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The First Case Report of Spontaneous Idiopathic Bilateral Vertebral Arteriovenous Fistula

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Abstract

We present a 43-year-old male patient who has bilateral spontaneous idiopathic Vertebral Arterio Venous Fistula (VAVF) which is rarely seen. The importance of our case is that the third case of bilateral VAVF which was reported in the literature and the first case of spontaneously developed with uncertain etiology. Due to risk factors, bilateral and sub axial cases are difficult to treat and their surgical complications can be life-threatening and sometimes unsolvable.

Keywords: Bilateral; Idiopathic; Spontaneous; Vertebral arteriovenous fistula

Introduction

Arteriovenous fistulas were first described by Hunter in 1762 [1]. Arteriovenous fistulas are defined as abnormal anastomoses between an artery or its branches and the adjacent venous system [1,2]. While Vertebral Arterio Venous Fistulas (VAVF) is rarely seen diagnosis is often confirmed by catheter cerebral angiogram. Most commonly, they occur due to hyperextension of the neck, penetrating and iatrogenic injuries (during vascular catheterization and spinal surgery), and may occur congenitally or spontaneously [1-4]. Diseases such as neurofibromatosis and fibro muscular dysplasia may also cause congenital VAVFs [3,5]. Congenital VAVFs result from the continuity of embryonic connections between arteries and veins in the head and neck region [6,7]. Congenital VAVFs are extremely rare pathologies [2-4,6-12]. A spontaneous VAVF case was first reported by Norman in 1950 [13,14]. In some patients, VAVFs are asymptomatic, while others suffer from neurological symptoms such as tinnitus, weakness, pain, and vertigo [15-17]. Treatment aims are complete occlusion of the fistulas, which can be achieved by both endovascular and/or surgical methods. In this study, we present a case of spontaneous and idiopathic bilateral VAVF.

Case Study

A 43-year-old male patient was admitted to the neurosurgery clinic with headache complaints. After the cranial Computed Tomographic Angiography (CTA) and diagnostic catheter angiography, we detected a right-middle cerebral artery bifurcation aneurysm and bilaterally dilated VAVF (between the left and right vertebral arteries and both deep cervical veins) in the paravertebral region. We found that these dilated vascular fistulas located bilaterally at C1, C2, and C3 levels in the cervical region of the vertebral column (Figure 1A and 1B). The patient was discharged after the aneurysm clipping and was asked to return for a follow-up visit a month later for endovascular VAVF intervention. However, when the patient showed up for a follow-up visits, he complained of dizziness, tinnitus and fatigue. The patient was found to have had no history of neck and head trauma, and no Cafe Au Lait spots related to neurofibromatosis. When the patient was examined for cardiac insufficiency due to a possible high-flow VAVF, we found no clinic or radiological pathology was observed. The cardiothoracic index was smaller than 0.5. Endovascular fistula embolization was again proposed as a treatment for the patient, and the risk factors of the intervention were explained in detail. However, because the patient did not experience difficulties with his day-to-day activities, and due to the risk factors that might occur as a result of the intervention, he did not accept the treatment and was discharged.

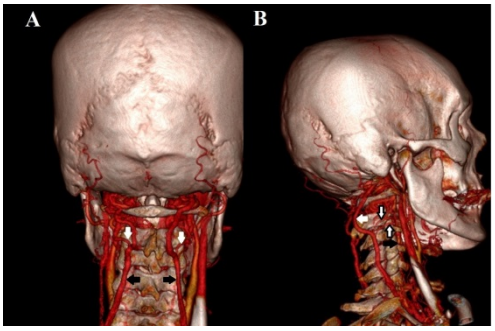

Figure 1: 3D images showing bilateral VAVF appearance (white arrows) between vertebral arteries and the deep cervical veins (black arrows) at C1-C3 level in the paravertebral region (A). In oblique view of the image (B) demonstrating vertebral artery (black arrow), deep cervical vein (white arrow) and bilateral VAVFs (white and black coloured arrows).

Results and Discussion

Arteriovenous fistulas of the vertebral artery are rarely seen and usually develop after trauma [4-6,11-18]. Most of the traumatic vertebral fistulas are found in the intraforaminal region while spontaneous vertebral fistulas are located in the region where the artery moves away from the foramen of the atlas and enters the foramen magnum [19]. Fistula surgery in the intraforaminal region of the artery can be challenging. Abundant hemorrhages may develop in the Batson plexus in this region during surgery. This can damage vital anatomical structures such as the phrenic nerve, the brachial plexus, or the cervical spinal cord. Even though the etiology of the congenital arteriovenous fistulas is not fully known, it is reported to be associated with its anatomic localization [3,4,20]. Whereas pathologies leading to arterial wall diseases such as arteriosclerosis and neurofibromatosis lead to congenital or spontaneous VAVFs in the neck region due to the delicate wall structure [21,22]. Because of the vascular structure and low blood pressure of the venous plexus surrounding the vertebral artery in this region, the arterial pressure in the vertebral artery or its branches can easily drain into the venous system. Spontaneous VAVF

is much less common and its etiology is uncertain. Some are thought to be congenital [23,24]. Congenital or spontaneous VAVF is usually found in the C1-C2 region and in the upper segments of the vertebral artery [25], while traumatic VAVFs are usually located in the C5-C6 region [26].

In a review they prepared, Vinchon et al. reported a total of 224 VAVFs, 72 of which were congenital and 152 were traumatic. They stated that congenital VAVFs were associated with Fibro Muscular Dysplasia (FMD), neurofibromatosis or the Ehler-Danlos disease, and most commonly seen in the elderly, while traumatic VAVFs were more often seen in young people. Bilateral cases are very rare, as indicated in the largest series in the literature. In this series of 49 cases studied by Vinchon et al. only one bilateral VAVF with FMD in its etiology was reported. In the second case presented by Hiroshi et al. a concomitance of neurofibromatosis and atlantoaxial dislocation in the etiology was reported.

Endovascular embolization and/or occlusion techniques are most commonly used for the treatment of VAVFs [27-29]. Especially if surgical stabilization of the subaxial (C1, C2) traumatic unstable vertebral lesions is planned using screws, detection of the preoperative vertebral artery course and its neighboring anatomical structures by using CTA and/or digital subtraction angiography is important in terms of avoiding life-threatening complications.

Conclusion

The importance of our case lies in the fact that it is the third reported case of bilateral VAVF in the literature and the first spontaneously developing case with uncertain etiology. Due to these factors, bilateral and sub axial cases are difficult to treat and their surgical complications can be life-threatening and sometimes unsolvable. In particular, patients with a murmur in the neck region and a history of trauma should be examined for these pathologies.

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