

비정형 결핵성 림프절염의 악화로 오인된 경동맥 가성동맥류 1예

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A Case of Carotid Artery Pseudoaneurysm Misdiagnosed as Aggravation of Non-tuberculous Mycobacterial Lymphadenitis

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= Abstract =

경동맥에서 발생하는 가성동맥류는 비교적 드문 질환으로 주로 외상, 감염, 수술의 합병증 또는 방사선 치료 후에 합병증으로 발생할 수 있다. 비정형 결핵성 림프절염에 의한 경동맥 가성동맥류는 거의 보고된 바 없으며 따라서 비정형 결핵성 림프절염이 동반된 환자에서는 급성악화에 의한 농양형성으로 오인될 수 있다. 비정형 결핵성 림프절염으로 치료중인 81세 남자 환자에서 이러한 경험을 하였기에 문헌고찰과 함께 보고하고자 한다.

An 81 year-old male patient presented with rapid enlargement of left lateral neck mass, diagnosed two months earlier as cervical nontuberculous mycobacterial lymphadenitis. Abscess formation and impending rupture related to aggravation of nontuberculous mycobacterial lymphadenitis was highly suspected. Unexpectedly, blood flow was detected by Doppler ultrasonography, which indicated possible vascular mass. Computed tomography demonstrated a 6.0 cm x 4.0 cm sized enhancing mass consistent with pseudoaneurysm of internal carotid artery. The patient underwent pseudoaneurysmectomy. Surgical drainage without adequate evaluation might have led to potentially life-threatening condition. We describe this rare case with importance of imaging screening in a neck mass.

Key Words : Pseudoaneurysm; Carotid artery; Non-tuberculous mycobacterial lymphadenitis

Introduction

Carotid artery pseudoaneurysm is an uncommon condition that can occur secondary to trauma, infection, surgery, or radiotherapy, along with other causes.^{1,2)} Its incidence due to non-tuberculous Mycobacterial (NTM) lymphadenitis is extremely rare. Clinically, pseudoaneurysm

and NTM lymphadenitis-associated abscess present with similar symptoms with the exception of the pulsatile mass. This can lead to confusion between pseudoaneurysm and aggravation of NTM lymphadenitis. Mistaking a pseudoaneurysm for a cervical abscess can result in fatal outcomes to patient. Therefore, when a pseudoaneurysm is suspected, imaging is necessary prior to any surgical intervention.

This report highlights a case of carotid artery pseudoaneurysm that was referred for surgical drainage because it was mistaken for an aggravation of NTM lymphadenitis.

Case report

An 81-year-old male patient with NTM lymphadenitis

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Fig. 1. CT scan showing a rim-enhancing mass with central necrosis in the area of the carotid bifurcation. The internal carotid artery was partially surrounded by the mass (arrow).



Fig. 2. Left neck of the patient was swollen with reddish color change of the skin and central scar formation.

was referred to our ENT clinic for abscess drainage of a left lateral neck mass. The patient, with a previous history of bladder cancer, had presented two months prior to this visit with a left lateral neck mass for differential diagnosis of tumor recurrence. At that time, the patient had been evaluated with Computed-tomography (CT) scan, Positron emission tomography-Computed tomography (PET-CT), and Fine needle aspiration (FNA). The CT scan showed a well-defined rim-enhancing mass with central necrosis near the carotid bifurcation area of the left neck, suggesting necrotizing lymphadenitis (Fig. 1). Cytology using FNA revealed necrotizing granulomatous lymphadenitis with positive PCR results for tuberculosis (TB) and interferon (INF)-gamma. Tuberculous lymphadenitis was suspected; however, the patient had no pulmonary complaints and no irregularities on chest radiograph.

The patient was referred to the Pulmonary Department and started on antituberculous therapy (Isoniazid, Rifampicin, Ethambutol and Pyrazinamide). However, the patient was later confirmed to have NTM infection finally based on the results of bacterial culture; Therefore, the anti-tuberculous medication was changed to Rifampicin, Ethambutol, and Clarithromycin.

The neck mass had worsened two days prior to this visit and was suspected to be aggravation of NTM lymphadenitis. Therefore, incision and drainage of abscess were recommended by the Pulmonary Department. On physical exam, the left neck of the patient was swollen with overlying reddish skin discoloration and central scar formation (Fig. 2).

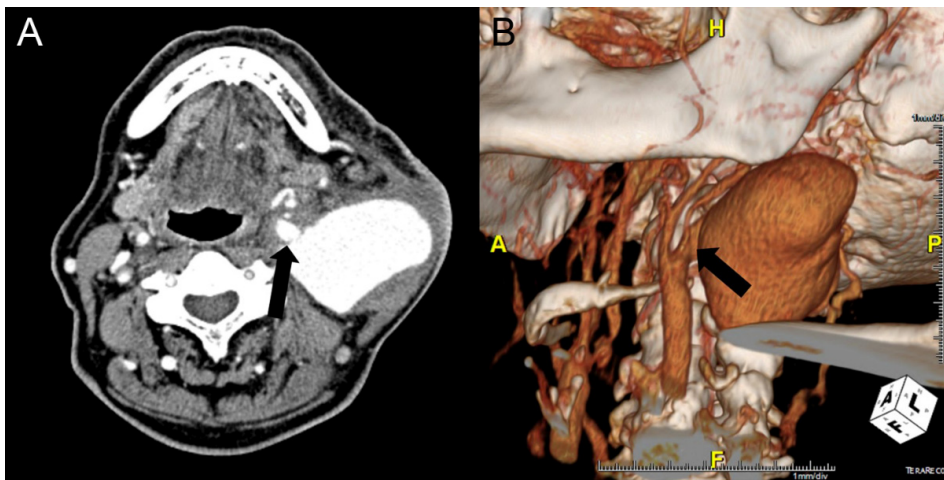


Fig. 3. (A) Computed tomography scan of the neck showed a 6 cm x 4 cm sized pseudoaneurysm in the left internal carotid artery. (B) Three-dimensional reconstruction of the CT scan demonstrated leakage of contrast media from the internal carotid artery (arrow). On both images, external and internal carotid arteries were located in front of a fistula tract (arrow)

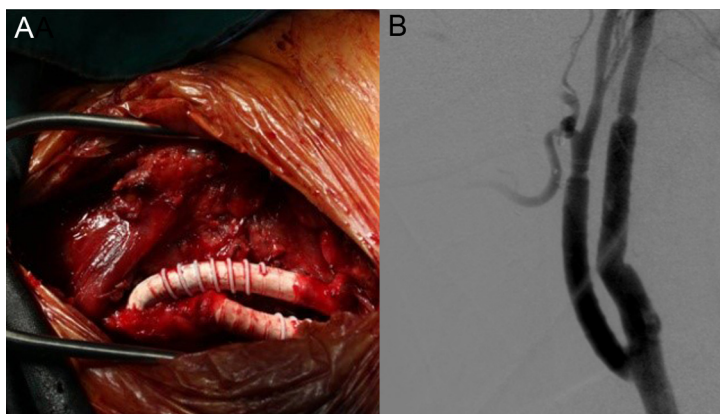


Fig. 4. (A) Pseudoaneurysm was resected, and interposition grafting was performed in both the external and internal carotid arteries due to pseudoaneurysmal fistula close to the carotid bifurcation. (B) Postoperative angiogram shows no leakage of contrast dye.

Abscess formation and impending rupture related to aggravation of NTM lymphadenitis was highly suspected. However, the swollen mass was pulsatile so surgical drainage was postponed, and Doppler ultrasonography was conducted in the out-patient clinic. The Doppler ultrasonography showed blood flow in the mass, which was therefore suspected to be of vascular origin. A CT scan with angiography was performed to better evaluate the mass. Neck CT scan showed a 6.0 cm X 4.0 cm pseudoaneurysm presumed to be caused by NTM lymphadenitis (Fig. 3-1). The internal carotid artery just above the bifurcation was found to be the source of the pseudoaneurysm using three-dimensional reconstruction of the CT scan (Fig. 3-2). Laboratory results were notable for a white blood cell count of 18,060/mm³, an erythrocyte sedimentation rate of 39 mm/hr, and a C-reactive protein concentration of 20 mg/dL.

The patient was referred to the Cardiovascular Surgery Department and underwent emergency pseudoaneurysmectomy with graft interposition (Fig. 4-1).

The pseudoaneurysm was confirmed intraoperatively as originating from the ICA 5mm above the bifurcation. Postoperative angiography showed restoration of vascular continuity with normal flow of the carotid artery (Fig. 4-2). After the surgery, the patient was treated with anti-mycobacterial therapy for six months and was free of disease.

Discussion

Pseudoaneurysm is defined by loss of integrity of the three layers of the arterial wall, resulting in a communication with the lumen of a ruptured blood vessel. This commu-

nication forms a hematoma that appears similar to a cystic mass. Pseudoaneurysm can arise from any artery in the body. In the neck, the carotid artery is most commonly involved.³⁾ Carotid artery pseudoaneurysm is usually related to trauma, surgery, vasculitis, infection, tumor, radiotherapy, or arteriosclerosis.^{1,2)} This report highlights a carotid artery pseudoaneurysm related to NTM lymphadenitis.

Cervical lymphadenitis caused by NTM infection is a rare disease and most commonly occurs in children.⁴⁾ However, many reports have noted that infection with NTM is increasing with the increase in the number of immunocompromised adult patients.^{5,6)} Carotid artery pseudoaneurysm from NTM lymphadenitis is important because it is a potentially lethal condition that can occur in any NTM lymphadenitis patient, although it is a rare complication.

The natural history of NTM lymphadenitis is not well documented. It is known that the infected lymph nodes form abscesses and increase in size. In time, they become adherent to the skin and result in fistula formation with subsequent drainage.^{7,8)} However, in this case the proximity of the infected lymph node to the carotid artery resulted in destruction of the arterial wall and fistula formation between the carotid artery and the infected tissue.

To our knowledge, this is the first case report to describe pseudoaneurysm formation related to cervical NTM lymphadenitis. The most common site of infectious pseudoaneurysm is the thoracic aorta and the pseudoaneurysms are predominately caused by tuberculosis due to the anatomical proximity of the lung.⁹⁾ Therefore, most previous cases reported pseudoaneurysm of the thoracic aorta occurring in patients with pulmonary tuberculosis. The importance of our

rare case is the report of a pseudoaneurysm of the carotid artery in a patient with NTM lymphadenitis during anti-mycobacterial therapy.

The most common presenting symptom of pseudoaneurysm is a pulsatile neck mass with swelling.¹⁰⁾ Other symptoms can include pain, fever, shock, embolism, and neurologic changes related to the mass.¹¹⁾ Therefore, a pseudoaneurysm with deep neck space infection would not be easily differentiated if a pulsatile mass was not palpated. Many imaging modalities are available to differentiate pseudoaneurysm, including Doppler ultrasonography, CT scan, MRA, and angiography. Doppler ultrasonography is non-invasive and cost-effective. It shows the presence of blood flow within the pseudoaneurysm with a communicating channel to the artery. CT angiography shows an enhancing mass projecting outside of the arterial wall. MRA is more sensitive than contrast angiography and avoids angiographic complications related to contrast.¹²⁾ Among the various diagnostic modalities, Doppler ultrasonography has many advantages as an initial diagnostic test. It is simple, portable, repeatable and non-invasive. Since pseudoaneurysm and abscess from NTM infection can be difficult to differentiate clinically, misdiagnosis of carotid artery pseudoaneurysm can lead to potentially life-threatening conditions if surgical drainage is performed without adequate evaluation. Doppler ultrasonography is very helpful screening tool, although most pseudoaneurysms require angiography for definitive diagnosis. It can be conducted easily in the outpatient clinic if a mass is vascular in origin. It is important that simple and easily performed imaging is obtained before treatment is initiated.

If diagnosis is confirmed with imaging, carotid artery pseudoaneurysm should be treated immediately. Without prompt treatment, major hemorrhage and hemodynamic shock can develop due to rupture or perforation of the pseudoaneurysm.¹³⁾

This report describes a patient with cervical NTM lymphadenitis and subsequent carotid artery pseudoaneurysm. Since NTM lymphadenitis is a rare condition in adults,¹³⁾ this presentation is extremely rare. The initial misdiagnosis of pseudoaneurysm is extremely concerning. Therefore, we strongly recommend Doppler ultrasonography for the initial evaluation of suspected pseudoaneurysm caused by NTM lymphadenitis. Ultrasonography can allow for prompt bedside or outpatient examination to identify abnormal vascular

structures such as pseudoaneurysms. If patients with NTM lymphadenitis are experiencing rapid neck mass enlargement, carotid artery pseudoaneurysm should be considered, and testing should be performed immediately due to the high mortality rate of this event.

Conclusion

Imaging screening of neck mass is very important. Surgical drainage without adequate evaluation might have led to potentially life-threatening condition. Also, Carotid artery pseudoaneurysm is rare but should be considered in NTM lymphadenitis patients showing rapid enlargement of a neck mass.

중심 단어 : 경동맥, 가성동맥류, 비정형 결핵성 림프절염

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