

# Coronary subclavian steal syndrome due to thrombosis of the left subclavian artery aneurysm: a case report

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Background	Coronary subclavian steal syndrome (CSSS) is an often easily overlooked cause of angina that may occur after a coronary artery bypass graft (CABG) procedure. The onset of CSSS several years after coronary revascularization has been described in case reports, and in the few retrospective reviews that compare the endovascular approach with surgical treatment. Subclavian stenosis can naturally coincide with coronary artery disease and may already be present during the initial CABG.
Case summary	A 59-year-old male with a history of three-vessel disease who had a left internal mammary artery (LIMA) bypass graft, exhibited a gradual worsening of angina that coincided with numbness and impaired function of the left fingers, hand, and arm. Myocardial per- fusion imaging showed reversible ischaemia, and coronary angiography suggested a thrombotic lesion proximal to the LIMA ostium. Calcified and partially thrombosed proximal left subclavian artery (LSA) aneurysm was visualized using computed tomography im- aging, whereas Doppler ultrasound revealed a partially reversed vertebral flow. The lowest risk treatment was a bypass between the left common carotid artery and the LSA. The procedure was immediately successful, with cessation of symptoms and a favourable medium-term outcome.
Discussion	As no guidelines exist for such cases, the importance of multidisciplinary co-operation in diagnostics and devising a treatment plan is underlined. Moreover, screening for subclavian artery stenosis in CABG candidates should be warranted as part of the initial pre- operative assessment.
Keywords	Coronary subclavian steal syndrome • Subclavian aneurysm • Subclavian stenosis • Case report
ESC Curriculum	2.1 Imaging modalities • 3.4 Coronary angiography • 7.5 Cardiac surgery • 3.1 Coronary artery disease • 9.3 Peripheral artery disease

#### Learning points

- Suspicion of subclavian artery stenosis should arise when inspecting a patient with previous coronary artery bypass graft (CABG), recurred angina pectoris, and differential blood pressure in upper extremities.
- Screening for subclavian artery stenosis in CABG candidates may be warranted as part of preoperative vascular assessment.
- Guidelines for optimal screening and treatment of coronary artery disease patients with coinciding, or secondary, proximal subclavian stenosis are lacking.

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#### Introduction

Aneurysms involving the supra-aortic arteries are rare, and 63% of such cases are a result of degenerative atherosclerosis.<sup>1</sup> The most common subclavian artery aneurysm (SAA) complications are thrombosis, rupture, and embolism.<sup>2</sup> Thus, thrombotic aneurysm can have a similar effect on distal haemodynamics to those of stenosis and occlusion.

Subclavian steal refers to a flow inversion in any branch of the subclavian artery (SA) due to significant proximal stenosis.<sup>3</sup> It develops in 1–4% of patients with left internal mammary artery (LIMA) graft, and is possibly an overlooked cause of angina pectoris (AP)<sup>4,5</sup> defined as coronary subclavian steal syndrome (CSSS). Subclavian artery stenosis in patients with prior coronary artery bypass grafting (CABG) can initiate myocardial ischaemia when a section of the coronary circulation is supplied via a LIMA graft<sup>3</sup> and there is at least >75% stenosis proximal to the LIMA ostium.<sup>5</sup> Such a stenosis results in an insufficient blood supply to maintain both myocardial perfusion and upper limb perfusion.

Here, we describe CSSS caused by thrombosed SAA and treated with open surgery.

### Timeline

Timeline of the patient's history, clinical presentation, diagnostics, and treatment. CABG, coronary artery bypass graft; LIMA-LAD, left internal mammary artery-left anterior descending (artery); 3VD, three vessel disease; RCA, right coronary artery; CCS, the Canadian Cardiovascular Society angina score; CSSS, coronary subclavian steal syndrome; SA, subclavian artery.

#### **Case presentation**

A 59-year-old male had a long history of coronary artery disease (CAD) and had undergone CABG 16 years before. Concurrent comorbidities included obesity (body mass index 33.4), Type 2 diabetes (T2DM), hypercholesterolaemia, sleep apnoea, and ankylosing spondylitis. The patient quit smoking 16 years before presentation but had smoked for 25 years prior to quitting.

The contemporaneous symptoms coincided with upper-limb steal syndrome, i.e. numbness and impaired function of the left fingers, hand, and arm. These symptoms persisted even without significant physical exertion. Blood pressure levels of the left limb were 30 mmHg lower than those of the right limb. Daily medication at the time of admission comprised bisoprolol, isosorbide mononitrate, aspirin, metformin, rosuvastatin, meloxicam, and buprenorphine patch, all within the recommended dosages.

Myocardial perfusion imaging with positron emission tomography (PET) (*Figure 1*) showed a perfusion defect. However, there was no significant stenosis visible in the left circumflex artery (LCX) in coronary angiography, and no suitable percutaneous coronary intervention (PCI) target could be identified (*Figure 2*). A non-critical ostial stenosis on a relatively small obtuse marginal branch was visualized and left to pharmacological treatment at this point. The left subclavian artery (LSA) appeared to have a stenosis proximal to the LIMA ostium. Doppler ultrasound flow spectre in the left vertebral artery was inversed during systole (*Figure 3*), and distal LSA exhibited attenuated, collateral-type currents, as did the axillary and the radial arteries.

Diagnostics were followed by a contrast CT-angiogram of the aorta and neck vessels, which showed a partially thrombosed aneurysm at the





Figure 1 Polar map images of myocardial perfusion at stress and at rest. A calculated polar map of perfusion reversibility. Reversible ischaemia in the left circumflex artery vascular region. Perfusion defect in the LAD supplied anterobasal region. No areas of significant perfusion deficiency at rest.



**Figure 2** Coronary angiography. (A) Right coronary artery remains clotted, with slow collateral inflow. (B) No stenosis in left main (LM) coronary artery, dominant branch of LM is left circumflex artery of and proximal narrowing in left obtuse marginal (LOM) artery, total blockage in LAD. (C) Plausible subclavian aneurysm is suggested by vessel lumen narrowing and turbulent flow in proximal left subclavian artery. Left internal mammary artery graft is open—no narrowing is visible proximally (C) or distally (D). Sternotomy fixation is visible in all projections.



Figure 3 Preoperative Doppler-ultrasound of the left vertebral artery. Flow is inverted during systole.

the presence and the extent of SA stenosis and its relation to other vascular structures, whereas Doppler ultrasound is essential in showing blood flow abnormalities confirming CSSS.

Most common pathologies behind the development of SAA are trauma, atherosclerosis, and thoracic outlet syndrome.<sup>6</sup> An iatrogenic traumatic mechanism could be ruled out for our patient, because neither the proximal SA nor the aortic arch was manipulated or cannulated during the primary procedure, which was performed using the aortic no-touch-technique. We were also unable to find any of the CABG-related iatrogenic SAA that are described in the existing literature.

A limited number of publications address the treatment of CSSS.<sup>3,3</sup> In previous case reports, angiography and stenting have been shown to have excellent outcomes in carefully chosen patients. Individual risk factors and the anatomic distribution of disease must be carefully weighted. For example, patients with ostial or diffuse atherosclerotic disease are less suited for percutaneous transluminal angioplasty and stenting, and patients who are elderly, or, have a poor medical condition overall, may be less tolerant of operative bypass.<sup>7</sup> We considered that stenting the aneurysm could have blocked the vertebral ostium in this case, and might have elevated the risk of iatrogenic thrombi migration towards the LIMA.

Surgical bypass (subclavian–subclavian or carotid–subclavian) can be a worthy alternative to angiography and stenting<sup>3,4</sup> and in some cases might be the only possible course of treatment for a patient with a challenging anatomy.<sup>8</sup> Possible surgical approaches include subclaviancarotid transposition,<sup>9</sup> carotid-subclavian bypass,<sup>10</sup> or relocation of the LIMA.<sup>11</sup> In cases of combined left SAA with ostial stenosis of the LIMA, extensive surgery, sternotomy, preoperative CABG, and repair of SAA may be necessary.<sup>7</sup> In a retrospective study by Song *et al.*,<sup>8</sup> both endovascular stenting and extrathoracic surgical bypass were found to be safe and effective treatments for subclavian steal syndrome (SSS) in the short and medium term. They also found that extrathoracic surgical bypasses were more durable in the long-term.<sup>8</sup>

A population with CAD often has concomitant extracardiac atherosclerosis, and it is not rare to find a significant (asymptomatic) SA stenosis.<sup>12</sup> Screening for SA stenosis in CABG candidates may be warranted, and relevant imaging should be performed for patients with a history and physical examination findings that suggest SA stenosis.<sup>13</sup> The surgical approach of coronary revascularization can be modified prospectively when stenosis or occlusion of the left SA is identified before CABG.<sup>4</sup> Guidelines for optimal screening and treatment approaches to CAD patients eligible for CABG with combined proximal subclavian stenosis are lacking.<sup>14</sup> Moreover, to the best of our knowledge there are no studies that compare endovascular vs. surgical revascularization of SAS in a CABG candidate.<sup>15</sup>

Here, we showcased a rare case of CSSS caused by a thrombosed SAA. The ever-increasing life expectancy after CABG may also be associated with an increase in the incidence of CSSS. Thus, a suspicion of SA stenosis should be kept in mind when examining a patient with previous CABG, recurred AP, and differential blood pressure of the upper extremities.

As no guidelines exist for such cases, the value of multidisciplinary cooperation is especially important in both diagnostics and in devising of the treatment plan.

#### Patient perspective

The patient in question appeared to understand both the severity of his disease and the risks involved in the treatment. He had no opinion as to which route to take for diagnostics or the choice of treatment. Overall, the patient remained exceedingly satisfied with the treatment he received and the outcome after several months. The patient now has

**Figure 4** Computed tomography angiography; preoperative (A and B) and postoperative (C and D) 3D reconstructions (A and C) and key CT (B and D) images of the subclavian aneurysm and surrounding vascular structures. Bypass graft (k) is open and functioning in the postoperative images (C and D). e: subclavian aneurysm, f: distal subclavian artery, g: left vertebral artery, h: left internal mammary artery-LAD graft, i: left carotid artery, j: cross-sections of the aneurysm (1–4; superior to inferior slice), and k: carotid-subclavian bypass graft.

base of the LSA with significant distal calcification and stenosis (Figure 4A and B).

Imaging results outlined a thrombosed LSA aneurysm and stenosis that were causing hypoperfusion of the upper limb and the LIMA graft. The patient had near-disabling symptoms, severe CAD, and the coronary bypass graft that provided most of myocardial blood supply was at risk of losing the entire inflow.

A multidisciplinary team concluded that the procedure with the lowest risk would be a prosthetic carotid-subclavian bypass. The procedure was performed under general anaesthesia with cerebral oxygenation and oesophageal echocardiography monitoring. The patient recovered well and was discharged on the third postoperative day.

The patient reported asymptomatic upper-limb function and normal physical performance at one month after the operation. The systolic finger pressures were symmetrical and the graft(s) appeared open upon Doppler ultrasound. Three months after the procedure, the bypass graft (*Figure 4*) LSA and vertebral artery showed regular contrast agent enhancement. The patient remained asymptomatic.

#### Discussion

According to the existing literature, CSSS is an often overlooked cause of AP that can easily remain undiagnosed.<sup>5</sup> Reaching the correct diagnosis for our patient required the utilization of several imaging modalities. Magnetic resonance angiography (MRA)/computed tomography angiography (CTA) and standard angiography are crucial to revealing



significantly improved physical performance and cessation of cardiac symptoms.

#### Lead author biography



Dr Max Kiugel (MD, PhD) graduated with MSc in Drug Development and Pharmacology from the University of Turku, Finland, in 2010, and completed his PhD at Turku Pet Centre in 2018, where he was working with novel PET-tracers in translational cardiovascular models. In 2019 Dr Kiugel graduated from the same university as MD, and, after working in general medicine, and emergency medicine, was accepted into the Vascular Surgery Residency Program at Turku University Hospital

in 2020. He moved to the private sector in September of 2021, and has now been working as a Research Physician in a private clinical research company CRST-OY, where he is involved in designing and conducting Phase I–IV clinical drug trials.

#### Supplementary material

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

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**Slide sets:** A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

**Consent:** The patient gave his written consent to publish this case report in accordance with COPE guidelines.

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#### References

- Cury M, Greenberg RK, Morales JP, Mohabbat W, Hernandez AV. Supra-aortic vessels aneurysms: diagnosis and prompt intervention. J Vasc Surg 2009;49:4–10.
- Vierhout BP, Zeebregts CJ, van den Dungen JJ, Reijnen MM. Changing profiles of diagnostic and treatment options in subclavian artery aneurysms. *Eur J Vasc Endovasc Surg* 2010;40:27–34.
- 3. Potter BJ, Pinto DS. Subclavian steal syndrome. Circulation 2014;129:2320-2323.
- Faggioli G, Pini R, Cremonesi A, Grattoni C, Longhi M, Mauro R, et al. Endovascular treatment of late coronary-subclavian steal syndrome. J Thorac Cardiovasc Surg 2014; 148:2112–2116.
- Lak HM, Shah R, Verma BR, Roselli E, Caputo F, Xu B. Coronary subclavian steal syndrome: a contemporary review. *Cardiology* 2020;**145**:601–607.
- Maskanakis A, Patelis N, Moris D, Tsilimigras DI, Schizas D, Diakomi M, et al. Stenting of subclavian artery true and false aneurysms: a systematic review. Ann Vasc Surg 2018;47: 291–304.
- Douglas DA, Adelman MA, Esposito R, Rockman C. Surgical repair of a left subclavian artery aneurysm causing stenosis of a left internal mammary graft-a case report. Vasc Endovascular Surg 2005;39:281–285.
- Song L, Zhang J, Li J, Gu Y, Yu H, Chen B, et al. Endovascular stenting vs. extrathoracic surgical bypass for symptomatic subclavian steal syndrome. J Endovasc Ther 2012;19: 44–51.
- Cinà CS, Safar HA, Laganà A, Arena G, Clase CM. Subclavian carotid transposition and bypass grafting: consecutive cohort study and systematic review. J Vasc Surg 2002;35: 422–429.
- Paty PS, Mehta M, Darling RC III, Kreienberg PB, Chang BB, Roddy SP, et al. Surgical treatment of coronary subclavian steal syndrome with carotid subclavian bypass. Ann Vasc Surg 2003;17:22–26.
- Chung DA, Large SR. Relocation of the internal mammary artery graft in a case of coronary-subclavian steal. *Thorac Cardiovasc Surg* 2000;48:39–40.
- Marques KM, Ernst SM, Mast EG, Bal ET, Suttorp MJ, Plokker HW. Percutaneous transluminal angioplasty of the left subclavian artery to prevent or treat the coronarysubclavian steal syndrome. Am J Cardiol 1996;78:687–690.
- Osborn LA, Vernon SM, Reynolds B, Timm TC, Allen K. Screening for subclavian artery stenosis in patients who are candidates for coronary bypass surgery. *Catheter Cardiovasc Interv* 2002;56:162–165.
- Westerband A, Rodriguez JA, Ramaiah VG, Diethrich EB. Endovascular therapy in prevention and management of coronary-subclavian steal. J Vasc Surg 2003;38:699–703.
- Cua B, Mamdani N, Halpin D, Jhamnani S, Jayasuriya S, Mena-Hurtado C. Review of coronary subclavian steal syndrome. J Cardiol 2017;70:432–437.