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CASE REPORT

Carotid artery ligation via sternotomy as a palliative surgery: Case report of advanced intramediastinal malignant soft tissue tumor

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1 | INTRODUCTION

The extent of surgical risk that can be tolerated in a palliative setting remains controversial. We present a case in which emergent carotid artery ligation via sternotomy was performed due to imminent carotid blow out. Fortunately, the patient was still alive at home 10 months postoperatively without any sequelae.

The extent of surgical risk that can be tolerated in a palliative setting remains debated. Generally, the surgical indication depends on the risks and benefits, and noncurative extensive surgery basically is contraindicated. However, the validity of palliative surgery has been of increasing interest in recent research and the indications for each procedure are being refined.¹

Highly advanced head and neck tumors often have a risk of carotid blow out syndrome (CBS), which is a fatal

Abstract

Under extreme conditions and in palliative settings, shared decision making with the patient is vital; narrative decisions beyond evidence could be considered. If there is a chance of symptom palliation, extended surgery should not be avoided merely because of the limited life expectancy.

KEYWORDS

carotid blow out syndrome, malignant Triton tumor, palliative care, palliative surgery, sternotomy

complication of head and neck tumors. Prevention often requires carotid artery sacrifice with related neurologic risks.² Because of the potential devastating complication and urgency of the situation, no uniform suggestion exists for management of CBS, and highly individualized and rapid decisions are needed.

We report on a case of locally advanced malignant soft tissue tumor of the upper mediastinum, suspicious for malignant Triton tumor, invading the esophagus and left common carotid artery. Pseudoaneurysm formation and massive hematemesis occurred during palliative care. From our surgical experience with this case, we discussed the indication for palliative surgery. This report is in accordance with our local ethics protocol, which is approved by our institutional ethics review board (2018-179), and written informed consent was obtained from the patient.

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2 | CASE PRESENTATION

A 32-year-old woman presented elsewhere with a 2-month history of back pain and cervical swelling. Needle biopsy demonstrated spindle-shaped neoplastic cells that were fusiform and wavy with a severely atypical nucleus. Mitoses were abundant, and necroses were scant. Immunohistochemistry revealed tumor cells positive for CD99, HHF-35, CD34, desmin, myogenin, and myo D1, and negative for S-100, EMA, STST6, MUC4, MDM2, CDK4, and TLE1. Chimera genes of SYT-SSX1 and SYT-SSX2 were negative on realtime polymerase chain reaction testing. According to these findings, a malignant peripheral nerve sheath tumor with rhabdomyoblastic differentiation (malignant Triton tumor) was suspected. On fiberscopic and radiographic studies, the tumor invaded the esophagus, tracheal membranous mucosa, bilateral recurrent nerve, and major mediastinal vessels. The tumor was diagnosed unresectable and progressed after two courses of doxorubicin and 61 Gy of radiotherapy followed by an additional five courses of eribulin. Gastrostomy was performed after oral feeding was discontinued due to esophageal perforation and mediastinitis.

The patient presented to our institute 10 months after initial diagnosis, seeking for further treatment. She was informed of her severe condition and agreed to palliative care. After hospitalization, massive hematemesis occurred and computed tomography (CT) revealed bleeding from a left common carotid artery pseudoaneurysm (Figure 1A). Radiologists did not recommend radiologic intervention, so surgical ligation of the common carotid artery without preoperative neurologic evaluation was required. Due to massive mediastinal tumor invasion, sternotomy was necessary. Despite bilateral vocal cord palsy, tracheostomy was impossible because the tumor covered the trachea and the risk of long-term intubation was unavoidable.

After consulting our institutional medical safety official, the patient and her family were informed of the neurologic concerns, risk of CBS due to surgical maneuvers, and hemodynamic changes, airway risks, and potential intraoperative death. The patient strongly requested the procedure, and surgery was performed the following morning.

With the patient under general anesthesia, an L-shaped sternotomy was performed, exposing the upper mediastinum. The proximal side of the left common carotid artery was identified caudal to the tumor and ligated. The external and internal carotid arteries were identified, and the distal side of the left common carotid artery was ligated caudal to the carotid bifurcation. Total operation time was 158 minutes, with 57 mL blood loss (Figure 2).

Fortunately, she was extubated successfully immediately postoperatively and observed in the intensive care unit. No postoperative complication was seen. Neurologic sequelae were absent and hematemesis resolved postoperatively. CT on postoperative day 7 revealed complete disappearance of the pseudoaneurysm (Figure 1B). Left internal carotid artery blood flow was speculated to be supplied by back-flow from the external carotid artery (Figure 3). She was discharged from the hospital on postoperative day 16. Palliative care was continued at home by local medical staff. She was alive at our last follow-up 10 months postoperatively, enjoying her last days at home with her family.

3 | **DISCUSSION**

Our patient was receiving palliative care due to an unresectable upper mediastinal malignancy, suspicious for malignant Triton tumor. Prognostic evaluation in our patient was difficult due to the limited diagnostic evidence. Despite limited time and insufficient evidence for the disease, palliative surgery was conducted according to the patient's strong request.

The role of surgery in a palliative setting remains debated and has been of increasing interest in recent research. For palliative care, major surgery for advanced malignancy has



FIGURE 1 Pseudoaneurysm of the left common carotid artery. A, Preoperative coronal CT image shows a pseudoaneurysm of the left common carotid artery caused by tumor invasion. B, Coronal CT image on postoperative day 7 demonstrates successful disappearance of the pseudoaneurysm



FIGURE 2 Intraoperative views. A, Skin incisions are outlined, showing a left hockey stick incision of the neck with median incision of the thorax. B, The left common carotid artery is identified on the proximal and distal side of the tumor and pseudoaneurysm. C, The left common carotid artery is ligated with 2-0 and 3-0 silk

FIGURE 3 Blood flow of the left carotid artery. A, Preoperative sagittal CT image shows blood flow of the entire left carotid artery along with the pseudoaneurysm. B, Sagittal CT image on postoperative day 7 shows disappearance of left common carotid artery blood flow due to ligation. The blood flow to the left internal carotid artery appears to be supplied by back-flow from the left external carotid artery



been reported to result in good symptomatic relief.¹ Reports in the literature support its benefits if there are a few months of life expectancy.³ Surgical indication is assessed individually according to the risks and benefits. Regarding malignant Triton tumors in general, reported poor survival rates at 2 and 5 years were 15%-33% and 11%-12%, respectively.⁴⁻⁶ However, evidence of such tumors originating in the mediastinum is scarce.⁷ As in our case, vital structures, such as major vessels, tend to be invaded at diagnosis. Life expectancy could have been shorter than anticipated due to the risk of sudden death caused by major vessel collapse.

CBS is a catastrophic condition with a reported mortality rate of 60%.² The two major treatment methods are radiologic intervention and surgical ligation. Although each has its own advantages and disadvantages, both procedures require carotid artery sacrifice with potential hazardous sequelae.⁸⁻¹¹ In our case, radiologic intervention was contraindicated because of concerns of deterioration causing CBS due to the procedure. Surgical ligation was required without preoperative evaluation of neurologic risks with the balloon occlusion test. The intraoperative carotid artery clamping test with the patient under local anesthesia, proposed by Matsumoto et al,² was contraindicated. The procedure had to be performed under general anesthesia since ligation of the proximal side of the common carotid artery required sternotomy. Because our patient already was receiving palliative care, with refusal of resuscitation, the tolerability of the surgery was questioned. WII FY_Clinical Case Reports

Promotion of end-of-life care is an important phase of palliative care and, therefore, requires extra effort. However, this is often challenging in symptomatic patients with highly advanced head and neck malignancies, since the esthetic and functional burden along with pain and fetor can be devastating. Our patient had acute bleeding and faced the risk of sudden death. After urgent and careful communication, she strongly requested the surgery. Chan et al reported that six of eight patients (75%) who underwent carotid artery ligation successfully found their place of death outside the hospital.¹² Because CBS is an extreme condition with presumed sudden death, its management could resemble a form of end-of-life care, regardless of the risks. The surgical burden of additional L-shaped sternotomy is acceptable, and the procedure could be performed by head and neck surgeons alone. We believe that sternotomy should not be avoided merely because of its radical nature. As in our case, carotid artery ligation via sternotomy was an adequate procedure, perfectly meeting the needs of the patient.

4 | CONCLUSION

We report a case of a locally advanced intramediastinal malignant soft tissue tumor, in which carotid artery ligation via sternotomy was performed for palliation. Shared decision making and precise planning were essential.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Kohtaro Eguchi: main contributor and the first author and contributed substantially in the conception and design of the work, and in writing the manuscript. Kenya Kobayashi: main contributor and the corresponding author and contributed substantially in the conception and design of the work, and in writing the manuscript. Tomonari Takano: contributed substantially in revising the manuscript. Akiko Ito: contributed substantially in revising the manuscript. Azusa Sakai: contributed substantially in revising the manuscript. Atsuo Ikeda: contributed substantially in revising the manuscript. Yoshifumi Matsumoto: contributed substantially in revising the manuscript. Go Omura: contributed substantially in revising the manuscript. Fumihiko Matsumoto: contributed substantially in revising the manuscript. Seiichi Yoshimoto: main contributor and the last author and contributed substantially in the conception and design of the work, and in writing the manuscript.

ETHICAL STATEMENT

Written informed consent was obtained from the patient for publication of the report.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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