Vertebrobasilar insufficiency after subclavian flap aortoplasty for aortic coarctation

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ABSTRACT

The mainstay of treatment of pediatric aortic coarctation is open surgery. One option for repair includes subclavian flap aortoplasty, first described by Waldhausen and Nahrwold in 1966. Within this technique, several modifications have been made over the years as long-term follow-up data became available. Early outcomes revealed little concern for left upper extremity limb ischemia or subclavian steal syndrome. These complications are rare but can have a significantly delayed presentation years after coarctation repair. We present a case of subclavian steal syndrome with lifestyle-limiting vertebrobasilar symptoms experienced by a patient 36 years after subclavian flap aortoplasty for aortic coarctation. (J Vasc Surg Cases Innov Tech 2024;10:101409.)

Keywords: Aortoplasty; Coarctation; Steal syndrome

Long-segment aortic coarctation is common, encompassing 5% to 8% of all congenital heart defects and can be treated using several operative techniques, including resection and anastomosis, endovascular balloon dilation, patch aortoplasty, and subclavian artery turndown. Resection and anastomosis can lead to recurrent stenosis secondary to stricture, endovascular techniques can lead to recurrence, and patch aortoplasty can lead to aneurysmal degeneration.[1] In an attempt to avoid circumferential anastomosis, subclavian aortoplasty was developed. Subclavian aortoplasty procedures have evolved over time as long-term outcomes have suggested modifications in the procedure. The traditional subclavian flap procedure includes ligation of the left subclavian artery just proximal to the vertebral artery and transforming the proximal portion of the subclavian artery into a flap to widen the coarcted segment of the descending aorta (Fig 1).[1] Initially, there was little concern for upper extremity ischemia or subclavian steal syndrome. Therefore, the subclavian artery was not usually revascularized after ligation, and the vertebral artery was not ligated or reimplanted. Cases of limb-length discrepancy[2–6] and left upper extremity limb ischemia[7] have prompted modifications in the procedure to allow for preserved arterial blood flow to the left upper extremity by avoiding ligation of the left subclavian artery[8] or revascularization of the left subclavian with interposition grafting.[9] Additionally, there have been reports of delayed presentation with subclavian steal syndrome ≤24 years after treatment.[10,11] Thus, in some cases of subclavian aortoplasty, the left vertebral artery is ligated.[7,10] We present the case of a patient who developed symptoms of subclavian steal syndrome 36 years after subclavian aortoplasty without vertebral artery ligation. The patient provided written informed consent for the report of her case details and imaging studies.

CASE REPORT

Our patient is a 40-year-old woman with a history of possible coarctation of the aorta with open repair via subclavian flap aortoplasty without vertebral ligation in 1987. Because of the remote coarctation surgery, no operative reports were available. She was previously morbidly obese but underwent laparoscopic biliopancreatic diversion with duodenal switch with excellent weight loss. Her medical problems include hypertension and hypothyroidism, which are managed with levothyroxine and verapamil. The patient was referred to the vascular surgery clinic for subclavian steal syndrome. She had been experiencing symptoms of vertigo, diplopia, and lightheadedness for several years and seen multiple specialists, including her cardiologist and neurologist. She has undergone extensive diagnostic testing and tried multiple medications without improvement. She underwent carotid duplex ultrasound studies, which revealed 50% to 69% stenosis of the right internal carotid artery and <50% stenosis of the left internal carotid artery with retrograde flow in the left vertebral artery (Fig 2). She underwent aortic arch, vertebral/carotid artery, and intracranial angiography, which demonstrated significant reversal of flow in the left vertebral artery (Figs 3 and 4). The patient was subsequently referred to the vascular surgery clinic for further evaluation.

Her diagnostic angiogram revealed brisk, antegrade flow through the innominate artery and right subclavian, right carotid, and left carotid arteries. The left subclavian artery origin was not visualized, and the distal left subclavian artery was only seen on delayed images revealing reversal of flow through a large left vertebral artery supplying the left subclavian artery

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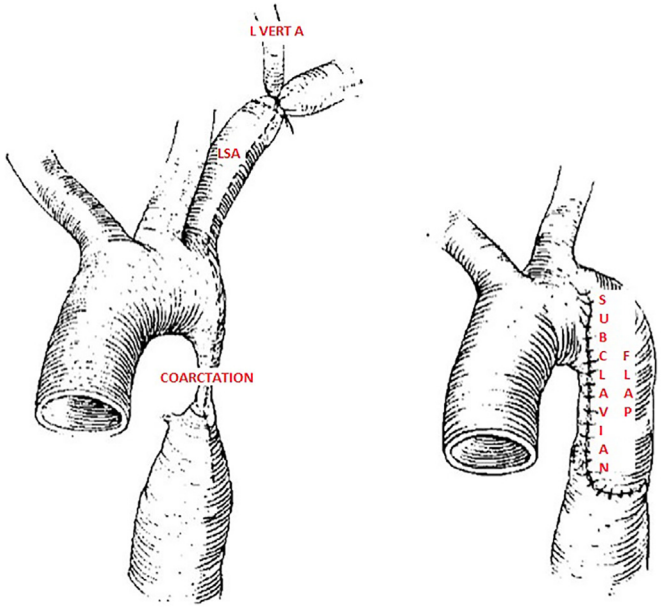

Fig 1. Subclavian flap repair for coarctation of the aorta. *LSA*, Left subclavian artery; *L VERT A*, left vertebral artery.

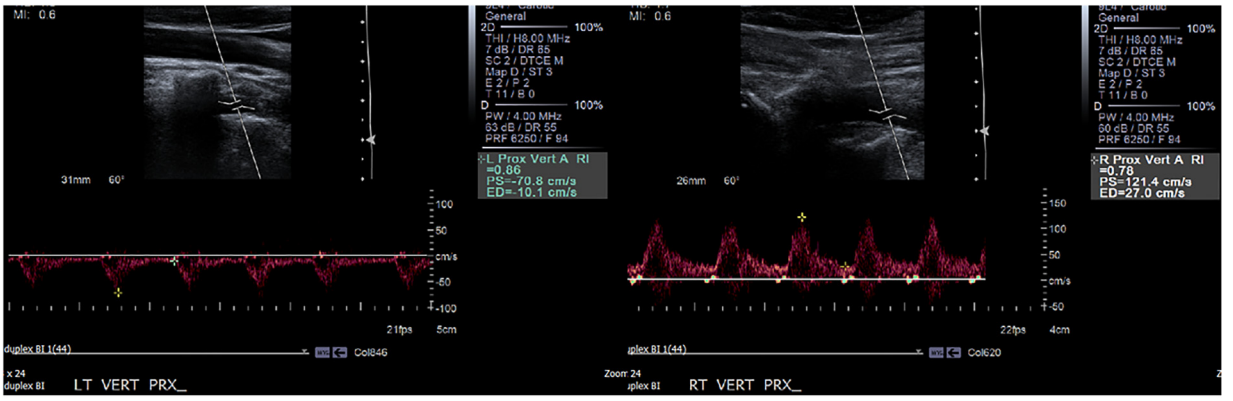

Fig 2. Carotid duplex ultrasound revealing retrograde flow in the left vertebral artery.

(Fig 3). This was demonstrated again when the right vertebral artery was selectively catheterized, with demonstration of antegrade flow through the right vertebral artery into the basilar artery and brisk retrograde flow into an enlarged left vertebral artery measuring 1 cm in diameter.

The patient continued to experience symptoms of diplopia and dizziness. She denied syncopal episodes. Her symptoms were not reproducible or associated with any specific positioning or activity, specifically the use of her left upper extremity. However, her symptoms were so profound, she was unable to perform daily activities. She denied any symptoms of left arm claudication. There was no upper extremity limb length discrepancy. On physical examination, she did not have a significant discrepancy between the right brachial and left brachial blood pressure measurements. Her radial pulses were palpable bilaterally. The patient was counseled that although her symptoms did have similarities, they were not discretely consistent with subclavian steal syndrome. However, because her symptoms were disabling, we believed surgical intervention was warranted. After extensive discussion, she wished to proceed with operative intervention with left carotid to subclavian artery bypass.

Therefore, patient underwent uneventful left carotid to subclavian artery bypass with an 8-mm Dacron graft. Her postoperative course was uncomplicated. She was discharged home on postoperative day 1. She was seen in the outpatient office at 2 weeks postoperatively, at which time all her vertebrobasilar symptoms had completely resolved. She denied any headaches, vertigo, diplopia, and lightheadedness. Her quality of life is vastly improved. Repeat carotid artery duplex ultrasound revealed antegrade flow in the left vertebral artery (Fig 5). She continues to do well at 3-month follow-up with no recurrence of her symptoms.

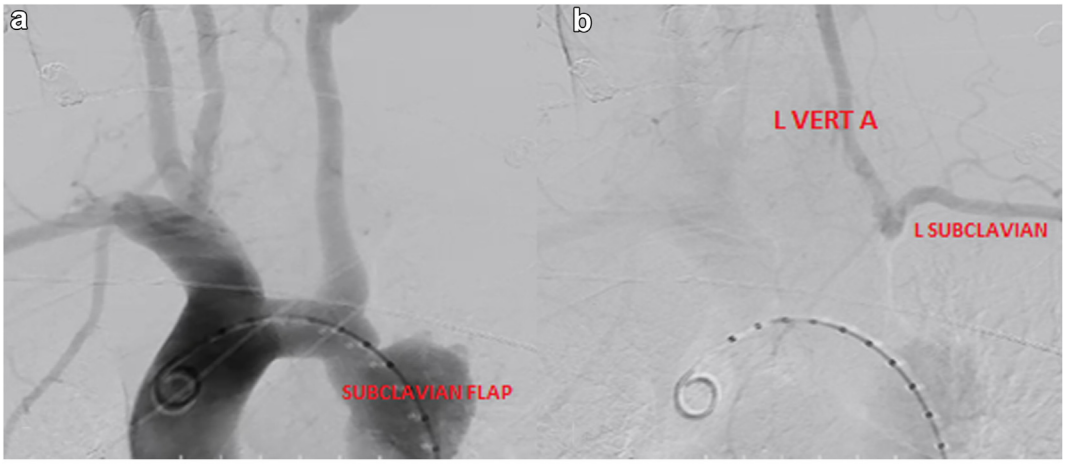

Fig 3. Aortogram showing ligated proximal left subclavian artery (**A**), with retrograde filling of the left vertebral artery (**B**). *L SUBCLAVIAN*, Left subclavian artery; *L VERT A*, left vertebral artery.

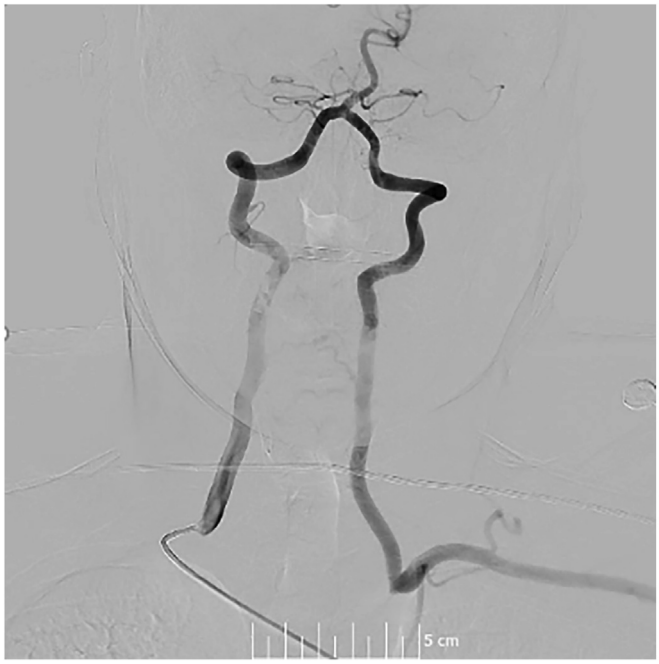

Fig 4. Cerebral angiogram revealing large vertebral arteries bilaterally with retrograde flow in the left vertebral artery.

DISCUSSION

Subclavian flap aortoplasty, as depicted in Fig 1, includes ligation of the left subclavian artery just proximal to the vertebral artery and transforming the proximal portion of the subclavian artery into a flap to widen the coarcted segment of the descending aorta. Subclavian steal syndrome is rare after subclavian flap aortoplasty. However, when it does occur, it seems to occur in a delayed fashion during adulthood.[10,11] It is unclear why patients might be asymptomatic for >1 decade before developing neurologic symptoms.

Carotid to subclavian artery bypass allows for antegrade left vertebral flow, resolving neurologic symptoms. Subclavian aortoplasty was performed often in the 1970s and 1980s but is now rarely performed due to recurrent coarctation rates of ≤42% and significant risks of limb ischemia, limb dysfunction, subclavian steal syndrome, and aneurysmal degeneration.[2,3,7,10,12] The incidence of these complications after subclavian flap aortoplasty is not well defined.

In a study performed by Khalil et al[13] comparing subclavian flap aortoplasty and extended end-to-end anastomosis, limb ischemia was a complication in 3 of 35 patients in the subclavian flap aortoplasty group vs 0 of 45 patients in the extended end-to-end anastomosis group. A study by Kopf et al[5] demonstrated no upper limb ischemia in 23 patients undergoing subclavian flap aortoplasty. Todd et al[3] demonstrated a significant limb-length discrepancy between the upper extremities after subclavian flap aortoplasty but reported that the patients had minor symptoms.

Retrograde vertebral flow rarely causes symptoms after subclavian flap aortoplasty. Saalouke et al[14] reported four cases of subclavian steal syndrome after coarctation repair with vertebrobasilar symptoms, which were treated with ligation of the left vertebral artery or subclavian angioplasty. However, most patients with evidence of subclavian steal syndrome on cerebral arteriography performed after coarctation repair were asymptomatic. Additionally, only one of the patients had undergone ligation of the subclavian artery with the coarctation repair.[13]

Aneurysmal degeneration is a long-term concern, most often seen with patch angioplasty, but is reported in ≤17% after subclavian flap aortoplasty.[15,16] Aneurysms occur 6 to 16 years after the initial coarctation surgery.[6] In cases of aneurysmal degeneration, repair can be performed via an open or endovascular approach. Chiesa

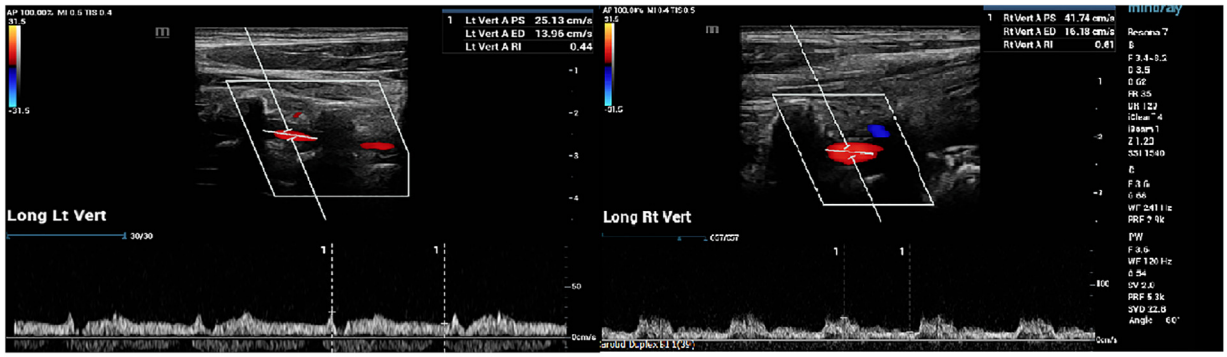

Fig 5. Postoperative carotid duplex ultrasound showing antegrade flow in the left vertebral artery. *Long Rt Vert*, Long right vertebral artery.

et al[12] described a case of aneurysmal degeneration 25 years after Dacron patch aortoplasty treated with open surgical repair with deep hypothermic circulatory arrest in a patient with anatomy unsuitable for endovascular repair. An open approach can involve complex reconstruction with a significant risk of paraplegia, nerve injury, and bleeding and mortality rates of ≤23.5%.[16] Therefore, endovascular or hybrid techniques, including the use of thoracic stent grafts to exclude the aneurysmal segment with or without debranching, are more frequently used. Thoracic endovascular aneurysm repair has been shown to be successful in excluding the aneurysmal segment with low mortality and morbidity and long-term patency without the need for reintervention.[6]

CONCLUSIONS

Due to the poorly defined complication rates, modifications to subclavian flap repair are made on a case-by-case basis, because no consensus has been reached regarding ligation of the vertebral artery or revascularization of the subclavian artery.[3,7,10] Today, most coarctation repairs are performed with an extended end-to-end anastomosis, with ≤5% performed with subclavian flap aortoplasty.[1] There are several known complications that can occur in a delayed fashion, ≤25 years or more after surgery for aortic coarctation. Subclavian steal syndrome can develop and become debilitating, necessitating surgical repair. Aneurysmal degeneration can occur with a significant risk of rupture. Patients with previous coarctation repair will need lifelong surveillance and consideration for repair if aneurysmal degeneration occurs. Patients with vertebrobasilar symptoms and a history of subclavian aortoplasty should be evaluated for subclavian steal syndrome and offered surgical intervention if their symptoms are significant.

DISCLOSURES

None.

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