

Repercussions of a giant major aortopulmonary collateral aneurysm adjacent to vertebral artery origin

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ABSTRACT

An aneurysm of major aortopulmonary collateral in an adult with congenital cyanotic heart disease was detected incidentally following a hemoptysis episode. The location and size of the aneurysm needed special concern during treatment to avoid aneurysm-related complications and thromboembolism secondary to intervention.

Keywords: Cerebral embolic complications, hemoptysis, major aortopulmonary collateral aneurysm, major aortopulmonary collateral embolization

INTRODUCTION

Major aortopulmonary collaterals (MAPCA) are commonly associated with reduced pulmonary blood flow in congenital heart diseases. Long term outcomes of untreated MAPCAs are less known as early detection and surgical or endovascular repair are available in the current era. We present a case of untreated MAPCA in an adult patient who had congenital heart disease, who got lost to clinical follow-up and presented as a complication of MAPCA.

CLINICAL SUMMARY

A 28-year-old male who underwent Glenn surgery for tricuspid atresia and infundibular pulmonary stenosis [Figure 1a] at the age of 3 years before Fontan repair was lost to clinical follow-up and presented after 25 years with an episode of massive hemoptysis. At presentation, he had cyanosis (Spo₂ of 78%) and clubbing. Hemogram revealed polycythemia (PCV - 69) with deranged INR (4.3) and normal leukocyte counts.

Contrast computed tomography (CT) angiogram revealed preserved flow in Glenn with adequately sized pulmonary arteries (right pulmonary artery 14 mm and left pulmonary artery 10 mm) and hypertrophied collaterals supplying both lungs [Figure 1b and e]. A 3.1 cm × 2.6 cm × 2 cm fusi-saccular aneurysm [Figure 1b and c] involving the proximal course of a major aortopulmonary collateral (MAPCA) arising from the first part of the right subclavian artery was noted and the distal branches of this MAPCA were enlarged and supplying bilateral lung segments. The aneurysm was not showing any thrombus or features of rupture.

He was not considered for the completion of Fontan surgery due to high pulmonary arterial pressures (mean of 25 mmHg).

After correcting coagulopathy, palliative embolization of enlarged MAPCAs to reduce the chances of further bleeding and reduce chances of further hemoptysis

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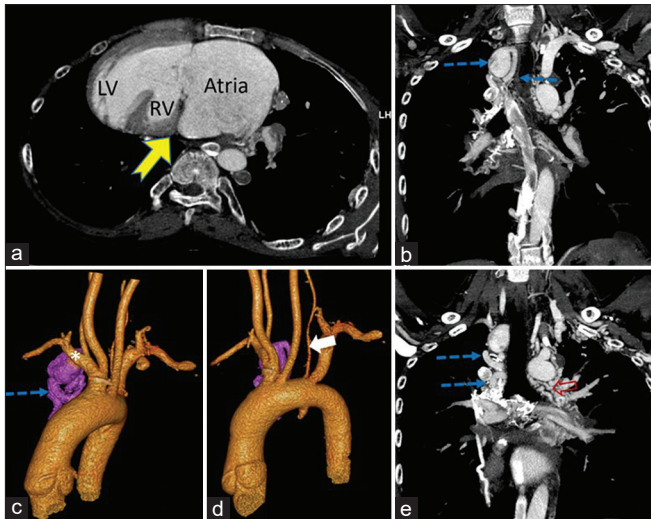


Figure 1: Computed tomography angiogram images. (a) Oblique axial section showing atric tricuspid valve (tailed arrow) and hypoplastic right ventricle and enlarged atria. (b and e) Coronal section showing a large aneurysm along the course of collateral (blue dotted arrows) and branching to both lungs. Lots of tiny collaterals are seen in the left mediastinal hilum (open red arrow). (c and d): Volume rendered reconstruction with aneurysmal major aortopulmonary collateral in pink color seen arising from right subclavian artery (*) close to right vertebral artery origin. The left vertebral artery (white arrow) is having a direct origin from left-sided aortic arch and is hypoplastic compared with right one

was performed [Figure 2]. Selective angiograms were performed using a 2.7 French (F) progreat microcatheter (Terumo, Somerset, NJ) keeping 5 F right coronary catheter as a guide close to MAPCA origin. 50% n-butyl-2-cyanoacrylate glue (Histoacryl, B. Braun, Melsungen, Germany) was slowly injected in the distal segment of MAPCA by sandwich technique to avoid distal embolization [Figure 2c-e] and subsequently, a 20 mm × 50 cm fibred coil (Concerto detachable coil system, Medtronic, Minneapolis) was deployed in the proximal aneurysm attaining complete occlusion of the collateral and aneurysm. All large collaterals of diameter >3 mm were subsequently embolized using fiber coils and glue [Figure 2f]. The hemoptysis was controlled, and the patient got discharged with Spo2 maintained around 80%.

DISCUSSION

Long-term outcomes of untreated MAPCAs are less known as early detection and surgical or endovascular repair are available in the current era. Although the occurrence of stenosis and poststenotic dilations is common in MAPCAs, reports of aneurysm along course are only hand full.^[1-4] Flow-related dilation is common in unobstructed MAPCAs resulting from segmental pulmonary hypertension.^[5]

Aneurysm from MAPCA may present as dyspnea or hemoptysis as a result of its mass effect.^[3,4] Rupture of aneurysm in MAPCA is a medical emergency and various

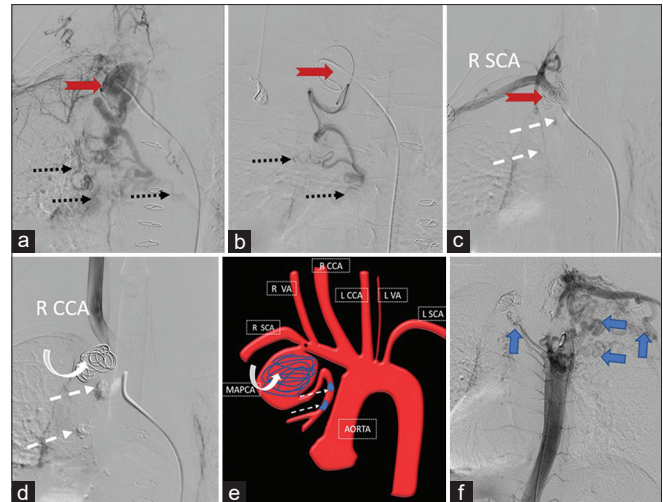


Figure 2: DSA images of aneurysmal major aortopulmonary collateral during embolization. (a) Angiogram from origin of major aortopulmonary collateral with aneurysm along the course of major aortopulmonary collateral beginning from its ostium (red tailed arrow) and extensive branching to bilateral lung segments (black dotted arrows). (b) Selective angiogram from major aortopulmonary collateral distal to aneurysm before embolization. The location of aneurysm is shown by red tailed arrow. (c) Right subclavian artery angiogram post coiling showing good opacification without any prolapse or thrombosis in subclavian branches. The aneurysm (red) tailed arrow is totally thrombosed. (d and e) Glue cast in vertical segment of major aortopulmonary collateral (white dotted arrows) and coil mass inside aneurysm (white curved arrow) close to right vertebral artery origin (*) and good flow in right vertebral artery when injected from innominate artery. F-Aortogram from descending aorta showing multiple collaterals in left and right hemithorax (blue solid arrows) which were embolized subsequently. L CCA: left common carotid artery, L SCA: left subclavian artery, L VA: left vertebral artery, R CCA: Right common carotid artery, R SCA: Right subclavian artery, R VA: Right vertebral artery

treatment options such as covered stent or vascular plug or surgery have been tried.^[1,2,4] Conservative management for giant MAPCA aneurysm considering the intraoperative risks had a dismal outcome.^[3]

The proximity of vertebral artery and other subclavian artery branches [Figures 1c and 2d] close to this MAPCA warranted against a covered stent in the subclavian artery, jailing the MAPCA. Hypoplasia of the contralateral vertebral artery [Figure 1d] mandated the right vertebral flow preservation to avoid arterial insufficiency or stroke in the posterior brain circulation.

Posterior circulation thromboembolic complications can occur during treatment of subclavian artery aneurysms close to vertebral artery origins.^[6] Injecting liquid embolic agent directly in the aneurysmal sac may result in inadvertent cerebral or pulmonary thromboembolic complications because of proximity of vertebral artery origin or unpredictable flow dynamics in aneurysm [Figures 1c and 2e]. Intraprocedural rupture or occlusion of the adjacent artery is common with vascular occlusive devices. The purpose of deploying

a large fibred coil in the aneurysm after occluding the distal MAPCA with glue was to contain the accelerated thrombosis occurring in the aneurysm and avoiding its extension to subclavian artery [Figure 2c-e]. The coil mass which is larger than ostium of the MAPCA will act as a scaffold for thrombosis and prevents its extension in to subclavian artery.

CONCLUSION

MAPCA-related aneurysms may present as a catastrophe if not treated. Combination of embolic agents can be used in occlusion of MAPCA aneurysms. Thromboembolic complications during treatment should be avoided if MAPCAs or aneurysms are close to cerebral blood vessels using suitable interventional or hybrid techniques.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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