Journal of Surgical Techniques and Procedures

Case Report
Published: 08 Jan, 2020

Ruptured Aneurysm at a Rare Location: Azygos Anterior Cerebral Artery

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Abstract

Anatomic variations of the circle of Willis may cause cerebral aneurysm formation. Azygos Anterior Cerebral Artery (aACA) is the existence of A2 segments of the anterior cerebral artery as a single vessel. In the presence of aACA, aneurysm development is more common than normal anatomic structure. Azygos Anterior Cerebral Artery (aACA) aneurysms often develop at the level of the corpus callosum genu and bifurcation. We present a patient who was admitted to the emergency department for intracerebral hematoma, and was operated due to saccular aneurysm developed at the bifurcation of callosomarginal artery, the first branch of aACA.

Keywords: Aneurysm; Azygos anterior cerebral artery; GCS; Corpus callosum genu

Introduction

Azygos Anterior Cerebral Artery (aACA) is a rare variant of A2 segments of the Anterior Cerebral Artery (ACA). In angiography and autopsy series, it was reported to be 0.3% to 2% [1]. In this variation, there is only one A2 segment. Typically, this artery is divided into two branches at the level of the corpus callosum genu and continues as callosomarginal and pericallosal arteries. Azygos Anterior Cerebral Artery (aACA) feeds the medial side of the frontal and parietal lobes. Studies have shown that the incidence of aneurysms in aACA is higher than in normal A2. This has been reported to be caused by increased blood flow velocity and hemodynamic stress in aACA bifurcation [2]. We presented a patient with saccular aACA who presented to the emergency department with subarachnoid hemorrhage and we also evaluated the literature on this topic. Between March 2015 and October 2019, only one out of 136 patients operated for aneurysm in our clinic had aACA aneurysm.

Case Presentation

A 44 years old male patient who had headache for 1 week was brought to emergency service due to loss of consciousness. In his neurological examination in the emergency department, his Glasgow Coma Scale (GCS) score was 14 and there was no motor deficit in his extremities. The patient's brain Computed Tomography (CT) revealed a right frontal intracerebral hematoma (Figure 1). Brain CT angiography was performed and a 5.3 mm long saccular aneurysm was seen on the aACA at the level of the corpus callosum genu where the callosomarginal artery branch was exited (Figure 2). Because his GCS score was lowered to 12, he was operated on immediately. Bifrontal craniotomy was performed. The interhemispheric fissure was passing and intracerebral hematoma was evacuated in a controlled manner. Callosomarginal artery was seen and dissected proximally and just below the aneurysm. The aACA was exposed for proximal control. But there was no need to place temporary clips. The aneurysm was closed with a single clip on the neck. Azygos Anterior Cerebral Artery (aACA) and callosomarginal artery flow were checked by Doppler. The patient stayed in the intensive care unit for one day postoperatively. He had normal neurological examination and control DSA angiography was performed. The aneurysm was completely closed (Figure 3). The patient had mild psychiatric symptoms and medical treatment was started. He returned to work two months later postoperatively.

Discussion

Anterior circulating aneurysms are most commonly seen on the anterior communicating artery and middle cerebral artery bifurcation [3]. The incidence of distal ACA aneurysms has been reported to be between 2% and 6.7% of all intracranial aneurysms. Azygos Anterior Cerebral Artery (aACA) is defined as the fusion of A2 arteries and the presence of aACA increases the incidence of distal ACA aneurysms [4]. At the same time, occlusion in this single vessel causes cerebral ischemia in areas fed

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Received Date: *19 Nov 2019*
Accepted Date: *02 Jan 2020*
Published Date: *08 Jan 2020*

Citation:
Eren B, Karagoz Guzey F, Gulec I, Serdar Bas N. Ruptured Aneurysm at a Rare Location: Azygos Anterior Cerebral Artery. J Surg Tech Proced. 2020; 4(1): 1030.

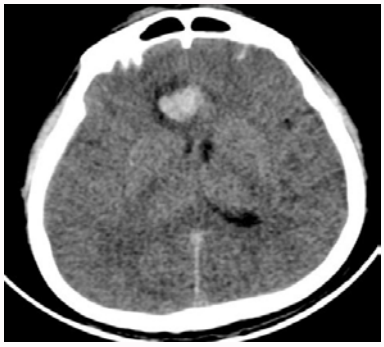

Figure 1: Axial brain CT shows right frontal intracerebral hematoma.

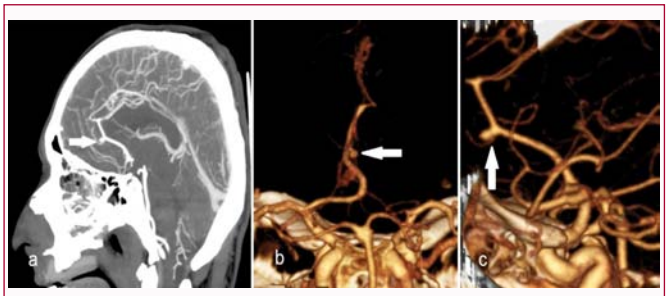

Figure 2: CT angiography of the brain a) sagittal, b) 3D reconstruction posterior view, c) 3D reconstruction lateral view of azygos ACA and saccular aneurysm (white arrows) on it.

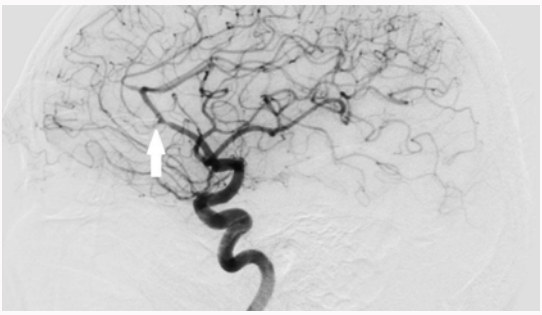

Figure 3: Postoperative cerebral DSA image showed a total closure of the aneurysm (white arrow) with one clip.

by the distal ACA, resulting in neurological and neuropsychological sequelae. The natural course of aneurysm formation begins with the effect of endothelial cells [5]. The slowing/acceleration/turbulence of the blood flow causes damage to endothelial cells covering the vascular wall. It is characterized by inflammatory changes in the aneurysm wall, loss of internal elastic membrane, thinning and hyperplasia in tunica media, proliferation of smooth muscle cells, degeneration of extracellular matrix and development of fibrosis. The process starts with endothelial damage and continues with phenotype changes in smooth muscle cells. In humans with aACA, the blood flow velocity in this vessel was measured faster than normal A2. In locations where the vessel is divided into branches, this increased flow causes stress in the vessel wall and may cause aneurysm formation [6]. In a study of experimental bifurcation in cerebral arteries, it was found that two types of modeling were performed in histopathological examination of the vascular structure [7]. First, blood flow impinging on the cerebral vascular bifurcation wall caused intima hyperplasia, and in the vascular wall where the blood flow accelerated, internal elastic membrane deterioration, loss of smooth muscle cell and loss of fibronectin tissue were detected. In cerebral artery bifurcations, the type of flow, angle of bifurcation and diameters of the main vessel and its branches vary [8]. On bifurcation aneurysms, blood flow from the afferent artery is divided into two or more currents. In patients with aACA, aneurysm is usually seen in the bifurcation of the branches of the pericallosal artery and callosomarginal artery. Bifurcation, trifurcation and even quadrifurcation can be seen here [9]. However, our patient had only one early callosomarginal artery and the aneurysm was located there. In addition to flow velocity and hemodynamic stress theories, congenital anomalies of aACA vessel structure have been reported. Kasperra et al. [9] reported in a study performed with transcranial color-coded sonography in patients with aACA aneurysm that, the cause of aneurysm formation was not attributed to increased vascular flow velocity, but to hemodynamic stress caused by bending of this artery around the corpus callosum genu. In the patient we presented, aACA was upward at a sharp angle at the level of the corpus callosum genu according to the opinion of Kasperra et al. [9].

Conclusion

Azygos anterior cerebral artery is a rare anatomical variation. Nevertheless, aneurysm is seen at a high rate on this vessel. Angiography of the patients should be carefully examined before surgery. Because vascular occlusion that may occur during operation in these patients it will cause infarction in the brain regions fed by the distal anterior cerebral artery.

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