year follow-up physical and radiologic examinations.

Four years after the CCRT, regular laryngoscopic evaluation found disease recurrence on the left true vocal fold that extended into the subglottis and left false vocal fold(Fig. 1). The CT scan showed no neck metastasis, and a carcinoma on the left true vocal cord that extended to subglottis. Histopathologic examination of the laryngeal biopsy identified squamous cell carcinoma(graded as T3N0M0). The patient underwent total laryngectomy, bilateral neck dissection, left thyroid lobectomy, and left paratracheal lymph node dissection to treat the recurrent carcinoma of the larynx(Fig. 2). The surgical procedure and initial recovery were uneventful. According to the histopathologic report, all resection margins were free from carcinoma. And there was noted no neck lymph node metastasis.



Fig. 1. Flexible laryngoscopy showed recurrent lesions on the left true vocal fold.



Fig. 2. The operation field after total laryngectomy with bilateral neck dissection was unremarkable.

After gastrographic swallow study revealed no evidence of leakage around the pharyngeal closure, the patient resumed oral feeding postoperative 14 days later. Sixty-six days after the operation, the patient developed acute bleeding from the nasal cavity. The bleeding was quickly stopped. There was no bleeding focus in the nasal cavity. Eight hours later, the patient developed fresh hematemesis. Urgent gastroscopy was performed, but failed to identify the site of bleeding. The patient did not have any pulsating neck swelling, thrill, or bruit. However, the patient continued to experience massive bleeding from the nasal and oral cavities. The surgical site was immediately examined with the patient under general anesthesia. We first performed pharyngoscopic approach. And we thought the bleeding was occurred from left pharyngeal wall. We contacted the interventional radiologists, but they were not available at that moment to examine the patient. Unavoidably, we performed external approach, and explored the left neck and examined carotid artery and branches. But, we failed to identify the site of bleeding. Secondly we examined the right neck. The bleeding was found to have originated from the right lingual artery. Branches of the right external carotid artery including the lingual artery were ligated. The tissues of arteries, adjacent pharvngeal mucosa, and surrounding structures were inflamed, friable, necrotic, and easily bled when touched. Despite our treatment, the patient developed severe hypotension, disseminated intravascular coagulation(DIC) and multi-organ failure. The family of the patient wanted a DNR(Do not resuscitate) and no further procedure or surgery. The patient finally died the next day.

Discussion

Organ preservation treatment is widely administered to patients with advanced laryngeal cancer. Despite the efficacy of CCRT for treating laryngeal cancer,¹⁻³⁾ a considerable number of patients still require salvage total laryngectomy due to persistent or recurrent disease. However, increases in wound complications, such as pharyngocutaneous fistula development, wound infection, or bleeding are observed in patients who undergo salvage laryngectomy following chemotherapy and radiation thearpy.⁴⁾

Patients who had radiotherapy for laryngeal cancer are prone to developing multifocal iatrogenic arteriopathy in the radiation field.⁵⁾ In addition, surgery, malignancy, infection, or poor nutrition are factors that predispose patients to blood vessels damages.⁵⁾ The patient in our case had malignant disease, had previous received radiotherapy, and underwent surgery. Therefore, the patient had a high risk for developing postoperative arterial bleeding.

Post-operative arterial bleeding after salvage total laryngectomy in patients who have received radiation therapy and/ or chemotherapy is diagnosed on the basis of the patient's history and physical examination. However, if the patient has a massive bleeding from the nasal and oral cavities without pulsating neck swelling, thrill, or bruit, it is very difficult to immediately identify the site of bleeding. In our case, we performed nasal endoscopy, pharyngoscopy and gastroscopy, but we did not find the bleeding site. Ultrasonography of the neck was not performed because there was no palpable pulsatile neck mass, bruit, or thrill. Additionally, we could not perform angiography because the interventional radiologist was unavailable. We finally found the site of bleeding during surgical exploration. However, by this time it was too late to save our patient's life.

After the patient died, the authors reviewed the hospital course and thought about the reason of bleeding. Actually, on about the fifty days after operation, focal floatness of neck flap on right submentum was observed by another doctor (supervisor) on in-patient rounding. However, we ignored this information because there was not any evidence of infection and any particular symptoms. If this was related to the cause of bleeding, radiotherapy contributed partial dehiscence in pharyngeal closure postoperatively(around fifty days) and led to rupture of arterial wall.

There are two approaches for hemorrhage management in patients who have received chemotherapy and radiation therapy : endovascular intervention and surgical exploration.^{5,6)} Endovascular repair offers the advantage of allowing vessel preservation. However, if infection is found or suspected, early surgical exploration may be preferable to endovascular intervention.⁶⁾ The choice of management strategy depends on the clinical presentation and urgency of the situation. Furthermore, the use of vascularized reconstruction flaps has

been recommended to reduce the rate of wound complications after salvage laryngectomy following chemotherapy and radiation thearpy.⁴⁾

Late life-threatening arterial bleeding after salvage total laryngectomy following chemotherapy and radiation therapy is a rare complication. However, awareness of this condition is necessary for effective management. According to the clinical presentation and urgency of the situation, appropriate management procedures should be executed immediately to treat these patients.

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