Case Report

An infundibulum of thalamoperforator arteries: Importance of angiographic images for appropriate diagnosis

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Abstract

Background: The identification of infundibula on noninvasive imaging modalities may be challenging. Because these lesions have generally been viewed as nonpathological, distinguishing them from small or micro-aneurysms is important.

Case Description: A 39-year-old male was diagnosed with recurrence of typical orgasmic headache. An outpoutching arising from the distal part of the right P1 at the take-off of thalamoperforator arteries was visualized on noninvasive investigations. The patient was referred to neurosurgery for surgical management of a right P1 aneurysm. Its unusual location and morphology led to be suspicious of an infundibular dilatation. Catheter angiography with 2D projections and 3D rotational reconstruction revealed an infundibulum at the common origin of two thalamoperforators, giving rise to a double-peaked shape, mimicking a true aneurysm, rather than the more characteristic conical shape of an infundibulum.

Conclusion: Although noninvasive modalities may identify typical infundibula, the catheter angiogram with 2D projections was critical to establishing the diagnosis. The 3D rotational reconstruction enabled a straightforward understanding of the 3D vascular anatomy. This pyramidal variant of infundibular dilatation should be included in the differential diagnosis of a wide-based nonsaccular arterial contour deformities located in an area of multiple perforators.

Key Words: Aneurysm, angiography, dissecting aneurysm, infundibulum, perforator, posterior cerebral artery

INTRODUCTION

The identification of infundibula on noninvasive imaging modalities may be challenging. Although computed tomography angiography (CTA) and magnetic resonance angiography (MRA) have become the primary imaging screening techniques for detection of possible aneurysms, the limitations of these modalities in identifying outpouchings and rendering detailed assessment of their relationship with branches/perforators has been recognized. Because infundibula have generally been viewed as nonpathological, distinguishing them from small or micro-aneurysms is important. We present a unique case of an infundibulum at the common origin of two thalamoperforators, giving rise to a double-peaked shape that mimics a true aneurysm, discuss the differential

diagnosis, and emphasize the importance of catheter angiography for appropriate diagnosis and management.

CASE REPORT

Clinical history
A 39-year-old male was referred for management of a possible right P1 aneurysm. The patient was in good general health and his family history was negative for any cerebrovascular pathology. He recently experienced recurrence of his typical coital headaches, which had been diagnosed in his twenties. For completeness, the referring physician requested brain imaging.

Investigation
The initial head computed tomography (CT) scan performed within 48 h of his last headache failed to show acute subarachnoid hemorrhage. The CTA revealed a rounded bulge arising from the right distal P1 segment of the right posterior cerebral artery (PCA) [Figure 1a and b], suspicious for a saccular aneurysm. No other abnormality was identified. The brain magnetic resonance imaging (MRI) showed no hyperintense signal on T1WI or Flair sequences to suggest subarachnoid blood, no enhancement on the T1WI postgadolinium sequence, and no mass in the interpeduncular or crural cisterns, making a dissecting aneurysm much less probable [Figure 1c]. The GRE sequence showed no hypointense hemosiderin deposit. The brain MRA showed the same right P1 bulge, with no additional information gained. The patient agreed to undergo a catheter angiogram in order to clarify the angioanatomy at the site of the right P1 bulge. A catheter angiogram with 2D projections and 3D rotational reconstructions revealed a small 3 mm outpouching just proximal to the junction of the right posterior communicating arteries (PcomA) and PCA [Figure 2a and b]. In addition, two small thalamoperforators were now clearly seen arising from the dilatation creating a double-peaked shape to the infundibulum, rather than the conical shape more typically associated with infundibula. The catheter

angiogram also enabled visualization of two additional small infundibula, each measuring approximately 2 mm in maximal diameter at the origins of the left anterior choroidal artery (AchoA) and left PcomA [Figure 2c and d]. No true aneurysm was seen.

DISCUSSION

Infundibulum: Definition and variation
By definition, an infundibulum is a conical, triangular, or funnel-shaped dilatation of the origin of a major branch of the internal carotid artery (ICA).[6,8] Although Salkman's initial description addressed only PcomA infundibula, the dilatations have also been detected at branching sites of the AChoA.[19] The typical infundibulum has a widening of pyramidal shape, with its base facing the parent artery and measuring ≤3 mm, and its apex giving rise to the branching artery (generally the PcomA or AChoA).[8] In areas rich in perforating vessels, such as the P1 segment of the PCA and the M1 segment of the MCA, infundibular dilatations may incorporate the origin of more than one perforator, giving the infundibulum a double-peaked shape, rather than the more conical form typically associated with an infundibulum. At spatial resolutions below those of conventional catheter angiography, the double-peaked shape may be blurred into a more rounded appearance that can mimic a true aneurysm.

Therefore, in the context of a wide-based nonsaccular bulge located in an area of multiple perforators, the differential should include not only an atypical aneurysm or a dissecting aneurysm, but also a variant infundibular dilatation. Although we have encountered infundibular dilatations at the origin of the lenticulostriate arteries, this is the first observation and, to the authors' knowledge, the first description in the literature of an infundilum at the origin of thalamoperforators.

Identification of infundibular dilatations
Infundibular dilatations were initially described on cerebral angiograms by Saltzman in 1959.[9,12,13,15,18]

Figure 1: Brain CTA axial source image, (a) Brain CTA reconstruction, (b) Brain MRI axial T1WI postgadolinium, (c) Both noninvasive modalities show a small dilatation in the region of the right PCA (white arrow)

Figure 2: Left vertebral artery injection, (a) 2D angiography and, (b) 3D rotational angiography and left common carotid artery injection (c) 2D angiography and, (d) 3D rotational angiography. a and b show the double peaked shape infundibulum (white arrow) arising proximal to the junction of the right PcomA and PCA. (c and d) the two infundibuli (white asterixis) at the origins of the left AchoA and left PcomA

Both CTA and MRA have become the primary imaging screening techniques for detection of intracranial aneurysms. Although multiple studies have assessed the accuracy of these noninvasive modalities for evaluating unruptured aneurysms, few studies have evaluated their sensitivity to diagnose infundibula.[9,12,13,15,18] Recognizing the limitations of these modalities in identifying outpouchings and rendering detailed assessment of their relationship with branches/perforators is important. This is because the pixel size in conventional catheter angiography is 1024 × 1024, while that of conventional CTA is generally 512 × 512, and that of MRA is generally no higher than 384 × 256. This means that the spatial resolution of conventional catheter angiography is up to four times higher than CTA, and far higher than that of MRA. The impact of a lower spatial resolution is that fine image detail is blurred, and that two structures that are located close together may appear to be fused on CTA or MRA. Image data acquired by CTA and MRA using limited spatial resolution may be further compromised by the tendency of postprocessing software to blur image borders when creating 3D volume rendered or multiplanar reformatted images, through the use of smoothing algorithms designed to create visually appealing images. Despite the fact that CTA and

MRA have become the primary imaging techniques for detection and characterization of vascular outpouchings in many neurosurgical centers, conventional catheter angiograms remain the gold standard for the diagnosis of infundibula, and for the accurate depiction of small and very small vascular angioanatomy.[6,18]

Infundibula have been identified in 7-25% of catheter angiograms.[6,18,26] Although they may be more frequently documented in patients with multiple or familial aneurysms, up to 25% of nonfamilial cases have bilateral infundibula. In the present case, the 2D catheter angiography projections were essential for the diagnosis as other modalities failed to visualize the perforators arising from the dilatation. The 3D rotational reconstruction accomplished by the rotational angiography equipment confirmed the findings of the 2D projections and enabled a straightforward understanding of the 3D vascular anatomy, making sure the perforators were not vessels superimposed on the outpouching. Three-dimensional rotational angiography with volume rendering has shown superiority to 2D angiography in diagnosis of both cerebral aneurysms and infundibular dilatations.[19] Interpretation of 3D images acquired during rotational angiography should be done cautiously in conjunction

with the 2D angiography to exclude potential artifacts resulting from the 3D reconstruction processing.[19]

Clinical significance of infundibula

The clinical significance of a particular infundibulum is impossible to predict solely by imaging. Hassler and Saltzman have described several cases of PcomA infundibula, which presented defects in the media and splitting of the internal elastic lamina, histological changes similar to those found in aneurysms. However, none of the patients of their original series had presented with subarachnoid hemorrhage. Although for most clinicians the typical infundibular dilatations have minimal pathogenic significance and risks, some authors consider them to be pre-aneurysmal lesions.[1,4,14,16,17,19,21,22] Documentation of evolution from an infundibulum to an aneurysm with subsequent rupture is present, although scant in the literature.[1,4,14,16,21,22] Among the reported cases, progression to aneurysm occurred between 0.5 and 9 years following the diagnosis of infundibulum.[1,4,14,16,21,22] Distinguishing an infundibulum from a small aneurysm is important. Although historical data from the ISUIA study suggested a low risk of rupture for small (2-7 mm) unruptured aneurysms in patients without a previous subarachnoid hemorrhage,[24] the median size of ruptured cerebral aneurysms is also small.[11,20,25] In addition, the growth rate of unruptured intracranial aneurysms is highly variable and unpredictable, with reported annual incidences varying between 1.51% and 22.7% depending on the definition of aneurysm growth and length of follow-up.[3,7,10,20] Importantly, all aneurysms, even those of small size, carry a risk of enlargement over time.[3,5,7,10,20] Aneurysm growth has been associated with a higher annual rupture risk after growth has occurred, hence the importance of radiographic follow-up.[10]

To date, there is no consensus regarding if, when, how, and for how long imaging follow-up should be coordinated in the presence of a well characterized infundibulum or even small asymptomatic aneurysms.[2,23] In addition to the rareness of evolution from an infundibulum to an aneurysm, the lack of systematized radiographic follow-up impedes assessment of the true incidence of this evolution. In the present case, although the CTA did not allow to clearly visualize the two small thalamoperforators arising from the dilatation, the size and configuration of the outpouching were well documented. Given the higher spatial resolution of the CTA, we recommended a CTA in 6 months to follow-up on this unusually located infundibulum, and then yearly for the first 3 years. Although studies have documented that MRA 3.0T technology has comparable sensitivity to multidetector CTA for the diagnosis of aneurysms, we found, as well as other groups, that the reformatted 3D images of aneurysms from noncontrast-enhanced MRA are inferior compared with those from CTA.[9,15]

CONCLUSION

In summary, this is the first description of an infundibulum at the common origin of two thalamoperforators, giving rise to a double-peaked shape that mimics a true aneurysm, rather than the more characteristic conical shape of an infundibulum. This variant of infundibular dilatation should be included in the differential diagnosis of a wide-based nonsaccular arterial contour deformities located in an area of multiple perforators. Although noninvasive modalities may identify typical infundibula, the catheter angiogram with 2D projections may be critical to establish the diagnosis in some patients, with the 3D rotational reconstruction allowing a straightforward understanding of the 3D vascular anatomy.

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