Distal Marginal Stenosis: A Contributing Factor in Delayed Carotid Occlusion of a Patient With Carotid Blowout Syndrome Treated With Stent Grafts

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Distal marginal stenosis is rarely reported to be a factor associated with poor long-term patency of patients of head and neck cancers with carotid blowout syndrome treated with stent grafts. We report a case of laryngeal cancer with rupture of the right common carotid artery. A self-expandable stent graft was deployed, but bleeding recurred. Another stent graft was deployed for the pseudoaneurysm located distal to the first stent graft. Rebleeding occurred because of pseudoaneurysm formation from reconstituted branches of the right superior thyroid artery. We performed direct percutaneous puncture of the proximal superior thyroid artery for successful embolization. Distal marginal stenosis and asymptomatic thrombosis of the carotid artery were noted at 3.5- and 5-month follow-ups, respectively. We suggest aggressive early follow-up and reintervention for distal marginal stenosis by combined antibiotic therapy and angioplasty and stenting to improve the long-term patency of stent-graft deployment for management of carotid blowout syndrome. [*J Chin Med* Assoc 2010;73(5):271–274]

Key Words: carotid blowout syndrome, distal marginal stenosis, stent graft

Introduction

Carotid blowout syndrome (CBS) refers to rupture of the carotid artery.¹⁻⁴ It is a devastating complication associated with radical head and neck surgery. The reported incidence of carotid rupture after radical neck dissection is 4.3%.^{4,5} Emergency surgical ligation of the diseased carotid artery results in an average mortality rate of 40% and an average major neurologic morbidity rate of 60%.^{1,4} Endovascular therapy with permanent balloon occlusion has been reported to be an excellent alternative for management.¹⁻⁴ For patients at high risk of cerebral ischemia by carotid occlusion, endovascular reconstruction with stent grafts is reportedly a viable alternative management.⁵⁻⁷ However, such endovascular management may be associated with various delayed complications such as carotid artery thrombosis, brain abscess formation or rebleeding.^{5,7,8}

We report the complex delayed complications of a patient with laryngeal cancer associated with CBS who had received a self-expandable stent graft in the carotid artery. Delayed complications after this reconstructive management included 2 rebleeding episodes from a pathological process extending beyond the margin of the stent graft and from the collaterals, distal marginal stenosis of the carotid artery, and asymptomatic septic thrombosis of the stent grafts. Reintervention with deployment of a second stent graft and direct percutaneous puncture of the branch of the external carotid artery (ECA) was successful in achieving hemostasis in our patient. Distal marginal stenosis is a contributing cause of delayed carotid artery occlusion that is rarely recognized.



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Case Report

A 66-year-old man had received the diagnosis of laryngeal cancer (stage T1N1M0) 4 years before this presentation. He had received definitive conventional radiotherapy with a total dose of 7,000 cGy to the larynx and neck region. The tumor was in complete remission after radiation therapy. The patient was admitted to our hospital because of bleeding from his mouth and tracheostomy. Physical examination showed swollen soft tissue on the right side of his neck. Laboratory tests showed a white blood cell count of 11,400 cells/mm³ (segments, 85%), a platelet count of 251,000 cells/mm³, and hemoglobin level of 9.8 g/dL.

Another episode of uncontrollable active bleeding from the tracheostomy with hypovolemic shock status

was observed, and emergency angiography showed a pseudoaneurysm in the right distal common carotid artery (Figure 1A). Emergent antithrombotic medications were also given.⁷ An 8 × 50-mm self-expandable stent graft (WallGraft; Boston Scientific, Natick, MA, USA) was deployed from the right proximal internal carotid artery (ICA) to the common carotid artery. The active extravasation was stopped immediately. One and a half months later, bleeding from the tracheostomy recurred. Angiography revealed pseudoaneurysm formation in the right carotid bulb just distal to the previous stenting site. Because of suspected propagating radiation necrosis with rebleeding, a second 8×50mm, self-expandable stent graft (WallGraft; Boston Scientific) was deployed in the right ICA, overlapping the first one (Figure 1B). It stopped the active bleeding.



Figure 1. (A) Selective angiogram of the right common carotid artery (CCA), right anterior oblique view. Pseudoaneurysm in the right distal CCA (arrow). (B) Selective angiogram of the right CCA, lateral view. Two self-expandable stent grafts were deployed from the right internal carotid artery to the right CCA. (C) A right subclavian angiogram shows a pseudoaneurysm (arrow) in a branch of the right superior thyroid artery reconstituted from branches of the ipsilateral costocervical trunk via the ipsilateral external carotid artery. Direct percutaneous puncture of the orifice of the right superior thyroid artery with a spinal needle (arrowheads) was carried out for embolization. (D) Selective angiogram of the right CCA, lateral view. Distal marginal stenosis (arrowheads) was observed in the distal cervical internal carotid artery. (E) Contrast-enhanced axial computed tomography scan of the neck. Septic thrombosis of the stent graft of the right carotid artery with gas collection (arrow) was noted. Adjacent necrotic soft tissue was also found.

Since the contaminated wound was located in the soft tissue of the right neck region, we gave empirical prophylactic antibiotics (ampicillin and gentamicin) for 1 month.

Recurrent active bleeding from tracheostomy was noted 1 month after this second stent graft was deployed. Emergency subclavian angiography demonstrated a pseudoaneurysm in the branches of the right superior thyroid artery (Figure 1C). Numerous collaterals from the right costocervical trunk anastomosed with the branches of the right ECA and fed the pseudoaneurysm. Because the branches of the right costocervical trunk may anastomose with the right vertebral artery, embolization of branches of the right costocervical trunk posed a risk of stroke. To target the source of bleeding, we performed direct percutaneous puncture of the orifice of the right superior thyroid artery using a 21-gauge spinal needle (Figure 1D). The pseudoaneurysm was embolized by slowly injecting 25% N-butyl cyanoacrylate (Histoacryl; B. Braun Melsungen AG, Melsungen, Germany) and 75% lipiodol oil through the spinal needle. Active extravasation stopped immediately after embolization.

A follow-up angiogram 1 month after the final treatment showed a long-segmental stenosis in the cervical ICA distal to the end of the deployed stent grafts (Figure 1E). We closely observed the patient because he was asymptomatic. A computed tomography scan of the head and neck region 1.5 months later revealed septic thrombosis of the stent grafts with occlusion of the right carotid artery. The patient was alive without rebleeding during a 2.5-year follow-up.

Discussion

CBS is clinically classified into 3 types: threatened, impending, and acute.^{1,3} Patients with acute CBS have uncontrollable acute hemorrhage caused by complete rupture of the diseased carotid artery. Our patient had acute CBS, for which the first stent graft was deployed. Endovascular techniques, such as permanent balloon occlusion, are efficient in arresting hemorrhage and have substantially improved patients' outcomes.^{1,4} However, in as many as 15–20% of patients with CBS treated with permanent balloon occlusion, immediate or delayed cerebral ischemia develops.^{1,3,6} A balloon occlusion test is usually not possible in acute cases.

Endovascular stent grafts are useful in patients at high risk for occlusion of the carotid artery.⁶ In our case, deployment of stent grafts achieved only immediate and short-term hemostasis. This result indicates that stent grafts have a limited role in achieving durable hemostasis in the management of CBS in patients with head and neck cancers.^{4,5,7} Recurrent extravasation from the branches of the ECA of our case also favored the associated embolization of the ipsilateral ECA and its branches before deployment of a stent graft to treat CBS if the pathological lesions are close to the carotid bifurcation.⁷ This embolization can be carried out by simple deployment of fiber coils in the main trunk of the proximal ECA. In conjunction with the occlusion of the orifice of the ECA by the stent graft, complete thrombosis of the ECA close to the carotid bifurcation can be achieved.

Distal marginal stenosis is rarely reported as a complication of stent-graft deployment in patients with CBS.⁹ We speculate that the stenosis in our case was caused by vascular remodeling due to a high radial force and the shortening character of the continuously braided self-expandable stent grafts (WallGraft; Boston Scientific).^{10,11} The smaller diameter of the carotid artery in the distal side of the stent graft may suffer from stronger pressure than its proximal side and thus cause distal marginal stenosis. For our patient, the 2 overlapped stent grafts covered a long segmental carotid artery that created a strong vascular remodeling effect at their ends. This stenotic lesion could be differentiated from severe atherosclerotic stenosis by its rapidly progressive change, a well-patent ICA distal to it, and no other atherosclerotic lesions in the head and neck arteries on angiography. Since the lesion showed a rapid temporal change (3.5 months after the initial management), we suggested a close follow-up and early reintervention by angioplasty as well as stenting to prevent carotid artery occlusion. A follow-up imaging study, such as computed tomography angiography every 3 months in the first post-procedural year, may be useful to detect this delayed complication of stent-graft placement. We suggest aggressive management of this delayed complication with angioplasty and stenting. However, a better design of stent graft with lower radial force and better conformity is needed.

The use of foreign material in a field with contaminated necrosis may result in localization and persistence of infection as a result of colonization of the foreign material by infective organisms.¹² Although we gave prophylactic antibiotics, our patient suffered from asymptomatic septic thrombosis of the stent graft. It is possible that the cause of carotid artery occlusion of our patient was due to distal marginal stenosis rather than the septic process. For septic thrombosis of the stent graft without associated distal marginal stenosis and carotid occlusion, the patient may have complications with intracranial infectious processes such as brain abscess formation.⁸ The rapid progression of the distal marginal stenosis of our patient contributed to carotid artery occlusion and protected the patient from intracranial infectious processes. This implies that a longer duration of antibiotic use until the infected wound stabilizes is important in the management of distal marginal stenosis by angioplasty and stenting.

In conclusion, application of self-expandable stent grafts for managing patients with head and neck cancers associated with CBS can be associated with complex delayed complications such as rebleeding, distal marginal stenosis and septic carotid thrombosis. We suggest close follow-up of patients after stent-graft deployment. Combined antibiotic treatment and angioplasty and stenting for distal marginal stenosis are important to improve long-term carotid artery patency.

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