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# Non-Tuberculous Mycobacterium Induced Pseudoaneurysm of the Common Carotid Artery

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An 81-year-old male patient presented with complaint of a pulsating neck mass. The patient had a previous history of cervical lymphadenopathy by non-tuberculous mycobacterium infection. Rapid growth of the mass on admission and contrast enhanced computed tomography of the neck resulted in a diagnosis of non-tuberculous mycobacterium induced pseudoaneurysm. The patient underwent emergency open repair of the pseudoaneurysm. Pseudoaneurysm of the common carotid artery is regularly reported, but here we report a rare case of non-tuberculous mycobacterium induced pseudoaneurysm of the common carotid artery.

Key words: 1. Carotid arteries

- 2. Infection
  - 3. Nontuberculous mycobacterium
  - 4. Aneurysm, false
  - 5. Vascular disease

# **Case Report**

An 81-year-old male patient presented with complaint of a pulsating mass on the left side of the neck, which enlarged rapidly for three admission days. An infected pseudoaneurysm was diagnosed by neck computed tomography (CT) scan, and the patient underwent emergency operation against impending rupture.

Three months prior to admission, the patient was prescribed anti-tuberculosis medications (isoniazid, ethambutol, rifampicin, pyrazinamide [HERZ]), initially for cervical lymphadenopathy by tuberculosis infection and followed serially in the ENT (ear, nose, and throat) department and pulmonary internal medicine (PI) outpatient department (OPD). Two months prior to admission, cultures from sampling of the neck lymph node revealed the presence of *Mycobacterium avium* and diagnosis was confirmed as nontuberculous mycobacterial (NTM) lymphadenitis. HERZ medication was continued, and erythromycin and macrolide series antibiotics were added to the treatment regimen.

An ultrasound-guided fine needle aspiration of the neck mass was planned (OPD). Two days prior to admission for the aspiration procedure, hoarseness and neck mass enlargement developed and the patient was then admitted via PI OPD.

Two days after admission to the PI OPD, neck CT scan showed huge pseudoaneurysm with concealed rupture of left carotid artery surrounded by cervical lymph node with inflammation. Emergency open repair was necessary due to the rapid growth of the mass observed over 3 days, and was accomplished by the thoracic and cardiovascular surgery depart-

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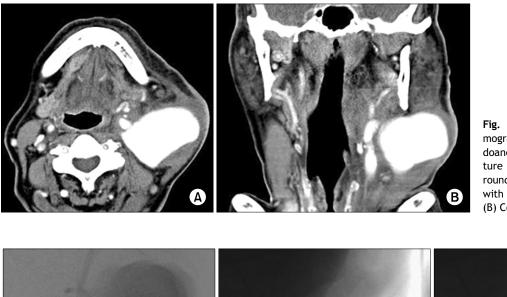


Fig. 1. Preoperative computed tomography scan showed a huge pseudoaneurysm with concealed rupture of the left carotid artery surrounded by cervical lymph node with inflammation. (A) Axial view. (B) Coronal view.



**Fig. 2.** Intraoperative angiography. (A) Pre-ballooning digital subtraction angiography image of contrast enhanced huge mass originating from the CCA. (B) Fluoroscopic image of inflated intraluminal balloon catheter without contrast injection via CCA. (C) Post-ballooning fluoroscopic image showed successful intra-arterial occlusion of breach at vessel wall of CCA without contrast leakage into pseudoaneurysm. CCA, common carotid artery.

ment (Fig. 1).

The pseudoaneurysm was friable and the preoperative CT scan showed indefinite enhancement of the distal runoff of the left internal carotid artery (ICA). Therefore, a hybrid operation was planned using intra-arterial occlusion using a balloon catheter under C-arm guidance to prevent intraoperative disastrous bleeding following rupture.

Under general endotracheal anesthesia, we performed a cutdown of the left common femoral artery. We checked for intra-arterial occlusion of the pseudoaneurysm orifice using a 2.5 mm ballooning catheter via 7Fr Flexor Shuttle guiding sheath (Cook Medical, Bloomington, IN, USA) on a guidewire rail (0.3556 mm) after left common carotid artery (CCA) selection using 5Fr Headhunter catheter (Terumo Co., Tokyo, Japan).

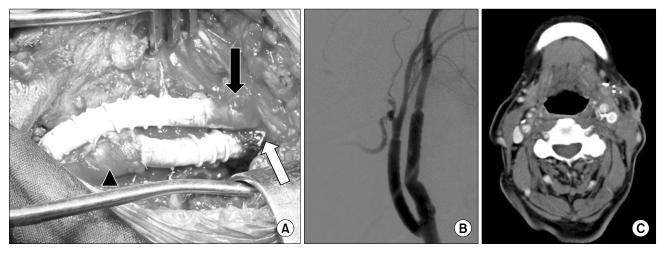
We detected remarkable changes of the contrast enhancement of pseudoaneurysm between pre- and

post-ballooning of CCA (Fig. 2), and then emptied the balloon.

Skin incision was made by straightened s-shape from the proximal insertion portion of the sternocleidomastoid muscle at the jugular notch, and was extended to just before the mandibular angle through the lower border of the left mandible above and anterior to the pulsating mass. The CCA and external carotid artery (ECA) were encircled for clamping. Intra-arterial occlusion of the distal ICA to the mass using the balloon catheter was prepared because control of the distal portion of the ICA by pseudoaneurysm was not possible. After clamping on ECA and CCA with intra-arterial occlusion, we incised the aneurysm directly.

The preoperative CT scan confirmed an intact circle of Willis. We decompressed the pseudoaneurysm by direct incision, and after withdrawal of the balloon catheter, found and clamped on the distal ICA.

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**Fig. 3.** (A) Postoperative field view of bypass grafting using ringed Gore-Tex 6 mm (WL Gore and Associates Inc., Flagstaff, AZ, USA) among distal external carotid artery (black arrow), distal internal carotid artery (white arrow), and common carotid artery (black arrow head). (B) Intraoperative final digital subtraction angiography showed patency of bypass graft without leakage. (C) Postoperative computed tomography scans at 3 month follow-up (postoperative) showed decreased but remnant lymphadenitis around artificial graft.

The location of defect was found just proximal to bifurcation of CCA. We made a confirmed diagnosis as CCA pseudoaneurysm of the left side.

The pseudoaneurysmal wall was covered with fresh blood clots, but without abscess or necrotic materials induced by active inflammation. The vascular wall and intrasaccular thrombus were sampled intraoperatively for pathology, and the postoperative report found no microorganism growth (e.g., NTM), relatively healthy ECA, stenotic change of ICA with chronic inflammation, and gross atherosclerosis.

Resection of the pseudoaneurysmal sac and trimming of the ends of CCA, ICA, and ECA were followed by plentiful saline irrigation. Bypass grafting anatomically was accomplished by interposition with ringed Gore-Tex 6 mm (WL Gore and Associates Inc., Flagstaff, AZ, USA) between ICA and CCA in an end-to-end fashion and between ECA and CCA in an end-to-end fashion (Fig. 3A).

Intraoperative final sheath angiography showed patency of bypass graft without leakage (Fig. 3B). Postoperative CT scans showed remnant lymphadenitis around the artificial graft (Fig. 3C). We prescribed continuous intravenous antibiotic and oral anti-tuberculous medications. The patient was discharged at postoperative day 21 (3 weeks) without any complication. Scheduled follow-up is ongoing at PI and cardiovascular surgery OPDs.

### Discussion

Common carotid artery pseudoaneurysm is derived from oropharyngeal or respiratory tract infection extending to deep neck infection. Examples of clinical manifestations of this rare disease are pulsating neck mass, Horner syndrome, and lower cranial neuropathy. Requirement of simultaneous vessel ligature should be guaranteed for prevention of lethal complications such as massive hemorrhage following rupture [1].

Liston reported this disease entity with high mortality for the first time in 1843. Since then, and the incidence rate declined remarkably due to introduction of empirical antibiotic therapy [2]. Incidence rate is 62%, 25%, and 13% in ICA, ECA and CCA, respectively. Open surgical ligation is recommended for the majority of cases, with surgical treatment with a mortality of 35% compared with mortality of 77% of cases with non-surgical treatment [3].

Endovascular arterial repair by exclusion using stent graft in the infected pseudoaneurysm has been reported [3]. Carotid pseudoaneurysm must be diagnosed early and undergo surgical treatment immediately.

Rare causes are bacterial infective endocarditis, penetrating trauma, postoperative infection (e.g., carotid endarterectomy), iatrogenic arterial or venous puncture, and pharyngeal foreign body [3]. In several reported cases of mycotic aneurysm, tuberculosis induced pseudoaneurysm was due to intravesical bacillus Calmette–Guérin instillation as immunotherapy for bladder cancer [4].

We report this rare case now, as NTM induced pseudoaneurysm of carotid artery maybe the first case report in the literature.

Previous reports documented that pseudoaneurysm develops after several days to 2 months of infection (14 days is the average). Pathogen identification is important for appropriate, pathogen-specific antibiotic therapy for pharmacological treatment. Ligation of the affected artery is recommended simultaneously with diagnosis for surgical treatment. Most cases involve the ICA, so ligation of ICA is generally the choice for surgical treatment. However, there are different views of ligation of ICA and CCA [5]. Due to the collaterals between CCA and ICA, it is inappropriate to ligate only the CCA (like ligation of only the ICA). When the aneurysm is on the external branch of carotid artery, ECA ligation is sufficient. Unless bleeding is controlled by proximal ligation, distal ligation should be required [6].

In addition to surgical treatment, intervention has been reported to control bleeding and prevent or decline the rate of embolization or mass effect. Successive antibiotic therapy is recommended for 4 to 6 weeks for treatment of remnant infection sources of the lesion [7].

When there is possibility of superimposed infection on artificial grafts, addition of rifampicin to the basic antibiotic therapy is more effective in some reports. Continuous follow-up is necessary in combination with application of appropriate antibiotic therapy [8]. Multi-drug resistant bacterial infection or septic condition might result in persistent and multiple abscess formation [3]. In this case, we are planning to apply anti-tuberculosis medication with rifampicin for an additional 1 year.

Additionally, we would like to suggest two important implications of our case. The first is the awareness of possible disastrous complications of routine percutaneous aspiration biopsy and drainage in cervical lymphadenopathy. The second is that, during the operation, intraluminal balloon occlusion guided by intraoperative DSA might be another option for prevention of massive bleeding.

In conclusion, NTM lymphadenopathy induced pseudoaneurysm is a very rare disease but can be easily differentiated from the typical clinical manifestations of the common mycotic disease. We suggest that CT angiography must be applied for early diagnosis of extent, characteristics, therapeutic plan, and prevention of lethal complications.

# **Conflict of interest**

No potential conflicts of interest relevant to this article are reported.

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