

Progressive cervical tumour in an HIV-patient: giant pseudoaneurysm of the carotid artery: a case report

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Background

Aneurysms of the extracranial carotid artery are a rare entity and correspond to <1% of all arterial aneurysms.

Case summary

A 58-year-old male with known human immunodeficiency virus (HIV) infection presented in the emergency department with a massive cervical tumour on the right side of the neck and a severe occipital pain. Contrast computed tomography demonstrated a pseudoaneurysm of the proximal right internal carotid artery (ICA). Open surgery of the ICA was performed with reconstruction of the posterior vessel wall, embolectomy of the ICA, and anterior pericardial patch reconstruction. After an uneventful postoperative course, the patient was readmitted 4 weeks later with a right retrobulbous haematoma caused by a carotid cavernous fistula. Therefore a coil-embolization of the fistula and finally of the distal right carotid artery was performed.

Discussion

In patients with a proven HIV infection, the occurrence of a vasculopathy in the extra- or intracranial carotid artery is significantly increased and the second most common site after the lower extremities. In patients with progredient neck swelling it should be considered in the differential. Surgical therapy is the preferred treatment strategy in the extracranial aneurysm type, especially in this patient collective.

Keywords

Human immunodeficiency virus • Pseudoaneurysm • Aneurysm • Carotid artery • Cardiovascular surgery • Case report

ESC Curriculum

9.1 Aortic disease • 9.3 Peripheral artery disease

Learning points

- In patients with a proven HIV infection, the incidence of HIV-related vasculopathy is increased, with carotid artery aneurysms or pseudoaneurysms making up to 25% of all HIV aneurysms.
- Therefore HIV infection must be excluded in younger patients with the occurrence of this entity.
- Diagnosis can be confirmed by computed tomography angiogram.
- In HIV-pseudoaneurysms, the surgical therapy should be preferred, due to increased stent-related complications in interventional therapy in this patient group.

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Introduction

Aneurysms of the extracranial internal carotid artery (ICA) are a rare entity, contributing to only 1.25% of all arterial aneurysms.¹ Before the introduction of antibiotics, infections like syphilis, tuberculosis, and local infections were the main underlying cause. Nowadays, the aetiology has shifted to atherosclerosis (80%), fibromuscular dysplasia, trauma, and dissection.²

Nevertheless, carotid artery aneurysms making up to 25% of all HIV aneurysms.³ In Germany the number of HIV infections has increased until 12/19 to 90.70 cases. Accordingly, nearly 11,000 are potentially undiagnosed and 37.9 million people were living with HIV worldwide.⁴

We present the case of a 58-year-old male with a proven human immunodeficiency virus (HIV) infection presenting at the emergency department with a painful right sided cervical tumour.

Timeline

Day	Event
0	A 58-year-old male with HIV infection presented with an increasing swelling of the right neck combined with occipital pain CT-scan showed a carotid pseudoaneurysm
3	Operative reconstruction with exclusion of the 'blow out' site, embolectomy of the internal carotid artery and patch plasty was performed
7	The patient was discharged after uneventful p.o. course on warfarin
23	Postoperative elective CT-scan: sufficient reconstruction of the carotid bulb with pseudo-occlusion of the internal carotid artery in the carotid siphon
35	Spontaneous right sided retrobulbous haematoma
42	Invasive angiography: carotid cavernous fistula, chronic occlusion of the right internal carotid artery before the ophthalmic artery
46	Interventional coiling of the carotid cavernous fistula
47	Discharge without neurological impairment

Case presentation

We report the case of a 58-year-old male who presented at the emergency department with an increasing right sided cervical tumour and occipital pain (Figure 1).

The HIV-1-infection was diagnosed in 1999, resulting in an anti-retroviral therapy from 1999 to 2005 and resumption of this therapy again from 12/2009 combining Emtricitabin and Dolutegravir. In 2010, an atypical mycobacteria-related infection was diagnosed which presented as ulcerating pangastritis (specific medication was given).

His past medical history includes coronary artery disease, myocardial infarction, and the history of percutaneous coronary intervention.

Further, the patient had undergone multiple procedures for peripheral arterial disease. After repeated percutaneous transluminal angioplasties for the left superficial femoral in 2015 a femoral endarterectomy and femoro-crural bypass was performed. Because of bypass-occlusion in 2019, an interventional revision without success was followed by a redo bypass, which was again occluded in October 2020 without any further options for revascularization. The patient had a history of heavy smoking for decades (30 py).

Before admission the patient had noticed an increasing right sided cervical tumour without any neurological impairment. No specific abnormalities of the cardiovascular system were found with normal cardiac biomarkers. Contrast computed tomography (CT) demonstrated a normal supraaortal vascular anatomy with a large pseudoaneurysm (38 × 31 × 35 mm) originating from the right proximal ICA (Figure 2), distal of the aneurysm the ICA was thrombotic occluded.

On an interdisciplinary vascular board, the decision for surgical reconstruction was made. After exposing the ICA and the carotid bifurcation the pseudoaneurysm was incised, the destructed posterior vessel wall (Figure 3A) was excluded and a recanalization of the thrombosed ICA was performed via embolectomy. Finally the anterior wall was reconstructed using a bovine pericardial patch (Figure 3B).

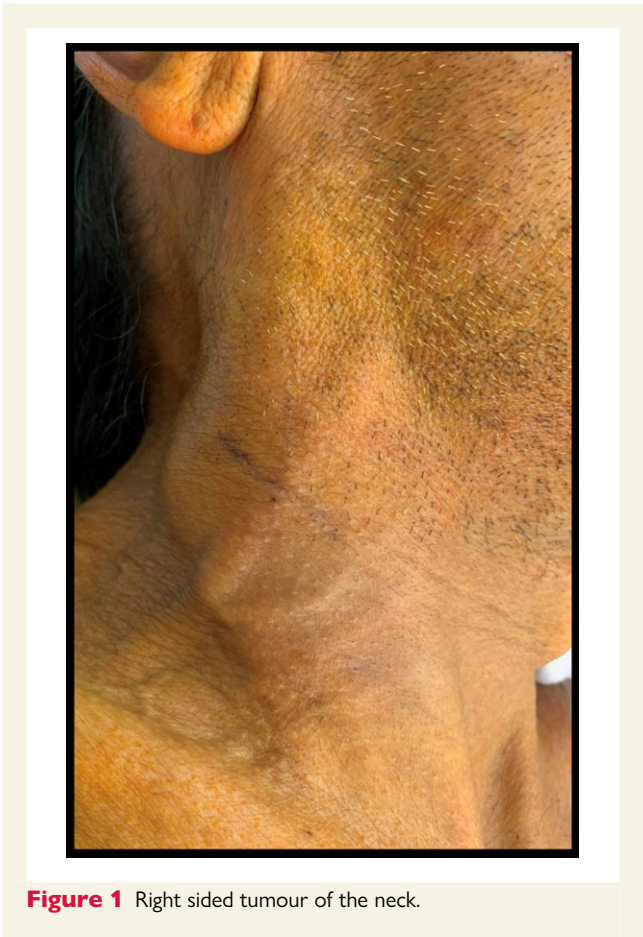


Figure 1 Right sided tumour of the neck.

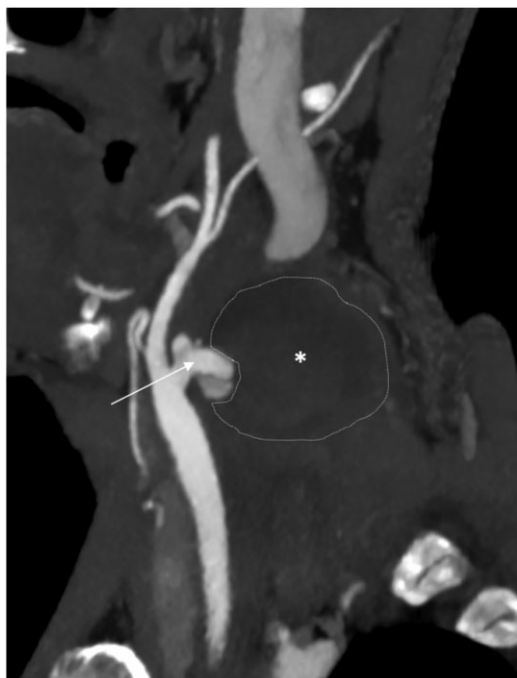


Figure 2 Preoperative contrast computed tomography: pseudoaneurysm \times (38 \times 31 mm) with origin of the right internal proximal artery with 6 mm neck (arrow). Internal carotid artery occluded.

The postoperative course was uneventful. He was discharged on the 4th postoperative day with a recommendation for warfarin for the next 3 months.

Microbiological examination of the intraoperative specimen was negative, as well as the blood cultures.

A CT-scan 14 days later showed an effective exclusion of the pseudoaneurysm (Figure 4) and a partly recanalized ICA with a small indistinguishable carotid siphon (differential diagnosis pseudo-occlusion). Four weeks later, the patient was readmitted with a retrobulbar haemorrhage. After urgent canthotomy an invasive neuroradiology imaging revealed a carotid cavernous fistula (CCF) (right carotid internal artery—cavernous drain per ophthalmic vein). The right distal ICA was re-occluded. Therefore, coil-embolization of the fistula (eight coils) and final occlusion of the distal right ICA with an Amplatzer device (8 mm, Abbott USA) were performed. The next day the patient was discharged without any neurological impairment.

The patient was last seen at HIV outpatient department 22 months after the first operation without any procedure-related neurological abnormalities, but in overall significantly reduced general and nutritional condition.

Discussion

Aneurysms of the extracranial carotid artery are a rare entity and correspond to <1% of all arterial aneurysms.¹ Infectious diseases like syphilis and tuberculosis were the main causes in earlier decades. Nowadays the two most predominate groups are atherosclerotic,

followed by pseudoaneurysms due to trauma or previous operations.² The mean age ranges around 60 years.^{5,6}

Arteriopathy associated with HIV was first described in children in 1987⁷; it includes vasculitis and perivasculitis,⁸ fibroproliferative occlusive disease,⁹ and aneurysm formation.¹⁰ The exact mechanism of aneurysm formation is unknown. Arterial wall weakening may result from the direct action of the virus or via immune complex mechanisms.⁸ Previous chronic infections are discussed as a cause of an inflammatory immune response. With 20% mycobacteria tuberculosis and second herpes viruses (13%) are the most commonly described infective agents. Also a vasculitis of the vasa vasorum resulting in transmural necrosis apparently plays a key role in the development of extracranial aneurysms of the carotid artery.^{10–12}

In the here presented case, the patients' medical history showed a severe atherosclerotic burden. A multifactorial aetiology might have led to the formation of carotid pseudoaneurysm. This is supported by the fact that the majority of HIV-associated carotid aneurysms were in young men, with a mean age of 30 years.¹⁰

The extracranial type of carotid aneurysms in HIV-patients tends to be saccular with development of a pseudoaneurysm.¹³ Whereas in noninfected individuals pseudoaneurysms only contribute to 10% of the extracranial carotid artery aneurysms,⁵ in HIV-patients this type sums up to 44%⁹ or even 85%.¹⁰

Diagnosis is usually performed by computed or magnetic resonance-tomography. An overall neurologic assessment is mandatory. Nerve compression and palsies. Dysphonia and dysphagia are common symptoms.

There have been numerous reports in the literature evaluating associated clinical and laboratory factors and outcomes. A low CD4 count (<200 cells/ μ L) combined with a low albumin count (<30 g/L) may predict a poor outcome.^{10,14} Serology for Syphilis, herpes, and tuberculosis might be helpful to allow goal-directed therapy.

Since the first description of proximal artery ligation in 1968 to treat a carotid aneurysm, the management and outcomes have changed substantially.

Due to localization, the configuration and aetiology of the aneurysms, multiple therapeutic interventional, or operative options are available. Surgical or endovascular repair is recommended due to inherent risk of neurological symptoms and/or significant risk of rupture.

In HIV-patients, surgical approach was most frequently reported for the management of extracranial carotid artery aneurysms.^{10,13} Surgical treatment is primarily provided with different vascular reconstruction techniques ranging from aneurysmectomy with end-to-end-anastomosis, patch plasty up to interponate, or reinsertion of the ICA into the common carotid artery to simple vessel ligation.

Also, endovascular techniques, e.g. implantation of covered stent-grafts and coiling procedures were described. In the study review of Silvestri dealing with carotid artery aneurysms in HIV the overall morbidity and mortality were both high with endovascular treatment carried the highest rates with 57% morbidity and 14.7% mortality vs. 10% and 5% for open surgery.¹⁰ These findings were supported by Padayachy and Robbs¹³ showing that the intervention group had worst outcome with 80% of the patients developing complications, e.g. endoleaks and thrombosed stent-grafts. The reasons for poor outcome after intervention might be due to clot embolization or

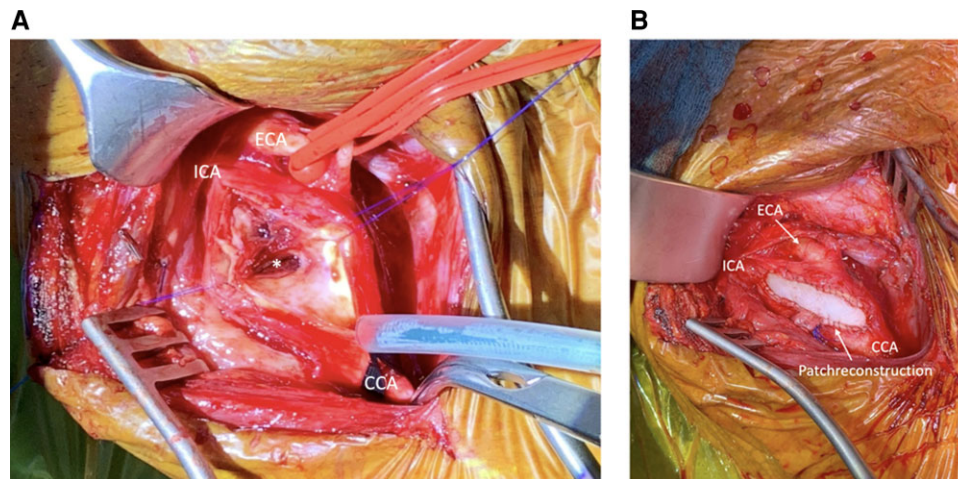


Figure 3 (A) Intraoperative situs: view from the right internal carotid artery into the false aneurysm *. (B) Final patch plasty after successful reconstruction.



Figure 4 Postoperative contrast computed tomography after 18 days showing sufficient vessel reconstruction (arrow) with pseudo-occlusion of the right internal carotid artery, strong opthalmic right artery.

the presence of vasculitis challenging the sealing in this area and increasing the likelihood of the development of endoleaks.¹⁰

In the here described case various therapeutic options were interdisciplinary discussed with decision for open surgical repair due to the aneurysm formation. Ligation of the ICA was also discussed. During surgery, the ICA revealed a thrombotic occlusion, followed by embolectomy.

This may have led to the CCF. Carotid cavernous fistula are commonly classified based on haemodynamic and aetiology, which is commonly secondary to trauma or can occur spontaneously in the setting of aneurysms or medical conditions predisposing to arterial wall defects.¹⁵ High-flow CCF may present with chemosis, proptosis, increased intraocular pressure, diplopia, and decreased vision. Cerebral angiography is the gold standard and first line treatment consists of endovascular embolization. In our case the CCF may have been caused by the intraoperative embolectomy of the occluded carotid artery, a complication described in the literature,¹⁵ but it also may be caused by HIV-related vasculopathy.

Yet, no specific guidelines are available. But despite the rapid advancement of endovascular techniques, we believe that open surgical repair remains the most appropriate option for extracranial carotid artery aneurysms, especially in HIV-pseudoaneurysms.

Lead author biography



Sandra Fraund-Cremer graduated from Heidelberg University School of Medicine in 1995. From 1995 to 2009 she underwent training to be a cardiac surgeon in Hannover Medical School (1995–1998) and University Hospital of Schleswig-Holstein, Kiel. Since 2009 she received additional vascular training and was board certified as a cardiovascular surgeon.

Supplementary material

Supplementary material is available at *European Heart Journal – Case Reports* online.

Slide sets: A fully edited slide set detailing these cases and suitable for local presentation is available online as [Supplementary data](#).

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

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