Massive Epistaxis from Internal Carotid Pseudoaneurysm during Acute Ischemic Stroke in a Patient with Nasopharyngeal Carcinoma

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Abstract-

Purpose: Carotid blowout syndrome due to rupture of internal carotid artery pseudoaneurysm in NPC patients with prior neck radiation is an uncommon but life-threatening complication. Concomitant carotid stenosis with ischemic stroke and carotid rupture from pseudoaneurysm is rare.

Case Report: A 71-year-old man had a history of NPC treated with radiation therapy 26 years ago. He was admitted to the hospital because of minor ischemic stroke and tarry stool. The carotid duplex sonography disclosed severe stenotic lesion in the proximal right internal carotid artery. A subsequent recurrent stroke on day three associated with nasal cavity bleeding resulted in an endotracheal intubation. Another episodic of massive epistaxis occurred on day 10 caused hypovolemic shock. Pseudoaneurysm of the left internal carotid artery was found by emergent angiography and was immediately obliterated by endovascular treatment with microcoils and glue.

Conclusion: Carotid blowout syndrome in NPC patients during acute ischemic stroke warrants further cervical angiographic study. Endovascular treatment provides immediate hemostasis and obliteration of ICA pseudoaneurysm.

Key Words: carotid blowout syndrome, nasopharyngeal carcinoma, pseudoaneurysm, radiation therapy, cerebral infarct

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INTRODUCTION

Head and neck tumor, especially nasopharyngeal carcinoma (NPC), is common in Southern East Asia including China, Hong Kong, and Taiwan [(1)]. The primary treatment of NPC is high-dose radiation with promising results [(2)]. However, there are many potential radiation complications such as temporal lobe necrosis, brainstem

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encephalopathy, myelopathy, cranial neuropathy, endocrine dysfunction, hearing loss, otitis, bone or eyeball damage, soft tissue necrosis/fibrosis/fistula, or severe epistaxis [3]. Beyond these, vascular complications due to radiation injury also remain challenge in treatment. Reported radiation-induced vascular lesions include obstructive arterial disease, either extracranial carotid or intracranial arteriopathy [4-6], and carotid rupture from pseudoaneurysm [3,7,9,10]. Carotid stenosis secondary to radiation injury to the carotid arteries is a well known long term complication of cervical radiotherapy [5]. Most of the previous studies aimed at the correlation between radiation-induced carotid stenosis and ischemic stroke. Pseudoaneurysm of ICA with carotid blowout syndrome is uncommon. It is even rare that simultaneous occurrence of both complications of carotid stenosis with ischemic stroke and carotid rupture from pseudoaneurysm. Here we present a patient who developed massive epistaxis due to rupture of radiation-related carotid pseudoaneurysm during acute ischemic stroke.

CASE REPORT

A 71-year-old man had NPC received radiation therapy (RT) 26 years ago. He had a habit of cigarette smoking for 30 years or more and alcohol drinking for about 10 years. He was noted to have hypertension 10 years ago but he did not regular take medication to control blood pressure. He suffered from an acute onset of slurred speech and unsteady gait with left neck pain. He did not pay attention to it. Tarry stool occurred three days later and he was sent to our emergency room.

On examination, his blood pressure was 176/91 mmHg. There was mild left facial palsy with dysarthria and dysphagia. The muscle power of his left limbs was grade 4/5 (Medical Research Council of Great Britain).

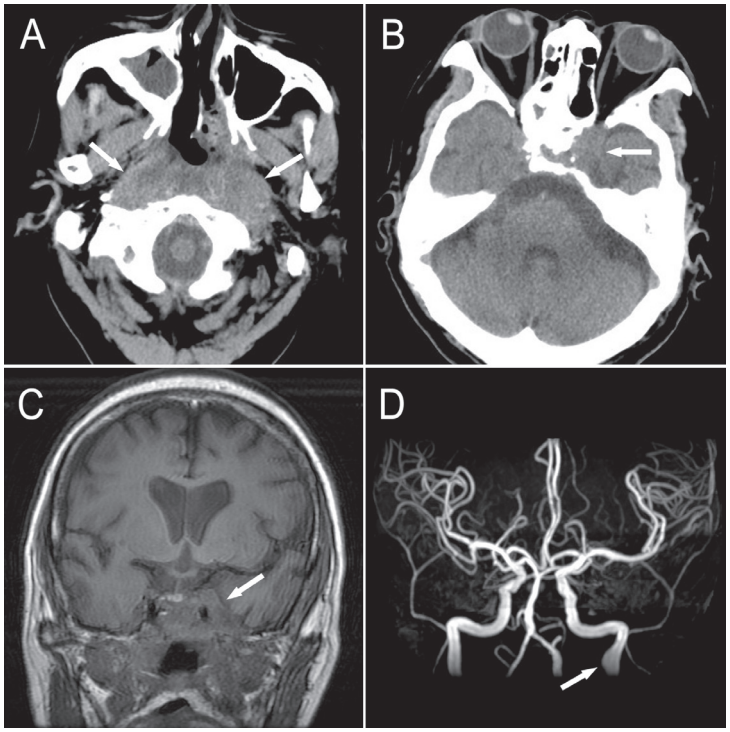

Figure 1. (A) Unenhanced CT images shows diffuse soft tissue thickening of nasopharynx (arrows). (B) Unenhanced CT images shows soft tissue lesion with left skull base and sphenoid bone destruction (arrow). (C) Unenhanced coronary T1-weighted brain MR image demonstrates soft tissue lesion with left skull base and sphenoid bone destruction, predominant on the left side (arrow). (D) Brain MR angiography discloses a saccular dilatation at the junction of lower part of petrous segment and distal cervical segment of the left internal carotid artery (arrow).

An emergent brain CT showed small old pontine infarct and nasopharyngeal soft tissue lesion with skull base and sphenoid bone destruction (Figs. 1A and 1B). He was admitted to neurological ward under the diagnosis of brainstem infarct and suspected upper gastrointestinal (UGI) bleeding. A brain MR angiography revealed old infarcts at cerebellum, pons and bilateral basal ganglia and diffuse soft tissue lesion in nasopharynx with predominance on the left side (Fig. 1C). There was no significant abnormality of visible cerebral arteries except for a suspected saccular dilatation at the high cervical left internal carotid artery (ICA) (Fig. 1D). The color-coded carotid duplex sonography found severe stenotic lesion in the right ICA bulb with elevated flow velocity (peak systolic velocity/end diastolic velocity = 192/73 cm/s). He received nasogastric tube insertion for intake owing to easy choking. We gave him oral clopidogrel and intravenous omeprazole for stroke and UGI bleeding treatment, respectively. Parenteral antibiotic was given for fever and aspiration pneumonia. An acute onset of disturbance of consciousness and right hemiplegia occurred with concomitant nasal bleeding and respiratory stridor on day three of admission. He received endotracheal intubation and was transferred to intensive care unit. An otolaryngeal consultation found septal erosion at posterior part of the nasopharynx with active oozing. Necrosis of both pharyngeal mucosa and underlying soft tissue was suspected. We discontinued clopidogrel. Bleeding was controlled by gauze packing of the nasopharynx and intravenous tranexamic acid. A panendoscopy disclosed much blood clots at his oropharynx and around the ehdotracheal tube. Only some small healing ulcers were found in the antrum. A follow-up brain CT the next day found a new infarct at the left frontal lobe.

Massive bleeding from nasal and oral cavity with hypovolemic shock occurred on day 10 of admission. An emergent digital subtraction angiography (DSA) under local anesthesia demonstrated more than 80% stenosis in the right proximal ICA, segmental dissection with moderate stenosis in the right ICA in the parapharyngeal space, and a large pseudoaneurysm about 25 mm in diameter located at the left high cervical ICA with moderate stenosis before the pseudoaneurysm (Fig. 2A). The left middle cerebral arterial territory was opacified from right vertebral injection and the left anterior cerebral artery was opacified from right ICA injection. The balloon occlusion test was not performed owing to relative adequate collateral circulation and emergent status with unstable vital sign. Therapeutic occlusion of the left ICA was achieved by 0.018 inch microcoils placement in the

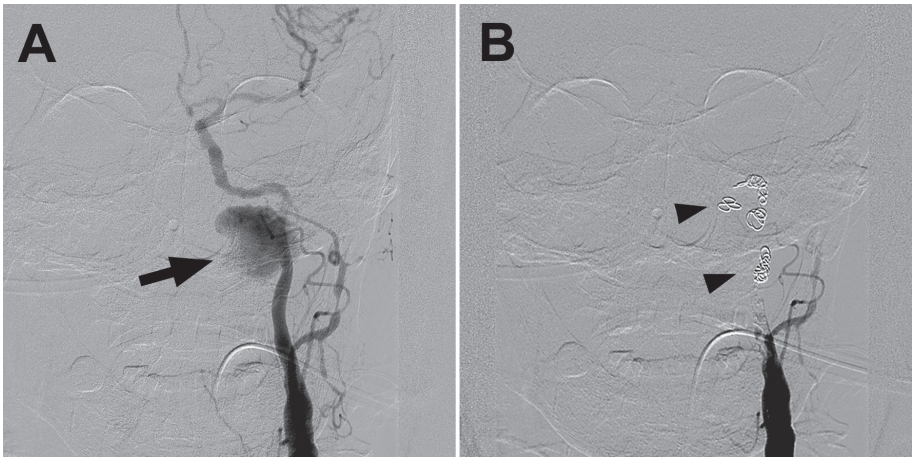

Figure 2. (A) Digital subtraction angiography displays a giant pseudoaneurysm at the lower part of petrous segment of the left internal carotid artery (arrow). (B) Carotid injection following microcoil embolization trapping internal carotid artery pseudoaneurysm. Note dense coil packing above (upper arrowhead) and below (lower arrowhead) pseudoaneurysm.

ICA distal to the pseudoaneurysm and both 0.018 and 0.035 inch microcoils with N-butyl-2-cyanoacrylate glue in the ICA proximal to the pseudoaneurysm (Fig. 2B). Post-embolization angiography revealed no more opacification of the pseudoaneurysm. The general condition of the patient stabilized after embolization. He gradual regained his consciousness yet remained right hemiplegia, and finally received tracheostomy owing to a ventilator-dependent respiration.

DISCUSSION

This patient presented as an ischemic stroke with severe stenotic lesion in the right ICA. Antiplatelet agent is the principle treatment in this situation. A recurrent stroke, which later is believed due to thrombotic emboli from ICA pseudoaneurysm, superimposed nasal bleeding caused a respiratory distress and required endotracheal intubation. However, epistaxis was initially thought from necrotic pharyngeal tissue and was primary controlled by local gauge packing. The saccular dilatation at junction of lower part of petrous segment and distal cervical segment of the left ICA on MR angiography was overlooked. The subsequent massive bleeding from nasal and oral cavity with hypovolemic shock aroused the attention of possible arterial bleeding. An emergent interventional angiography confirmed the speculation. The pseudoaneurysm was finally obliterated by therapeutic occlusion of ICA by microcoils placement with glue.

Diagnosis of ICA pseudoaneurysm is usually a challenge to physicians unless there appeared typical clinical presentations of aneurysmal rupture, for instance, repeated or intractable nasal, oral cavity bleeding, or even otorrhagia. Most of the previous studies focused on the radiation associated carotid stenosis and cerebral ischemic stroke. For a patient with NPC, recurrent tumor bleeding was supposed to be the first consideration for epistaxis. A recent angiographic case series study found that tumor bleeding was the most common cause for oronasal bleeding from patients with head and neck cancers following RT or chemotherapy [11]. Pseudoaneurysm bleeding comprises only 20% of 25 patients and simply one patient of NPC suffered active bleeding from ICA pseudoaneurysm. Lee AWM et al reviewed the incidence of late toxicities following radical RT in five series. There was only one out of 164 patients (0.6%) had carotid rupture in one series [3]. No carotid rupture was observed in other four series. Thus, when summarizing together with all five series, carotid rupture was such uncommon that occurred barely in one out all 7698 patients.

Carotid blowout syndrome (CBS) is one type of arterial injury that can occur following surgery, infection, RT, or chemotherapy [11]. The risk of CBS was reported to be higher by 7.6-fold in patients who received RT previously for head and neck cancers [12,13]. Described mechanisms about the formation of pseudoaneurysm include obliteration of the vasa vasorum, premature atherosclerosis, and weakening and necrosis of the arterial wall [14]. Experimental evidence suggests that ionizing radiation produces high concentrations of free radicals that damage all layers of the arterial wall [15]. The interval from previous radiation to CBS ranged from several months to 20 years or more. Most radiation-induced CBS in NPC occur in the petrous segment of the ICA [16]. For patients with excessive epistaxis from prior major head or facial trauma, it is not difficult to distinguish the traumatic ICA pseudoaneurysm, especially when Maurer's triad (history of prior facial trauma, delayed massive epistaxia, monocular visual disturbance) was present [17]. However, it is tough to make an accurate early diagnosis of radiation-related pseudoaneurysm for its infrequency and complexity.

A routine unenhanced brain CT or MRI study is not easy to identify the pseudoaneurysm since it can mimic an inflammatory or neoplastic mass. Saket el at reported the unenhanced CT and MRI findings of sphenoid sinus ICA pseudoaneurysm [17]. They concluded that a hyperdense, expansile, destructive, sphenoid mass on CT should arouse the suspicion of pseudoaneurysm. Cervical CT angiography or MR angiography provides better images to demonstrate the pseudoaneurysm. A recent study addresses that CT angiography demonstrates imaging findings not available with DSA and provides potentially useful prognostic information for patients with impending CBS [18]. Conventional angiography or DSA is the gold standard for conformational diagnosis as well as further endovascular treatment of pseudoaneurysm. Nevertheless, angiographic study is usually not an initial screening study for patients with epistaxis and might be preserved until succeeding massive bleeding.

Endovascular treatment of pseudoaneurym has becoming a better alternative method from traditional surgical ligation of ICA for CBS. Endovascular trapping of the ICA and pseudoaneurysm using coils, glue, or detachable balloons have been reported with high success rate for hemostasis [19,20,21]. To improve the procedure-related cerebral ischemic risk, reconstruction of the ICA with stents furnishes lower risk and recurrence rate [10]. The clinical severity of CBS was classified into three groups: acute, impending, and threatened [21]. Chang et al found that clinical severity is the significant factor affecting the hemostatic outcome of endovascular management and advised making this clinical classification as a guide for management of CBS [21]. The outcomes of most NPC patients with acute CBS were not satisfied. In this patient, endovascular treatment achieved immediate oblileration of ICA pseudoaneurysm. There was good collateral circulation to the left anterior and left middle cerebral arteries. Although no procedure-related complication occurred, this patient remained ventilator dependent from recurrent stroke and pneumonia. In Mak's series, 10 out of 15 patients who received successful endovascular treatment of ruptured ICA pseudoaneurysm after irradiation for NPC died within 11months [10]. Causes of death include sepsis, pneumonia, meningitis, myocardial infarction, and tumor progression.

In conclusion, simultaneous occurrence of ischemic stroke with carotid stenosis and CBS due to rupture of ICA pseudoaneurysm in NPC patients with prior neck radiation is a rare but life-threatening complication. Concomitant epistaxis and tarry stool in NPC patients during acute ischemic stroke warrants further cervical angiographic study to prevent subsequent catastrophic bleeding. Endovascular treatment provides immediate hemostasis and obliteration of ICA pseudoaneurysm.

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