

Case Report

Nontraumatic, posterior circulation pseudoaneurysm of the basilar artery summit with complete spontaneous resolution: Case report and literature review

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Abstract

Background: Intracranial pseudoaneurysms are rare vascular defects of arterial walls that are classically the result of traumatic injury, iatrogenic causes, or infection. Idiopathic pseudoaneurysms are seen even less frequently and are often related to atherosclerosis. Pseudoaneurysms are most commonly found along the distal wall of the internal carotid artery, however, can occur at any location in the cerebrovascular circulation. Treatment of these arterial defects is often challenging due to their frail nature.

Case Description: A 61-year-old male with a history of hypertension presented with a severe, atypical headache without history of trauma. Computed tomography (CT) and computed tomography angiography (CTA) demonstrated diffuse subarachnoid hemorrhage. Imaging demonstrated a 3.5 mm pseudoaneurysm projecting distally from the basilar artery at the apex. Repeated imaging (CTA, digital subtraction angiography) demonstrated decreased size and flow associated within the aneurysm over the following 2 weeks; as such, the patient was managed conservatively. The patient was discharged in neurologically intact condition when imaging at 14 days confirmed complete and spontaneous resolution of the pseudoaneurysm.

Conclusion: Idiopathic pseudoaneurysms that are commonly associated with atherosclerosis are most commonly managed surgically or endovascularly. Conservative approach may be considered in a select group of patients that exhibit decreased size and/or flow within the aneurysm in repeated imaging; spontaneous resolution was seen in the present case.

Key Words: Basilar artery, conservative management, pseudoaneurysm, subarachnoid hemorrhage

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INTRODUCTION

Intracranial pseudoaneurysms are potentially dangerous arterial lesions classically caused by trauma,^[10,28] iatrogenic injury,^[10,17,21,29,38] or bacterial infection.^[10] In contrast, idiopathic intracranial pseudoaneurysms—those with an unknown cause or source—are relatively rare. However, there have been several case reports of idiopathic pseudoaneurysms, often presenting with subarachnoid hemorrhage (SAH).^[8,9,11,21]

Unlike true aneurysms, pseudoaneurysms are not typically found on the bifurcation of vessels, but rather along a vessel wall, distal from a branch point.^[37] They differ pathologically from true or dissecting aneurysms in two manners. In pseudoaneurysms, all three layers of the vessel wall have been compromised, and focal thrombosis can be found external to the vessel, as opposed to a true aneurysm, where no layers have been compromised and the vessel dilates from within.^[1] These lesions often present with SAH. Postsurgical pathologic examination, when performed, has demonstrated that pseudoaneurysms are composed of blood clot and fibrous tissue.^[8] Considerable atherosclerosis is often present both on the parent vessel adjacent to the lesion and within the circle of Willis when pseudoaneurysms are present.^[5,8]

Pseudoaneurysms can occur on vessels in posterior circulation, as is our case, however, they most typically occur in anterior circulation on the internal carotid artery (ICA), and are also known as blood blister-like aneurysms. Nomenclature in the literature uses both terms interchangeably. Treatment is often challenging because of the frail nature of these false aneurysms, which have a tendency to rupture during treatment.^[1] Several methods of treatment have been proposed and demonstrated success, however, no one method has consistently shown superiority.^[4,5,10,11,13]

CASE REPORT

History and examination

A 61-year-old male with a history of hypertension presented to the emergency department with a severe, atypical headache without a history of trauma. The patient was neurologically intact, Hunt and Hess grade II, with an initial blood pressure of 172/96 mmHg.

Imaging

Computed tomography (CT) and subsequent computed tomography angiography (CTA) demonstrated diffuse SAH, contained largely within the basilar cisterns, but without clear source of the hemorrhage on CTA. Subsequent digital subtraction angiography (DSA) and digital rotational angiography (DRA) demonstrated a 3.5 mm idiopathic pseudoaneurysm projecting posteriorly

from the basilar summit/posterior cerebral artery junction without evidence of arterial dissection [Figure 1a].

Intensive care unit course and management

Subsequent imaging both with CTA and DSA/DRA demonstrated decreased size and flow associated within the aneurysm. Expectant management of the aneurysm was undertaken and no subsequent treatment was administered. The patient had an expected course in the intensive care unit (ICU), including temporary ventricular drainage, and was discharged home in neurologically intact in stable condition. Follow-up DSA and DRA 14 days after hemorrhage demonstrated complete resolution of the pseudoaneurysm [Figure 1b]. Subsequent follow-up, including imaging at 1 year, confirms the spontaneous and complete resolution.

DISCUSSION

“False aneurysms” have been described in the literature as early as 1928 due to trauma and iatrogenic causes. In a case series by Besser *et al.*, approximately 5% of the vertebrobasilar aneurysms were described as “false aneurysms.”^[4] At present, “pseudoaneurysm,” “blood blister aneurysm,” and “dissecting aneurysm” are the terms used interchangeably in the literature to describe aneurysms distant from branching points and share similar presentation and radiological and pathological findings. Pseudoaneurysms are histologically characterized by a layer of adventitia with or without encasing clot formation, therefore, lack all three layers of the true aneurysmal sac.^[5]

Idiopathic (nontraumatic and noninfectious) pseudoaneurysms, one of which is presented in the current case report, are relatively rare compared with pseudoaneurysms that are secondary to trauma,^[10,28] iatrogenic injury,^[10,17,21,29] or bacterial infection.^[10] As the present case had no history of trauma, iatrogenic injury, or infection, idiopathic pseudoaneurysm diagnosis was made. Similar to the present case, Ding *et al.* described

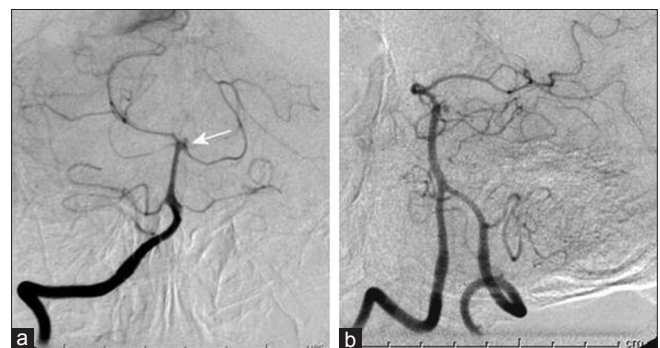


Figure 1: (a): Initial DSA shows a 3.5 mm pseudoaneurysm projecting distally from the basilar artery at the apex. (b) Complete spontaneous resolution was observed at 14 days after the initial imaging

two patients, each with an idiopathic pseudoaneurysm; one located along the posterior communicating artery and one located along the anterior communicating artery (ACoA).^[8] Charbel *et al.* described two cases of idiopathic anterior circulation pseudoaneurysms, both of which were located along the left ICA.^[5] All of these cases describe atherosclerosis on the parent vessel where the idiopathic pseudoaneurysm developed. It is hypothesized that atherosclerotic plaque deprives underlying tissues of oxygenation and nutrients, leading to subsequent tissue damage. Consequentially, these vessel walls may be more susceptible to dissection and disruption of the arterial wall with the formation of a pseudoaneurysm at the dissection site, particularly in hypertensive patients.^[30] The study by Takemoto *et al.* also supported this hypothesis that atherosclerosis may cause weakening of the arterial wall leading to pseudoaneurysm, demonstrating evidence of atherosclerosis such as intimal thickening and calcification in the media in histological specimen of spontaneous superficial temporal artery pseudoaneurysm.^[33] In children and young adults, however, idiopathic pseudoaneurysms can be observed without any associated risk factors.^[9,11]

Patients with this condition typically present with signs and symptoms consistent with SAH and include severe, thunderclap headache.^[5,10] Other features include neck rigidity,^[5,10] loss of consciousness,^[10] hypertension, and hypercoagulable state.^[5] Upon initial presentation in the cases reviewed, CT revealed SAH, and a subsequent four-vessel cerebral angiogram was typically performed.^[5,10] Initial DRA/DSA was successful in revealing pseudoaneurysms located in the posterior circulation in some cases as well as the present case.^[8] However, in cases where the pseudoaneurysm is found on the ICA, presence of the abnormality was only demonstrated after repeated angiograms, multiple imaging techniques, and/or during surgery, as Charbel *et al.* described two patients whose two four-vessel angiograms were initially either negative for aneurysm presence with only mild vasospasm or revealed a medial bulge on the left ICA but not a distinct aneurysm.^[5] Multiple imaging methods at the initial presentation and set intervals as clinically or radiologically indicated may provide clinicians multiple data points for more comprehensive care.

Treatment is typically necessary to repair the site of rupture of the pseudoaneurysm to avoid further subarachnoid hemorrhage and subsequent complications. Because a pseudoaneurysm is, by definition, a complete laceration or avulsion of all three layers of a vessel wall, they may be best treated as lesions whereby rupture or re-rupture is imminent. Typical variables to consider when selecting a treatment method include cause, location, and comfort of the treating physician with various treatment modalities. Because the procedural rupture rate is high

regardless of the treatment method, multiple modalities for treatment should be considered.

Three modes of treatment for pseudoaneurysms are open surgical, endovascular techniques, and conservative management. Surgical techniques that have been used to treat pseudoaneurysms include clipping, wrapping, clipping followed by wrapping, wrapping followed by clipping, and suturing. There have been multiple cases in which an encircling (Sugita) clip was successful in the treatment of an ICA and ACoA pseudoaneurysms.^[5,8,14] Use of rapid ventricular pacing was employed in a case of traumatic basilar pseudoaneurysm to reduce the turgor in the aneurysm wall to facilitate clipping.^[23] Another method of surgical treatment is wrapping of the pseudoaneurysm, either as the only method or combined with clipping using cellulose fabric or polytetrafluoroethylene.^[4,13,16] Endovascular options for the treatment of pseudoaneurysms are well described and are evolving rapidly with the incorporation of stent and flow-diversion devices. These techniques have demonstrated mixed results, but also undoubtedly improve with improvements in technology and techniques. Other methods include coiling, stent-assisted coiling, onyx embolization, balloon-assisted coiling, trapping, balloon occlusion followed by trapping, and flow diversion.^[2,7,10,12,15,16,19,25-28,34,35,38] Small wide necked pseudoaneurysms which are not amenable to endovascular treatment and high intraoperative rupture rates make these lesions challenging to treat.^[16,24] Unlike ruptured saccular aneurysms for which randomized clinical trials have been carried to determine the efficacy of treatment options, no data exists on which method of treatment is superior for managing pseudoaneurysms.^[20] On the other hand, conservative management and spontaneous resolution of pseudoaneurysms have been increasingly reported.^[3,18,22,31,32,36]

The patient in the present case report was treated conservatively due to decreased size and flow of the pseudoaneurysm in repeat CTA and DSA/DRA compared with the initial images. He was discharged from the ICU in neurologically intact condition when imaging at fourteen days revealed complete spontaneous resolution of the pseudoaneurysm. Spontaneous resolution of pseudoaneurysms is considered to be a rare phenomenon.^[3] However, many reports exist of spontaneous resolution in peripheral vessels and occasionally in intracranial vessels, including two cases in the middle meningeal artery,^[31,32] the vertebral artery,^[36] basilar artery pseudoaneurysm in a child,^[18] middle cerebral artery,^[3] and pericallosal artery.^[22] Common factors in these cases include history of head trauma and an absence of other radiographic abnormalities (such as atherosclerosis).

In the present case, both 14-day and 1-year follow-up imaging showed consistent resolution. Moreover,

spontaneous resolution of basilar artery perforator aneurysms has also been reported suggesting that aneurysms in the posterior circulation, possibly due to different flow patterns, may possibly undergo spontaneous resolution more frequently.^[6] Similar to the present case, spontaneous occlusion of a traumatic pericallosal pseudoaneurysm was seen 14 days after the first angiography.^[22] Furthermore, the histopathology of cadaveric pseudoaneurysms was shown to exhibit smooth muscle cell proliferation, macrophage accumulation, and lymphocytic infiltration around the injury site in vessel wall 7 days after SAH, which point to continuous remodeling process.^[30] It can be extrapolated that lack of atherosclerosis, as in our case, and physiological vascular remodeling response to injury as well as spontaneous thrombus formation leading to occlusion of the pseudoaneurysm may explain the mechanism of spontaneous resolution in the present case. Therefore, conservative management can be considered to be a treatment option when repeated imaging shows a decrease in pseudoaneurysm size and flow, taking into consideration the frail nature of these lesions and complications associated intervention. Some of the limitations of the study include lack of histological confirmation or intraoperative information about the lesion as it was managed conservatively and possible recall bias in patient history.

CONCLUSION

The present case is, to the best of our knowledge, the first reported incident of spontaneous resolution of an idiopathic pseudoaneurysm in the posterior circulation. All other reported incidences of spontaneous resolution have occurred where the pseudoaneurysm was caused by recent head trauma. None of these patients, including the present case, exhibited atherosclerosis of the parent vessel—this contrasts with the vast majority of pseudoaneurysm cases reviewed, in which treatment was necessary and atherosclerosis was present in both the parent vessel and the circle of Willis.

Treatment for an intracranial pseudoaneurysm should be carefully considered, and be based on the location of the aneurysm and the comfort level of a surgeon to perform a specific procedure deemed most appropriate. A conservative approach may be considered for selected pseudoaneurysms where potentially atherosclerosis is not present and where consecutive imaging shows a decrease in size and flow of the aneurysm.

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Conflicts of interest

There are no conflicts of interest.

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