# Brief Report

# Coil embolization of iatrogenic arterio-venous fistula from pulmonary collateral artery to subclavian vein

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**Abstract** A young woman with surgically treated tetralogy of Fallot with pulmonary atresia, with multiple aorto-pulmonary collateral arteries, developed an arteriovenous fistula subsequent to transvenous insertion of an automatic implantable defibrillator via the left subclavian vein. The fistula extended between this vein and a systemic-to-pulmonary collateral artery arising from the subclavian artery. Unilateral peau d'orange, and painful congestion of the left arm and breast, ensued. These were cured by coil embolization of a fistula-related aneurysm.

Keywords: Arteriovenous fistula; therapeutic embolization; peau d'orange (non-malignant); pulmonary atresia; pulmonary collateral artery

HEN FEASIBLE, PERCUTANEOUS EMBOLIZATION of arteriovenous fistulas is a desirable alternative to surgery. Such techniques have been used for coronary arterial fistulas<sup>1</sup> and iatrogenic femoral<sup>2</sup> and subclavian<sup>3</sup> arteriovenous fistulas occurring subsequent to invasive vascular procedures. We report a case of successful transcatheter closure using coils of an iatrogenic left subclavian fistula caused by transvenous insertion of an implantable defibrillator. There was dramatic relief of the symptoms of congestion and stasis of the left arm, and elimination of nonmalignant peau d'orange of the left breast.

### Case report

A young woman had a history of tetralogy of Fallot with pulmonary atresia and multiple systemicto-pulmonary collateral arteries. During childhood, she underwent several aorto-pulmonary shunt operations, as well as surgical division of large collateral arteries to the left lung via a left thoracotomy. An episode of Staphylococcal endocarditis, with a mitral valvar vegetation, was treated medically, except for

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surgical drainage of a sternal abscess. In 1991, she had repair of the pulmonary atresia, with a valved conduit placed from the right ventricle to the pulmonary arteries and dacron patch closure of the ventricular septal defect. The following year, she was resuscitated from a cardiac arrest and electrophysiological study demonstrated sustained monomorphic ventricular tachycardia. Despite antiarrhythmic therapy with amiodarone, ventricular tachycardia was still inducible, and she required insertion of an automatic implantable defibrillator via the left subclavian vein. Four years later, she required replacement for battery depletion. The old lead had a high defibrillation threshold but could not be extracted. Therefore it was capped, and a new lead inserted percutaneously via the left subclavian vein with a left infraclavicular pocket for the generator device. Bruising and haematoma occurred, and a thrill and continuous murmur became evident.

When the bruising settled, cardiac catheterization was performed. This demonstrated an arteriovenous fistula between a large collateral artery and the left subclavian vein. The collateral arose from the left subclavian artery and supplied the upper lobe of the left lung. The calculated shunt ratio was 1.2:1. In addition, there was an elevated right ventricular pressure and complex stenosis of the left pulmonary artery with a large distal aneurysm. A decision was made to continue conservative management of both

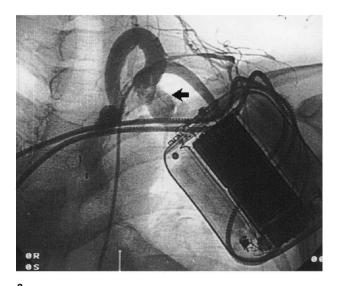
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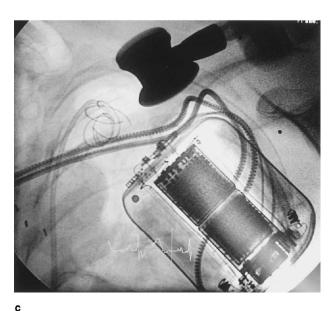
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#### Figure 1.

Clinical photog raph of patient's chest showing asymmetry of breasts and surgical scars. The left breast is enlarged and there is peau d'orange of the areola and surrounding tissue.

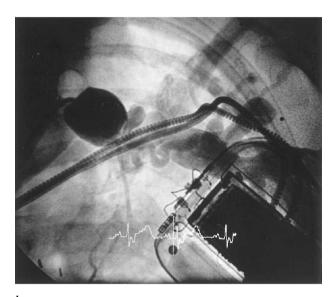




the fistula and the complex left pulmonary arterial anatomy because of surgical difficulties.

She subsequently developed a tender, swollen left arm and was rehospitalized six months later because of more acute pain and congestion. Venography showed apparent obstruction of the left subclavian vein at its origin, and extensive collateral veins in the chest wall. She was treated with heparin, warfarin, and the arm was elevated. The anticoagulant regime was later changed to aspirin. There was continued discomfort of her left arm, along with changed colour, size and consistency. She also developed enlargement and peau d'orange of the left breast (Fig. 1).

A further catheterization was performed, and the fistulous communication was entered selectively. A fistula-related aneurysm was found with near systemic blood pressure (Fig. 2a). Hand injections of contrast





#### Figure 2.

An implantable defibrillator (a) is seen with two leads in the subclavian vein. The uncoiled one could not be extracted. Angiography from a catheter in the left subclavian artery demonstrates a looping collateral artery arising from the subclavian artery to supply the upper lobe of the left lung. An aneurysm (arrowed) is related to the arterio-venous fistula, which arises near the origin of the collateral artery. A right Judkins catheter (b) was passed retrogradely from aorta to left subclavian artery to the origin of its collateral artery and directly into the aneurysm related to the arteriovenous fistula. The pressure in this aneurysm was near systemic levels. A hand injection of contrast has outlined the aneury sm and multiple dilated venous collaterals, as well as the subclavian vein. Coils (c) have been embolized into the aneurysm via a right Judkins catheter passed retrogradely from the aorta through the arteriovenous fistula. Already the continuous murmur had disappeared, as confirmed by the anaesthetist's stethoscope. Complete occlusion was achieved with a total of 18 coils.

filled the aneurysm, and filled multiple venous collateral veins retrogradely (Fig. 2b). The anatomy was deemed suitable for coil embolization. A total of 18 Cook embolization coils (38-5-12) were inserted (Fig. 2c). This resulted in formation of thrombus in the aneurysm, and an immediate decrease in the venous collaterals. The murmur disappeared and the patient began to notice relief of symptoms by the following day. Over the ensuing weeks, the swelling of the hand, arm and breast all resolved, and her unilateral peau d'orange was also cured.

Subsequently, she has required a new implantable defibrillation system. Both old leads were extracted surgically from the right ventricle and a new transvenous lead successfully installed.

## Discussion

Iatrogenic arterio-venous fistulas are recognized complications of vascular catheterization from femoral,<sup>2</sup> internal jugular, and subclavian approaches,<sup>3</sup> resulting from penetration of the vessels and trauma. Pacemaker and defibrillator leads were identified as culprits in several previous reports.<sup>4–6</sup> Coil embolization techniques have been used instead of surgery to block these channels since 1993, and are generally effective.<sup>4,7</sup>

In our case, there was the unusual involvement of a systemic-to-pulmonary collateral artery in the fistula. It arose from the subclavian artery, looped upwards, and then down into the chest cavity. This vessel had remained undiagnosed because it was not visualized by previous aortography. Multiple selective angiograms are usually needed for delineation. Such a vessel would have contributed to the less than ideal result from surgical correction of her congenital cardiac lesion. The need for a defibrillator also relates to an imperfect repair, with elevated right ventricular pressure. Of course, the original poor quality of her native pulmonary arteries made corrective surgery difficult.

Left arm venography in our patient suggested there was thrombus in the left subclavian vein.

Subsequently, it appeared that the near systemic arterial pressure in the fistula-related aneurysm was enough to prevent forward venous flow. Thus, the patient became symptomatic, with venous congestion and pain and heaviness in the left arm. This also prevented lymphatic flow into the venous system, producing non-malignant peau d'orange of the left breast. Presumably this lymphatic obstruction resulted from the high venous pressure in the veins to which the lymphatics drain. These include the internal mammary and intercostal veins. All these features resolved with therapeutic coiling. Pain and swelling, peau d'orange, and the continuous murmur all disappeared.

We conclude by agreeing with previously expressed views that catheter embolization of subclavian arterio-venous fistulas should be performed in asymptomatic patients to prevent potential complications.<sup>4,7</sup>

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