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CASE REPORT: SUBARACHNOID HEMORRHAGE DUE TO RUPTURED ANEURYSM IN A KNOWN CASE OF MOYAMOYA DISEASE

Clinical Research

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ABSTRACT

Moyamoya disease is a rare neurological condition of the cerebral vasculature that occurs mostly in Asians. It is characterized by steno-occlusion of cerebral arteries, leading to reduced blood flow. In severe cases, it may cause hemorrhage. This report describes a known case of moyamoya disease in a 24-year-old female who presented with a history of severe headaches and vomiting. On evaluation, MRI, DSA, and bilateral vertebral arterial injections suggested a ruptured left P1-PCA aneurysm with SAH associated with Moyamoya disease. She underwent flow diverter placement and endovascular coiling for a ruptured aneurysm.

KEYWORDS

moyamoya disease, endovascular coil embolization, flow diverter, ruptured aneurysm, hemorrhage.

INTRODUCTION

Moyamoya disease is a rare, progressive condition characterized by the narrowing or occlusion of the cerebral arteries, with an unknown cause [1]. It is most prevalent among Asians, particularly in Japan, and has a higher incidence in females compared to males, with a male-to-female ratio of approximately 1:1.8. The disease is commonly diagnosed in early childhood, typically within the first decade of life[2]. While it usually affects both sides of the brain, there have been instances where it is confined to one side [3].

The term "Moyamoya," which means "puff of smoke" in Japanese, refers to the appearance of the collateral vessels seen on angiography that form to compensate for the arterial blockage [4].

The disease primarily affects the median or anterior cerebral arteries, creating an abnormal web-like or fibrous structure when visualized on imaging [5]. Both children and adults can be affected, experiencing symptoms such as headaches, seizures, ischemic strokes, intracranial hemorrhage, or transient ischemic attacks [6].

Some research suggests that circulating angiogenic factors like growth factors, vascular progenitor cells, and cytokines may play a role in neovascularization, contributing to intimal hyperplasia and the formation of abnormal collateral vessels [7].

Although there is no specific cure for Moyamoya disease, various surgical options, such as indirect and direct bypass procedures, aim to improve cerebral blood flow and address ischemic complications [8]. Surgical revascularization is often used to prevent ischemic strokes, but its role in preventing future hemorrhagic strokes remains debated [9].

Hemorrhages associated with Moyamoya disease can be classified into intraparenchymal hemorrhage (IPH), intraventricular hemorrhage (IVH), and subarachnoid hemorrhage (SAH). The hemorrhages seen in Moyamoya are typically smaller in volume and more frequently involve the ventricles compared to primary intracerebral hemorrhages [10]. A prior study found that the severity of IVH, measured by the Graeb scale, is an independent risk factor for poor short-term outcomes in patients with hemorrhagic Moyamoya disease [11]. However, research on prognostic factors related to hemorrhagic Moyamoya disease is limited, and this study aimed to explore disease- and patient-specific factors influencing outcomes in these cases. In complex situations involving ruptured aneurysms, advanced endovascular techniques, such as a combination of flow diverter placement and coil embolization, are used to prevent re-bleeding.

Case Description

A 24-year-old female presented to the hospital with a history of severe headaches associated with two episodes of vomiting and a fall in the bathroom due to syncopal episodes. On admission, her vitals were stable, and the Glasgow Coma Scale (GCS) was 15. She has a history of hypothyroidism and is on Thyronorm 50 mg.

Lab investigations revealed normal hemoglobin, platelet count, sodium, and potassium levels. MRI and Digital Subtraction Angiography (DSA) suggested a bilateral aneurysm, a ruptured left P1-PCA aneurysm, and an unruptured right P1-PCA aneurysm with Subarachnoid Hemorrhage (SAH). Bilateral proximal anterior cerebral artery (ACA), middle cerebral artery (MCA) terminal, Internal cerebral artery (ICA) moyamoya disease. Bilateral vertebral arterial injections showed a ruptured left P1-PCA saccular aneurysm measuring 2.5×3.4mm with a neck measuring 1mm and a right PCA saccular aneurysm measuring 2.5×2.5mm. Patient attendees were counseled for further management.

The flow diverter placement and coil embolization were performed using a 3mm×6cm coil (target 360 ultra) of the ruptured left P1-PCA under general anesthesia after getting the patient's consent. The post-coiling angiogram showed no residual filling of the sac. There were no periprocedural complications. Subsequent monitoring in the intensive care unit (ICU), Postoperative day 1(POD-1) CT showed no new intracranial hemorrhage. The patient remained hemodynamically stable and was discharged.

DISCUSSION

In this study the patient was diagnosed with a ruptured aneurysm located in the left P1 segment of the posterior cerebral artery (PCA), alongside involvement of the proximal anterior cerebral artery (ACA), middle cerebral artery (MCA), and terminal internal carotid artery (ICA), occurring in the context of moyamoya disease (MMD). Since the rupture was immediate, emergency intervention was necessary to avert possible life-threatening hemorrhage because coil embolization was selected as the primary treatment; it is minimally invasive and will occlude an aneurysm and help prevent further rupture. This was relevant particularly because of the patient's background MMD, which would increase the risk for vascular dysplasia and formation and subsequent rupture of aneurysm. Thus, the endovascular coil embolization of the ruptured aneurysm was satisfactorily completed, thus preventing the imminent threat of hemorrhage and other serious neurological complications [12.13.14]. However, the treatment of MMD is challenging and sometimes a global approach has to be adopted in dealing with the vascular pathology as well as the complications accompanying it.

CONCLUSION

Continuous long-term follow-up becomes essential in monitoring disease progression, assessing the effectiveness of the employed treatment, and thus helping prevent recurrence or further development of new aneurysms. This method has two important challenges: first, it is located near vital neurovascular structures at their anatomical sites, and surgical access is extremely difficult, if not impossible. Second, proximal control with temporary clips is usually unfeasible, based on the damaged pre-existing anterior circulation system and the fact that the vertebrobasilar system becomes the only blood supply source. Third, high internal pressure in the vertebrobasilar system makes

clipping these aneurysms risky [15.16]. This indicates that surgically treating posterior circulation aneurysms is a seriously compromising situation, thus strongly underlining the usefulness of coil embolization in posterior cerebral artery aneurysm treatment particularly when additional surgeries could eventually become a risk factor. In such a scenario, intra-aneurysmal embolization is an effective treatment.

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