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Case Report

Choreoathetosis in a patient after pulmonary artery endarterectomy: A case report

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Chorea after pulmonary artery endartectomy is a rare entity. It has been described as a complication in children who has undergone hypothermic circulatory arrest. We present a case of a 41 year old lady who developed choreoatheosis after pulmonary artery endarterectomy.

Keywords: Choreoatheosis, pulmonary artery endarterectomy

INTRODUCTION

History

A 41 year old female with protein C and S deficiency, hypertension and asthma was diagnosed with pulmonary embolism in April of 2010. Despite medical compliance and therapeutic anticoagulation she was subsequently admitted to our medical center with worsening dyspnea. An echocardiogram revealed pulmonary hypertension with right ventricular systolic pressure of 65 mmHg and severely dilated right ventricle and right atrium. Of note, there was no history of known prior pulmonary hypertension, obstructive sleep apnea, use of weight loss medications or HIV. Past surgical history was positive for cesarean section x4, cholecystectomy and lumpectomy of her right breast. She had smoked 0.5 packs a day for over 10 years. She did not have any history of alcohol or illicit drug use.

Subsequent ventilation perfusion scan revealed multiple mismatched perfusion defects.

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A right and left cardiac catheterization revealed a mean pulmonary artery pressure of 42 (67/24), Left ventricular end-diastolic pressure 10mmHg, cardiac output 3.38 L/min, cardiac index 1.87 L/min/m , PVR 9.469 woods units (WU) and right atrium pressure (RAP) of 23 mmHg. There was no oxygen step up suggestive of a shunt and no response to inhaled nitric oxide challenge. There were no significant lesions in the coronary arteries. A pulmonary angiogram confirmed surgically amenable chronic pulmonary embolism. A pulmonary artery endarterectomy was recommended.

Operative details

Following a median sternotomy the patient was placed on cardiopulmonary bypass. Vents were placed in both the main pulmonary artery and the right superior pulmonary vein. While cooling, the head was packed in ice and Phenobarbital (1 gram) and Solumedrol (1 gram) were administered. The aorta was clamped and the heart was arrested with cold blood cardioplegia, which was re-administered every 20 minutes. The superior vena cava, aorta and main pulmonary arteries were extensively

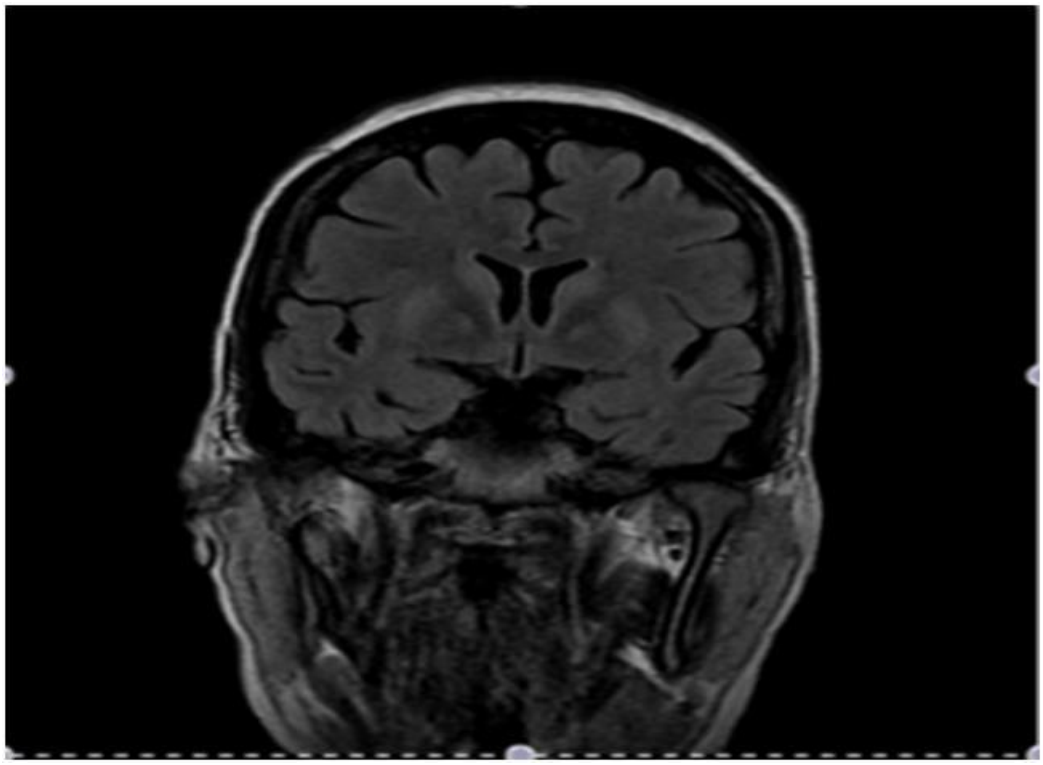

Figure 1. MRI of brain showing abnormal signal in putamina, caudate heads and globipallidi, consistent with hypoxic ischemic injury.

mobilized. After cooling for 57 minutes circulation was arrested with a bladder temperature of 18° C and an isoelectric electroencephalogram. Separate longitudinal arteriotomies were made in the left and right pulmonary arteries and using the techniques described by Jaimeson et al. and extensive endarterectomy specimen was retrieved from each side. Each side was done with a separate period of circulatory arrest (right-34 minutes and left-27 minutes) interrupted by a 15 minute period of reperfusion at 18°C. The patient was re warmed in a standard fashion to 37° C prior to separation from cardiopulmonary bypass. Total rewarming time was 76 minutes and total cardiopulmonary bypass time was 212 minutes.

Post operative course

Following extubation the patient displayed choreoathetotic movements in her extremities, eyes, mouth and tongue. She was able to follow simple commands but speech was slow and dysarthric. Her choreoathetotic movements increased with verbal questioning and tactile stimuli.

A CT scan of head revealed low attenuation and loss of grey/white differentiation within basal ganglia regions. MRI of brain showed an abnormal signal in putamina, caudate heads and globipallidi, consistent with hypoxic ischemic injury. (Figure 1)

She was started on propanolol and risperidone and later switched to haldol and amantadine in the neuro rehab. Patient was discharged home after 40 days with significant improvement but not completes resolution. This slowly improved as an outpatient over a period of 3 months resulting in complete recovery.

DISCUSSION

Neurological complications are not common after pulmonary artery endarterectomy (Fedullo et al., 2001). However, deep hypothermia with circulatory arrest, has been shown to cause stroke and cognitive impairment (Jamieson et al., 1993). Chorea on the other hand is mostly been described as a complication in children who have undergone hypothermic circulatory arrest (Medlock et al., 1993). Post pump chorea occurs in 1.2 % of children and is thought to be due to several factors including prolonged cardiopulmonary bypass times, prolonged circulatory arrest periods and cooling to lower temperatures (Medlock et al., 1993). Others have postulated a role for hypocapnia-induced cerebral vasoconstriction (Curless et al., 1994).

We could find only one clinical observation report which

described chorea in a patient who underwent pulmonary artery endarterectomy (Rob et al., 2008). They identified, younger age, longer period of circulatory time and quicker rewarming time as risk factors for developing chorea. MRI in one patient revealed hyperintensity at the globuspallidus. In all cases the chorea was transient.

Reviewing our surgery we found that her circulatory arrest time was slightly prolonged at 34 minutes and 27 minutes compared to the San Diego protocol of 20 mins circulatory arrest period for each side. Our patient was cooled to 18 degree celcius as compared to the San Diego protocol of 20 degree.

While choreoathetosis appears to commonly resolve, it does contribute to hospital stay and morbidity. We feel that reducing circulatory arrest time and avoiding deep hypothermia may be helpful.

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