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Intermittent right upper extremity ischemia in a patient with simultaneous aberrant right subclavian artery and patent foramen ovale: A case report

Abstract

Background: Upper extremity intermittent ischemia due to non-aneurysmal, not occluded aberrant right subclavian artery (ARSA) is rare.

Case presentation: We describe a 30-year-old male who suffered from PFO and non-aneurysmal, not occluded ARSA, and presented by intermittent right upper extremity ischemia. He was treated by right carotid subclavian transposition for ARSA and antiplatelet medication for PFO.

Conclusion: Authors assume that intermittent limb ischemia can occur secondary to anatomical changes in a patient without aneurysmal degeneration or occluded ARSA and the existence of pure PFO without any evidence of venous thrombosis is not enough to prove the paradoxical emboli scenario.

Keywords: Aberrant right subclavian artery, Patent foramen ovale, intermittent limb ischemia

Citation:

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A berrant right subclavian artery (ARSA) occurs in 0.5% to 1% of the population (1) and the upper extremity ischemia is a clinical presentation in the aneurysmal or occluded ARSA (2). In a patient with non-aneurysmal or not occluded ARSA, the acute ischemia of right upper extremity with the embolic pattern is due to other etiologies such as arterial fibrillation or paradoxical embolism from a patent foramen ovale (PFO) (3) or is a subsequent complication of arterial thoracic outlet syndrome. Patent foramen ovale (PFO) can increase the incidence of paradoxical embolism and followed by the risk of cryptogenic stroke as well as peripheral embolism (4). The size of PFO and the presence of the source of venous thrombi are two main risk factors for ischemic events (5). Despite previous reports for cryptogenic stroke that concerning paradoxical embolism through a PFO, the magnitude of this etiology as a risk factor for acute ischemic events in a patient with ARSA remains undefined, because deep vein thrombosis is infrequently detected in such patients (6). Therefore, in patients with non-aneurysmal or not occluded ARSA who suffer from PFO is a challenging discussion, which one is the main cause of intermittent acute ischemia and what is the best treatment of choice? We present a patient with both aberrant right subclavian artery and patent foramen ovale who had suffered repeated acute upper extremity ischemia. The patient's consent for publication was obtained.

Case Presentation

A 30-year-old male patient referred to the emergency department with acute right upper extremity paresthesia that gradually worsened.

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He was cautious and the vital signs were in the normal range (blood pressure: 120/80 mmHg, heart rate: 80 per minute, respiratory rate: 16 per minute and axillary temperature: 37.2 C). There was no history of any comorbidities and traumatic events. Physical examination revealed both radial and ulnar artery pulses were weaker on the right side. He was admitted with primary diagnosis of cerebrovascular events. Brain computed tomography (CT) scan was done and there was not any evidence of cerebrovascular pathologies. Cardiac monitoring showed normal sinus rhythm. After 4 hours, the right hand became numb and significantly colder than the left one. Also, the radial, ulnar and brachial pulses were not detected. CT angiography was requested which showed right brachial artery occlusion without any distal blood flow runoff (figure 1).



Figure 1: Reconstructed CT angiography show right brachial artery occlusion without distal runoff.

Vascular surgery consultation was requested for acute right upper extremity ischemia and heparin (1000 unit per hour) was started. Interestingly, the vascular surgeon's physical exam indicated that right upper extremity was not ischemic and radial and ulnar pulses were three plus. The numbness and paralysis of the forearm were resolved gradually. Chest and neck x-rays were taken and the cervical rib did not appear. CT angiography images were reviewed by the vascular surgeon again and another congenital anomaly was found out. He had a non-aneurysmal, not occluded ARSA; Group 1 "Kiefer et al" classification (7) (figure 2).

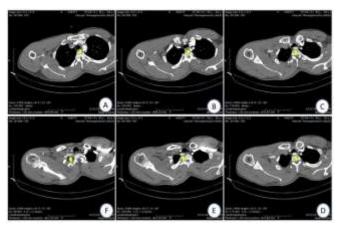


Figure 2: Right upper extremity CT Angiography; Right aberrant subclavian artery (Yellow arrow)

Anticoagulant treatment was continued for the patient and transesophageal echocardiography was done to check the cardiac thrombus or existence of PFO as a source of the probable embolic event. A 4 mm in diameter and 10 mm in length PFO (which bubbles were passed from) was reported. Doppler ultrasound of both lower limbs was performed on suspicion of paradoxical embolism but there was no evidence of deep vein thrombosis. After 48h, the patient had ischemia presentations (coldness and paresthesia) in the right upper extremity again while receiving heparin which was continued for 2 hours and was relieved gradually. After the second ischemic attack, the patient was a candidate for ARSA open surgical revascularization through right supraclavicular approach. A right carotid subclavian artery transposition with oversewing the origin of the aberrant subclavian was done (figure 3).

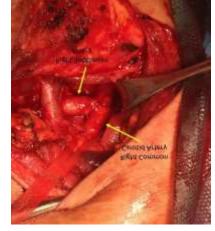


Figure 3: Carotid subclavian transposition was done for aberrant right subclavian artery revascularization through right supraclavicular incision.

He was discharged after 3 days and aspirin (80 mg) was continued for him daily as cardiologist prescription. The patient did not experience any ischemic attack in his right upper extremity for one year of follow up.

This case was approved by the Ethics Committee of Babol University of Medical Sciences with code number: IR.MUBABOL.HRI.REC.1399.118.

Discussion

Approaching to right upper extremity intermittent acute ischemia in a patient with ARSA and PFO is more challenging. At first, it is more important to identify the cause of acute ischemia. If the evidence is in favor of thromboembolic events, it is important to recognize the source of emboli. In such patients, both PFO (8) and aneurysmal degeneration or thrombus formation in proximal segment of ARSA are considered as the known causes of embolization (7). So, the main question in a patient with non-aneurysmal and not occluded ARSA who suffered from acute intermittent ischemia in the right upper extremity is that whether this can be attributed to paradoxical emboli through PFO.

In our report, which is the first report of repeated right upper extremity acute ischemia in the presence of simultaneous ARSA and PFO anomaly, there is no evidence of any deep vein thrombosis in lower extremities and although the size of PFO was smaller to pass a clot which could occlude the brachial artery. So, we emphasize, this is not the probable cause of ischemia.

On the other hand, the passageway of the subclavian artery from the back of the esophagus creates the possibility that when the upper limb is running, the proximal part of the artery will be under the pressure of the adjacent elements (9) and will be followed by stenosis and spasm. Therefore, the ischemic attack will occur and after stopping the activity, the stenosis will resolve gradually and the normal blood flow returns to affected limb.

After revascularization of ARSA by carotid subclavian transposition, the patient did not experience any ischemic attack for one year of follow-up. Thus, the authors suggest in the young or middle-aged adult diagnosed with an intermittent acute upper extremity ischemia, if there is not enough evidence of thromboembolic events do not hurry up to do surgery. Computed tomography angiography and echocardiography can help to identify the other probable pathologies. If the imaging reveals the presence of PFO and

ARSA, paradoxical embolization can be one of the embolic etiologies but in the presence of deep vein thrombosis. In this manner, antiplatelet, anticoagulation therapy or atrial septal device closure are useful for the prevention of a secondary event (10). Otherwise, the correction of the arterial pathway can be helpful in solving the patient's problem.

The authors conclude intermittent acute upper extremity ischemia can occur in the presence of ARSA and PFO. If there is not any evidence of paradoxical emboli and proximal aneurysmal degeneration or occluded ARSA, the treatment can include open surgical revascularization for ARSA and prescription of antiplatelet for PFO.

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Author Contributions

Study conception: PT; Data collection: PT; Investigation: PT, MD; Writing: PT; Funding acquisition: none; Critical review and revision: all authors; Final approval of the article: all authors; Accountability for all aspects of the work: all authors

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