



Novel Aneurysm Grown from a PcomA Infundibulum after Embolization of a Contralateral Posterior Communicating Aneurysm in an Isolated Polycystic Hepatic Patient: A Case Report

Rui Zhao[#], Chao Zou[#], Chuan-Chuan Wang, Qing-Hai Huang and Jian-Min Liu^{*}

Department of Neurosurgery, Stroke Center, Changhai Hospital, Second Military Medical University, China

[#]Equal Contribution

***Corresponding author:** Jian-Min Liu, Department of Neurosurgery, Stroke Center, Changhai Hospital, Second Military Medical University, 168 Changhai Road, Shanghai 200433, China, Tel: 8513916728169; E-mail: chstroke@163.com

Abstract

We present a case of a posterior communicating (Pcom) artery infundibulum that progressed to an aneurysm in a patient with Polycystic liver disease without kidney cyst after embolization of the contralateral Pcom aneurysm. The infundibulum had been documented on angiography 4 years earlier, while a ruptured contralateral Pcom aneurysm was embolized. The patient presented to the outpatient clinic with right postorbital headache and cranial III nerve palsy. Head CT scan found no gross subarachnoid hemorrhage. Though MR angiography note a 7-mm pcom aneurysm at the previous site of Pcom infundibulum. The patient underwent coiling for the novel aneurysm with a good result. This case illustrates a small but growing number of cases which developed from infundibula into aneurysms. It suggests that polycystic disease may pose an additional risk for infundibulum while what has traditionally been considered as a benign lesion.

Keywords

Aneurysm, Infundibulum, Polycystic liver disease

Introduction

Intracranial Arterial Infundibulum (IAF) is frequently regarded as incidental normal anatomical variant which rarely causes severe events. Up to now, reports of IAF rupture and aneurysms arising from IAF were rare. We report a case of development of aneurysm from IAF at origin of Pcom artery with a recurrent contralateral Pcom Aneurysm and a history of Polycystic Liver Disease (PLD).

Case Report

A 48-year-old man suffered sudden onset of headache with nausea and vomiting. CT scanning revealed a diffused SAH. He had a Polycystic Liver Disease (PLD) without kidney cyst, a ten-year history of HBP, no history of cigarette smoking, no familial history of intracranial aneurysm or Autosomal dominant PLD (ADPLD). After admission, cerebral angiography disclosed a left Pcom Aneurysm (Figure 1A) with an infundibulum (Figure 1C) at

the origin of right Pcom artery. The aneurysm measured 6.10mm in width, 2.86mm in neck, 5.50mm in height. The infundibulum is less than 3mm in diameter. The patient was discharged with no symptom after the aneurysm was embolized with GDC coils successfully. He has no neurologic symptom during two-year clinical follow-up at outpatient clinic and had no more follow-up until the 51st month after the treatment. The patient came back to our outpatient clinic and complained of paroxysmal right post-orbit headache with cranial III nerve palsy symptom. We found out that he hadn't had a regular visit to a physician to control his blood pressure well after aneurysm embolization. CT scanning revealed no SAH. Follow-up cerebral angiography suggested neck recurrence of L-Pcom Aneurysm (Figure 1B) and a novel saccular aneurysm (Figure 1D) at the previous site of right Pcom infundibulum. The size of novel aneurysm is 7.53 mm in length, 3.33 mm in width.

Endovascular treatment was considered as the first choice since the novel aneurysm was unruptured and the recurrent one had a small sac. The symptoms were assumed to be caused by the enlargement of right PcomAn. So, we performed the endovascular treatment of the novel aneurysm.

The novel aneurysm was successfully embolized by coiling with GDC coils. Post-embolization angiography suggested a neck residual (Figure 1E). Although the patient had no symptom, recurrence of R-Pcom aneurysm without obvious change of the left one was showed by 3-month follow-up angiography (Figure 1F). We considered retreating both aneurysms. But the patient refused and complained of no symptom in 4 more year's clinical follow-up.

Discussion

IAFs have variously been described as funnel-shaped or triangular symmetrical widenings at origins of major cerebral arteries, principally occurring at the junction of the ICA and PComA [1]. IAFs are revealed on 7% to 25% of otherwise normal angiograms [2,3], and the incidence seems to be greater in cases of multiple [2] or familial [3] aneurysms.

Citation: Zhao R, Zou C, Wang CC, Huang QH, Liu JM (2014) Novel Aneurysm Grown from a PcomA Infundibulum after Embolization of a Contralateral Posterior Communicating Aneurysm in an Isolated Polycystic Hepatic Patient: A Case Report. Int J Neurol Neurother 1:008. doi.org/10.23937/2378-3001/1/1/1008

Received: October 05, 2014; **Accepted:** October 28, 2014; **Published:** November 01, 2014

Copyright: © 2014 Zhao R. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

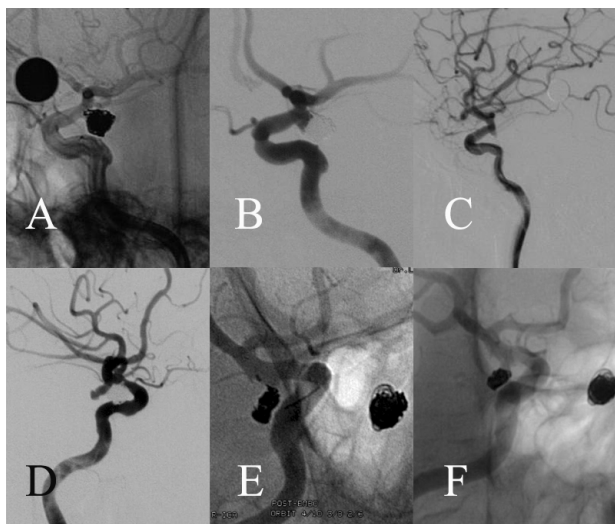


Figure 1: A: Postoperative angiogram of the ruptured left ICA PcomA aneurysm. B: Four-year follow-up angiogram of the left ICA PcomA aneurysm. C: A 3-mm, funnel-shaped infundibulum at junction of right ICA-PcomA with a branch arises from whose tip. D: A novel aneurysm was revealed at right PcomA at the site of previous infundibulum at 4-yr-FU. E: Postoperative angiogram of the novel right ICA PcomA aneurysm. F: DSA follow-up at 3 months revealed neck recurrence of the right PcomA aneurysm.

The significance of IAF is under controversy. It is frequently regarded as normal anatomical variants that without pathogenetic significance [4], for its not too low occurrence in general population but generally resulting in no event. Some authors believe this lesion is preaneurysmal [3], requesting more attention and regular follow-ups for its higher incidence in patients with multiple, familial intracranial aneurysms than general people. Further more, histological examinations shew medial defect with a disrupted internal elastic lamina in some IAFs, just the same as that in normal artery bifurcations and aneurysms [5]. Up to now, six cases of IAF rupture without aneurysmal formation were reported [3], and the authors suggested more attention be paid on some isolated IAF cases with SAH. These rare cases put us a not much too convincible but unoverlooked evidence of pathogenetic significance of IAF after all.

We present here the case of a man with isolated PLD and a history of HBP and a rupture left PCoA aneurysm in which a contralateral PCoA aneurysm developed at the site of a previously documented infundibulum during follow-up angiography. To our knowledge, this is the 14th known case about infundibulum growing into aneurysm in the literature [6-8]. According to our case and literature review the isolated PLD [9] and hypertensive may be risk factors for the novel aneurysm formation and recanalization of the embolized one. Cowan et al. [8] reported a case of progression from IAF to aneurysm in a man with Alagille Syndrome, a disease which may result in high incidence of systematic arterial lesions, including CVD.

Autosomal Dominant PLD (ADPLD), a rare inherited Mendelian disorder (with ADPLD gene mutation of *PRKCSH* [10] and *SEC63* [11]) that is characterized by development of multiple hepatic cysts, is genetically different from Autosomal-Dominant Polycystic Kidney Disease (ADPKD). The latter is a hereditary systemic connective tissue disease involving cardiocerebral vessels [12], especially intracranial aneurysm [13]. ADPLD has been reported significantly relative with cerebral aneurysms at autopsy [14] and patients [15]. The occurrence of vascular type in ADPLD patients is much higher than those in the general population [16]. We can't diagnose this patient ADPLD, because we did not perform the gene analysis. As our case is concerned, the vessel wall abnormality caused by polycystic disease may be a risk factor of aneurysmal occurrence and relapsing after occlusion the sac. So we suggest that patients with polycystic disease should accept screening and regular follow-ups for cerebrovascular disease, especially for intracranial aneurysm.

Acknowledgement

This work was supported by the National Natural Science Foundation of China (81000495).

References

- Yoshimoto T, Suzuki J (1974) Surgical treatment of an aneurysm on the funnel-shaped bulge of the posterior communicating artery. *J Neurosurg* 41: 377-379.
- Leblanc R, Worsley KJ, Melanson D, Tampieri D (1994) Angiographic screening and elective surgery of familial cerebral aneurysms: A decision analysis. *Neurosurgery* 35: 9-18.
- Coupe NJ, Athwal RK, Marshman LA, Brydon HL (2007) Subarachnoid hemorrhage emanating from a ruptured infundibulum. Case report and literature review. *Surg Neurol* 67: 204-206.
- Edelsohn L, Caplan L, Rosenbaum AE (1972) Familial aneurysms and infundibular widening. *Neurology* 22: 1056-1060.
- Archer CR, Silbert S (1978) Infundibula may be clinically significant. *Neuroradiology* 15: 247-251.
- Marshman LA, Ward PJ, Walter PH, Dossetor RS (1998) The progression of an infundibulum to aneurysm formation and rupture: case report and literature review. *Neurosurgery* 43: 1445-1449.
- Martins C, Macanovic M, Costa e Silva IE, Griz F, Azevedo-Filho HR (2002) Progression of an arterial infundibulum to aneurysm: case report. *Arq Neuropsiquiatr* 60: 478-480.
- Cowan JA Jr, Barkhoudarian G, Yang LJ, Thompson BG (2004) Progression of a posterior communicating artery infundibulum into an aneurysm in a patient with Alagille syndrome. Case report. *J Neurosurg* 101: 694-696.
- Schievink WI, Spetzler RF (1998) Screening for intracranial aneurysms in patients with isolated polycystic liver disease. *J Neurosurg* 89: 719-721.
- Drenth JP, TeMorsche RH, Smink R, Bonifacio JS, Jansen JB (2003) Germline mutations in *PRKCSH* are associated with autosomal dominant polycystic liver disease. *Nat Genet* 33: 345-347.
- Davila S, Furu L, Gharavi AG, Tian X, Onoe T, et al. (2004) Mutations in *SEC63* cause autosomal dominant polycystic liver disease. *Nat Genet* 36: 575-577.
- Arnaout MA (2000) The vasculopathy of autosomal dominant polycystic kidney disease: Insights from animal models. *Kidney Int* 58: 2599-2610.
- Niemczyk M, Gradzik M, Niemczyk S, Bujko M, Gołębowski M, et al. (2013) Intracranial aneurysms in autosomal dominant polycystic kidney disease. *AJNR Am J Neuroradiol* 34: 1556-1559.
- Geevarghese SK, Powers T, Marsh JW, Pinson CW (1999) Screening for cerebral aneurysm in patients with polycystic liver disease. *South Med J* 92: 1167-1170.
- Schievink W, Spetzler RF (1998) Screening for intracranial aneurysms in patients with isolated polycystic liver disease. *J Neurosurg* 89: 719-721.
- Qian Q, Li A, King BF, Kamath PS, Lager DJ, et al. (2003) Clinical profile of autosomal dominant polycystic liver disease. *Hepatology* 37: 164-171.