

Incidental Finding of Intercostal Artery Aneurysm During Coarctation Surgery

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Abstract- Intercostal artery aneurysms (ICAA) are very rare vascular complication of coarctation. The most cases are asymptomatic and could be found incidentally during thoracotomy for others vascular pathology or may present with fatal problems such as rupture that is leading to bleeding and hypovolemic shock. Intercostal artery aneurysm most commonly accompanies with neurofibromatosis, aortic coarctation, or in chest trauma. We report a 10-year-old girl who complained of lower extremities pain and hypertension for a few months. Her final diagnosis was coarctation of the aorta. She was scheduled for thoracotomy and coarctation repair by resection and end to end anastomosis. During thoracotomy at lateral side of coarctation, an ICAA distal to coarctation site was seen that by a collateral artery connects to its upper intercostals artery. The patient was treated by ligation and resection of the affected intercostal artery and subsequent coarctation repair. The postoperative course was uneventful and, she was discharged on 7th postoperative day.

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Introduction

The association of intercostals artery aneurysm (ICAA) with coarctation is an exceedingly rare phenomenon, and in our knowledge, only 8 cases reported in the medical literature so far. These ICAA may observe in the proximal or distal of stenotic segment of coarctation. Some authors suppose that this location related to the presence of connective tissue disease in the intercostals artery as was seen in histology of coarctation segment. Although others believe that presence of risk factors such as endocarditis, septic emboli, and turbulence of blood flow may contribute to aneurysm formation (1). In our case absence of history of infection and presence of hypertension led to suppose that cause of ICAA may be hypertension and turbulence of blood flow. The presence of ICAA distal to coarctation segment occur more in the older patient with the association of hypertension or congenital connective tissue abnormality (2). ICAAs are predisposing to serious complication that may be life threatening. ICAA could be rupture with minor trauma such as cough, exercise, or rapid rising of blood pressure. Other life threatening associating anomaly is berry aneurysm of the circle of Willis that occur in up to 10 % of patients with ICAA. In the most of the cases, the ICAA was not seen on the plain chest. Chest X-Ray except to huge

aneurysm that may be observed as a para vertebral mass is usually unremarkable (3-4).

Case Report

A 10-year-old girl presented to our department for elective surgery of descending aorta coarctation. Her past medical history included hypertension and lower extremities pain from 5 months ago. There was not any history of chest trauma, familial history of neurofibromatosis or Kawasaki disease. On the time of admission her lower extremities blood pressure measurement were approximately 90/60, while the upper extremity blood pressures were 160/70. Cardiac auscultation was normal. There was a continuous soufflé in the third intercostal space and in the interscapular space left hemithorax. In skin exam, coetaneous pigmentation or tumor specific of neurofibromatosis were not observed. Her neurologic exam was unremarkable and, her mental status was normal. Echocardiography showed coarctation of the aorta (CoA) in the initial examination (Figure1,2). Her blood hemoglobin level was 13 g/dL, with normal coagulation test. The chest X-ray did not show any rib notching or 3 sign were not detected). Aortography was demonstrating coarctation of the aorta and not only confirmed the diagnosis of coarctation (peak gradient of 80 mmHg) but

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also showed the flow of contrasted blood through the collateral vessels around the coarctation to the descending aorta. "Coarctation causes nearly complete obstruction of blood flow through the descending thoracic aorta. The descending aorta was perfused by collateral vessels from the axillary and internal mammary arteries that connected through the intercostal arteries to descending thoracic aorta, but intercostals aneurysm was not filled by contrast flow. The patient scheduled for thoracotomy and coarctation repair. During thoracotomy, in addition to the presence of short segment of coarctation in descending aorta, a large intercostal aneurysm was detected on the left side of descending aorta. The aneurysm with a broad base was attached to intercostals artery (Figure 3). The coarctated aortic segment reveals an intimal and medial lesion consisting of thickened ridges that protrude posteriorly and laterally into the aortic lumen. The ductus (i.e., patent embryonic remnant) or ligamentum arteriosus (closed and fibrotic) inserts at the same level anteromedially. Intimal proliferation and disruption of elastic tissue were seen at the coarctation level. After resection of the aneurysm and the coarctation, the repair was performed by the end to end anastomosis. The post operative course was uneventful and discharged on the 7th day of operation.

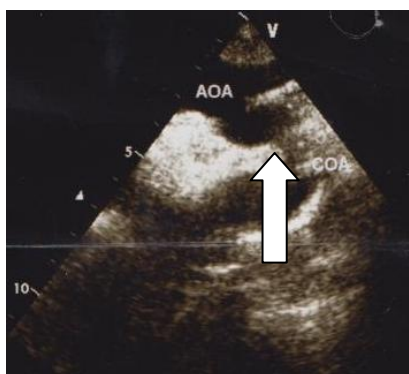


Figure 1. Shows site of coarctation (white arrow)

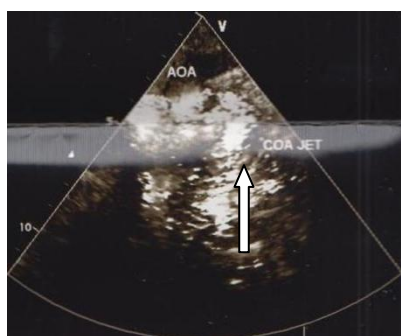


Figure 2. Shows coarctation jet of blood flow across of coarctation

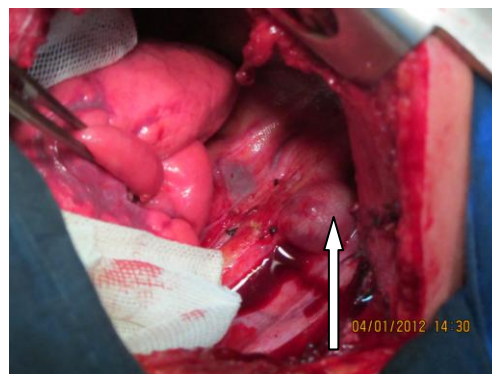


Figure 3. Intercostals artery aneurysm with a broad base was attached to intercostal artery (white curved arrows) and location of coarctation

Discussion

Incidental detection of the intercostals artery aneurysm has very rarely been reported during coarctation repair. The most cases of intercostals artery have been detected during aneurysm rupture. 80% of patients with intercostals artery's aneurysm have neurofibromatosis while 5% have coarctation of aorta and remainder cases related to autoimmune disorder and traumatic causes (5,6,7). The true pathology of aneurysm is still not obviously delineated but is believed to be a wall weakness in the base of connective tissue disease (Ehler-Danlos syndrome), neurofibromatosis or with Kawasaki disease 7. In some case, turbulent blood flow through these collateral arteries may contribute to aneurysm formation in the intercostals arteries. Pseudo aneurysm of the intercostal artery is also observed in association with penetrating or blunt chest trauma. Lu, C.C, and Aizawa (8,9). Report a spontaneous rupture of an intercostal artery in a patient with systemic lupus erythematosus. Pseudo aneurysm of intercostal artery could occur in the open heart surgical procedures (10). Alonso reports a case of intercostals artery aneurysm following midline sternotomy and damage to intercostals artery (11). The association of the ICAA in coarctation patients with no others risk factor is extremely rare, and we found only less than handful cases in the existing medical literature. In the most these cases, patients were diagnosed by sign and symptoms of spontaneous rupture of aneurysm or was detected incidentally by routine chest or C_T scan. In addition, the most of these patients have not coarctation and were elderly and had the risk factors such as, gender female, use of anticoagulation drugs, or having respiratory problem causing severe cough (12-15). Kawasaki disease, as a important cause of acquired heart disease in

children, may be associated with inflammation of medium or large size vessels (16). Some Kawasaki cases may be presented with acute chest pain and impending rupture of intercostal artery aneurysm. In a case report a patient recent history of thoracic surgery and empyema presented with multiple episodes of the arterial hemorrhagic event through chest tubes. In the evaluation, the bleeding cause was found to be an ruptured intercostal artery aneurysm (17). In a case study, the author described a patient with neurofibromatosis that presented with a hemorrhagic pleural effusion. Imaging studies revealed the presence of an aneurysm in the 11th intercostal artery in right hemithorax. With absence, of trauma to the chest wall, author diagnosed a ruptured aneurysm of intercostals artery provoked by coughing in the base of connective tissue in neurofibromatosis. Mycotic ICAA is a rare event sometimes seen in cases with a history of chest wall trauma or chest tube insertion or prior thoracic surgery. The most common presentation of intercostal mycotic aneurysms is brisk bleeding in the presence of a chest tube or bloody pleural effusion or hemorrhagic shock (17,18). The histological evaluation of resected aneurysm revealed the normal amount of elastin and collagen content in the intercostals artery wall that excludes connective tissue disease; tissue culture results were also negative for conventional Fungal, and bacteria that rule out mycotic aneurysm. A serum antitrypsin enzyme level was also normal. The absence of neurofibroma in the histological exam of intercostal nerve excluded neurofibroma related ICAA. Systemic lupus erythematosus was excluded from negative results for dsDNA and antinuclear antibody. Rheumatoid factor was also negative that rule out rheumatoid arthritis. The erythrocyte sedimentation rate that is raised in Kawasaki disease was normal. No any evidence of cystic medial necrosis or medial degeneration was found in the histological exam.

ICAA is a rare association with coarctation of the aorta, and careful literature search revealed less than handful cases of intercostals artery aneurysm in children. most of the cases have related to neurofibromatosis, coarctation of the aorta, and Kawasaki's disease and in rare cases this combination was found in systemic lupus erythematosus, septic emboli to intercostals artery, mycotic aneurysm, traumatic damage to artery and Ehlers-Danlos syndrome, but our case had neither of these association except to coarctation based on aforementioned testing. Unique feature of our case related to the site of intercostals artery aneurysm in relation to coarctation of

the aorta, the aneurysms usually origins in the upper intercostal arteries as in previously published case reports but in our case aneurysm occurred in lower intercostals artery regard to coarctation.

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