Rupture of Inferior Phrenic Artery Aneurysm*

An Unusual Complication of Mesenteric Arteritis Due to Postcoarctectomy Syndrome

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Postcoarctectomy syndrome was developed in a boy with Williams syndrome. Angiogram revealed widespread changes of visceral arteries and extravasation of the contrast material from the right inferior phrenic artery. Emergency laparotomy was successful. This unusual complication makes clear that the mesenteric arteritis after coarctectomy can cause aneurysm with a risk of rupture.

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Postcoarctectomy syndrome presents as severe abdominal pain, beginning 24 h to 8 days after coarctectomy, and can include mesenteric arteritis. Williams syndrome is characterized by hypercalcemia, a congenital heart defect, mental retardation, and elfin face. We report an unusual complication of rupture of the right inferior phrenic artery aneurysm due to mesenteric arteritis following coarctectomy in a boy with Williams syndrome.

CASE REPORT

A 10-year-old boy was admitted to the hospital with edema and proteinuria on March 9, 1989. The patient was diagnosed as having primary osteoma cutis and pseudohypoparathyroidism at 7 months of age.

At the time of hospital admission, the boy weighed 25.4 kg and was 121.7 cm tall. He had elfin face and was mentally retarded. On physical examination, a continuous murmur (Levine second degree) was heard in the fourth intercostal space at the left sternal border, and a diastolic murmur (Levine third degree) was heard at the apex. Blood pressure in the upper extremities was 160/100 mm Hg, but pulses were barely palpable in the lower extremities. Subcutaneous calcified nodules were noted on the thighs, hips, and abdomen. Laboratory data were normal except for hematocrit, 167 mg/100 ml (2.5 g/dl), and serum albumin, 2.0 g/dl. Ellesworth-Howard test showed type 2 abnormality with an increase in cyclic adenosine monophosphate levels. Cardiac catheterization revealed coarctation of the aorta with a pressure gradient of 80 mm Hg, and mild aortic regurgitation.

Coarctectomy with end-to-end anastomosis was performed on July 13, 1989. Immediately after surgery, arterial pulsation became normal in the lower extremities. However, hypertension continued (150 to 170/90 to 100 mm Hg), and the patient was given nifedipine, hydralazine, and other antihypertensive drugs. β-Blocker was not administered due to bradycardia. By these medications, blood pressure was gradually normalized on the third postoperative day; however, the patient complained of nausea and a stomach ache and vomited. On the fourth postoperative day, abdominal distention became obvious. The next day the platelet count fell to 10,000/mm³, and the patient developed melena and went into shock. Gastroscopy revealed acute gastric mucosal lesions, and abdominal paracentesis revealed hemoperitoneum. Angiogram of the abdominal aorta and its branches revealed widespread club-like dilatation, segmental spastic constriction, and stretching. The right inferior phrenic artery was dilated, and extravasation of the contrast material was noted (Fig 1).

We performed an emergency laparotomy. Blood was spurting from the area of the right diaphragm, and hemostasis was achieved by suture ligation of the inferior phrenic artery. Stomach and intestines were edematous and filled with blood; numerous petechiae were observed on the serosa of intestines and the mesentery. Total amount of hemorrhage was estimated to be about 850 ml. The patient recovered rapidly after laparotomy.

Three weeks after surgery, the patient’s systolic arterial pressure was 100 to 120 mm Hg, and the diastolic murmur had disappeared. Two months after surgery, angiography revealed resolution of pathologic changes, except for two small aneurysms in the jejunal and ileal arteries (Fig 2). The patient experienced no further cardiovascular complications or abdominal symptoms.

DISCUSSION

The mechanisms of paradoxical hypertension following repair of coarctation is thought to be due to hormonal (increased catecholamine release and activation of renin-angiotensin system) or neural disorders. It disappears for a few days with or without treatment in most of the patients, thus patients who require laparotomy are extremely rare. Pathologic findings of postcoarctectomy mesenteric arteritis include intestinal hemorrhage, and arterial and venous thrombosis. The pathogenesis of mesenteric arteritis following coarctectomy is thought to be similar to that of malignant hypertension. Nemes et al have suggested that progressive elevation of blood pressure induces vasoconstriction by autoregulation and release of vasopressor agents that resulted in necrosis of smooth
Peripheral Intrapulmonary Hamartoma Accompanied by a Similar Endotracheal Lesion*  

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We describe a case of peripheral intrapulmonary hamartoma accompanied by a similar endotracheal lesion. A 70-year-old asymptomatic man had a polyloid lesion in the trachea and a nodular shadow in a peripheral lung area. A biopsy specimen from the trachea revealed a chondroid hamartoma, and the surgically resected tissue of the intrapulmonary tumor showed the same histologic findings. This is the second case of multiple hamartomas showing coexistence of tracheobronchial and parenchymal tumors. (Chest 1994; 106:1291-93)

Hamartomas are the most common benign pulmonary neoplasms, and approximately 90 percent of them are located within parenchyma.1 Tracheal hamartomas are extremely rare, and only nine cases had been found in the available literature. Rarely, hamartomas are multiple, and most of these are the fibroleiomyomatous type occurring in female subjects. We report a rare case of a man with a pulmonary chondroid hamartoma accompanied by a similar endotracheal lesion. We discuss the relationship between parenchymal and endotracheal tumors and further speculate on the pathogenesis of hamartoma.

CASE REPORT

A 70-year-old Japanese asymptomatic man was referred to our hospital after a routine chest radiograph showed a well-demarcated nodule behind the heart (Fig 1). The patient had an 18 pack-year smoking history but a medical history that disclosed no abnormalities. Vital signs and laboratory data were essentially normal. The patient’s previous roentgenograms showed, in retrospect, the lesion to have been present for 6 years and to have slightly increased in size. Computed tomography showed a 4X3.5-cm mass of heterogenous density with speckled calcification in the left lower lobe without obvious involvement of adjacent structures.

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Figure 2. Superior mesenteric arteriogram performed 2 months after surgery revealed resolution of pathologic changes, except for two small aneurysms in the jejunal and ileal arteries.