Content available at: https://www.ipinnovative.com/open-access-journals

IP Journal of Nutrition, Metabolism and Health Science

Journal homepage: https://www.jnmhs.com/

Case Report

Fourteen intracranial aneurysms in a single patient—the largest number ever reported in the literature

AL Periyakappan[1], K Mohamed Rafi[1]*

[1]*Dept. of Inventional Radiology, Vinayaka Missions Medical College, Mansarover Garden, New Delhi, India*

ARTICLE INFO

Article history:
Received 16-12-2023
Accepted 31-01-2024
Available online 15-02-2024

Keywords:
Intracranial Aneurysm
DSA
Multiple Aneurysm
Sacular
Fusiform

ABSTRACT

A 52-year-old female came with complaints of intense headaches for the past 10 days. The CT brain revealed SAH in the bilateral Sylvain fissure (left more than right) with Hunt and Hess Grade 1. An urgent CT cerebral angiogram was done, which revealed multiple bilateral MCA, ACA, and basilar top aneurysms. DSA was done the next day, which revealed multiple aneurysms, a total of fourteen in number. Such multiple aneurysms in a single patient were never reported in the literature until now, according to the present author's knowledge.

1. Introduction

Multiple intracranial aneurysms usually range from 15% up to 35% of patients presenting with non-traumatic aneurysmal subarachnoid haemorrhage (SAH). Increased age and female sex are significantly associated with SAH in cases of multiple aneurysms.[1] The occurrence of multiple aneurysms in a single patient without any underlying systemic pathology or concurrent family history is rarely reported in the literature.

2. Case Report

A 52-year-old female came with complaints of intense headaches for the past 10 days. She was known to be hypertensive and had been on irregular medication for the past 10 years. She was also diagnosed with a case of medical renal disease two years ago. For this, she was put on a salt-restricted diet with conservative management. She was not a known diabetic or had any previous history of CAD. She does not have a significant past surgical history. There is no previous history of blood transfusions. She is G2P2AOL2, and both the girl babies are delivered via normal vaginal delivery. There is no history of similar complaints among the family members. There is no adverse social history.

On general examination, she was not icteric and had no signs of pallor or clubbing. There is no significant cervical lymphadenopathy. CVS, RS, and abdomen examinations were unremarkable and showed the presence of mild neck rigidity. The cranial nerve examination was unremarkable. The powers of the bilateral upper and lower limbs are normal (5/5). Bilateral plantar reflexes are flexors. Refluxes are not exaggerated. She was stable, conscious, and oriented with GCS-15/15.

A CT brain done outside revealed thin SAH in the bilateral Sylvian fissure (left more than right). There is no evidence of intra-ventricular haemorrhage. Hunt and Hess grade for SAH is Grade 1. An urgent CT cerebral angiogram was done, which shows multiple small bilateral MCA, ACA, and large basilar top aneurysms (Figures 2 and 3). A blood investigation revealed raised urea (84 mg/dl) and creatinine (4.6 mg/dl). mild hyponatremia (129 meq/l) and a normal potassium level (3.5 meq/l). Clotting parameters

* *Corresponding author*.
E-mail address: radiologistrafi1984@gmail.com (K. Mohamed Rafi).

https://doi.org/10.18231/j.ijnmhs.2023.032

and a complete blood count are within normal limits.

The echo cardiogram shows concentric left ventricular hypertrophy. There were no regional wall motion abnormalities. The ejection fraction was 66%. Normal left ventricular systolic function. Moderate MR with AML prolapsed. Trivial AR and mild TR No PHT/PE. There is no intracardiac mass like myxoma.

Rheumatological examination revealed no obvious features of connective tissue disorder. The USG abdomen revealed bilaterally increased renal cortical echoes with normal CMD (Grade 1 medical renal parenchymal disease). There is no evidence of ADPKD. The next day, DSA was done, which revealed multiple aneurysms of a total of fourteen in number (Figures 4, 5, 6 and 7). Such multiple aneurysms in a single patient were never reported in the literature until now, according to the present author's knowledge.

3. Discussion

In the adult population, the prevalence of IC aneurysm is about 1 to 3.2%. Of these cases, multiple intracranial aneurysms were noted in 2–45% of the patients. 40% of intracranial aneurysms and 5% of IC aneurysms are mirror aneurysms.[1] A mirror aneurysm is defined as the presence of a symmetrical aneurysm in the bilateral arteries of the same segment.

A true aneurysm is defined as the dilation of all the walls of the arterial lumen. The cause may be due to either congenital or acquired abnormalities that lead to a loss of vascular wall mechanical resistance. The most common aetiology is the absence or weakness of the elastic or muscular layer of the vessel.[2]

Based on morphological appearance, aneurysms are classified into saccular, fusiform/dolichoectatic, dissecting, posttraumatic, mycotic, and giant aneurysms without or with intra-aneurysmal thrombosis.[3] Saccular aneurysm is the most common type of IC aneurysm.

Atherosclerosis and arterial hypertension are important risk factors for the development of aneurysms in the elderly population. In young people, connective tissue disorder plays a significant role in the early development of aneurysm.[2] Other risk factors include smoking, alcohol, and a family history of aneurysm.

Genetics plays a vital role in the development of IC aneurysms in patients with a positive family history. Hereditable conditions like Ehlers-Danlos syndrome, Marfan syndrome, and neurofibromatosis-1 lead to the development of IC aneurysm, which is 10–20% more common than in the general population. It also causes an 8–25% higher rate of IC aneurysm rupture when compared to the general population.[4] This syndrome causes a mutation in the structural proteins like COL1A1 in Ehlers-Danlos syndrome, leading to weakness of the vessel wall and aneurysmal development. Genome-wide association studies (GWAS) from large cohorts from Dutch, Finnish, and Japanese populations have found that many single nucleotide polymorphisms (SNPs) are more common in individuals with IC aneurysms.[5]

The familial aneurysm had some predilection for involvement of the middle cerebral artery. Multiple studies have shown a statistically significant association between intracranial aneurysm formation, SAH, and variants of the genes CDKN2 (chromosome 9), EDNRA, and SOX17 (chromosome 8).

Patients with one affected family member have a 4% increased risk, and ones with two or more affected first-degree family members have an 8–10% increased risk of developing an aneurysm.[6] The prevalence of intracranial aneurysms is four times higher in patients with ADPKD than in the general population.[7]

There are various modifiable and non-modifiable risk factors associated with intracranial aneurysm development and rupture.[8–10] Non-modifiable factors include older age, women, history of prior aneurysm or subarachnoid haemorrhage, family history of intracranial aneurysm or subarachnoid haemorrhage, and Finnish or Japanese ethnicity. Modifiable factors include hypertension, smoking, and excess alcohol intake. The risk of subarachnoid haemorrhage is three to seven times higher in first-degree relatives of patients than in the general population, but similar to the general population in second-degree relatives.[11] The other causes of the development of IC aneurysm included vasculitis, Diamond-Blackfan anaemia, and atrial myxoma.[12]

Coiling is currently accepted as a standard treatment for ruptured or unruptured multiple intracranial aneurysms with or without stent or balloon assistance, depending upon the morphology and location of the aneurysm and the parent vessel. The flow diverter will also play a role, depending on the morphology of the aneurysm. Nevertheless, many cases of clipping for multiple aneurysms have also been reported.[13]

This young female does not have features of connective tissue disorder, mycotic cause, or intracardiac mass. The patient was explained regarding both endovascular and neurosurgical management. The patient deferred treatment due to the risk involved in the procedure. The patient was discharged and asked to attend OPD in case of an emergency.

4. Conclusion

Multiple aneurysms, totally fourteen in number in a single patient were never reported in the literature till now. It is First case to be reported in literature in worldwide, according to the present author knowledge at the time of authoring this article.

Table 1: A list of aneurysms is described in the table

S. No	Location of Aneurysm	Segment	Length(mm)	Width(mm)	Neck Diameter(mm)
1	Right ICA	Cavernous	4.8	3.3	2.5
2	Right MCA	M1	1.3	1.1	1.3
3	Right MCA	M2 Bifurcation	0.6	1.9	
4	Right ACA	A2 Segment	0.7	0.7	1
5	Right ACA	A2 Segment	2.4	1.8	1.8
6	Right ACA	A2 Segment	2.6	3.5	2.2
7	Right ACA	A3 Segment	0.8	0.8	1.4
8	Right ACA	A3 Segment	2.2	2.7	1.2
9	Right ACA	A3 Segment	4.5	3.8	3
10	Left ICA	Cavernous	1	1.1	1.4
11	Left ICA	Infundibular	1	0.6	1.3
12	Left ICA	Carotico-Opthalmic	5.1	3.6	4.1
13	Left MCA	M2 Bifurcation	0.6	0.7	1.2
14	Basilar Artery	Top	9.7	7.9	7

Figure 1: Photograph of the patient.

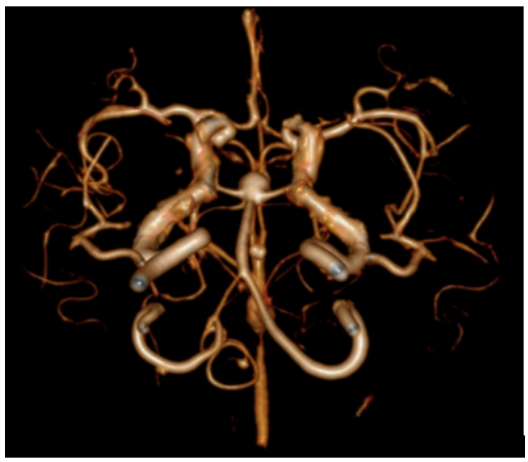

Figure 2: CT angiogram Image large Basilar top aneurysm

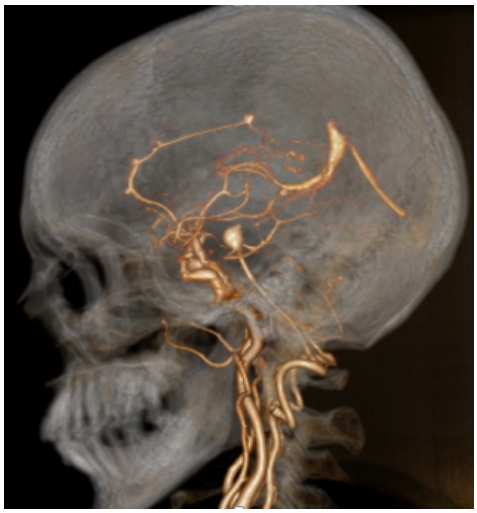

Figure 3: CT angiogram Image multiple small aneurysms along the ACA.

Figure 4: 3D DSA-reconstructed image of RT ICA Frontal projection shows multiple aneurysms in the right ICA, MCA, and ACA.

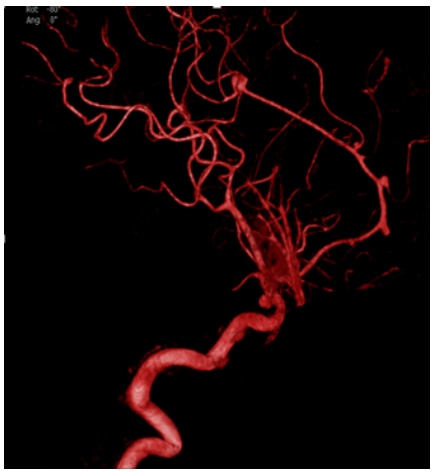

Figure 5: A 3D DSA-reconstructed image of the RT ICA lateral oblique projection shows multiple aneurysms in the right ICA and ACA.

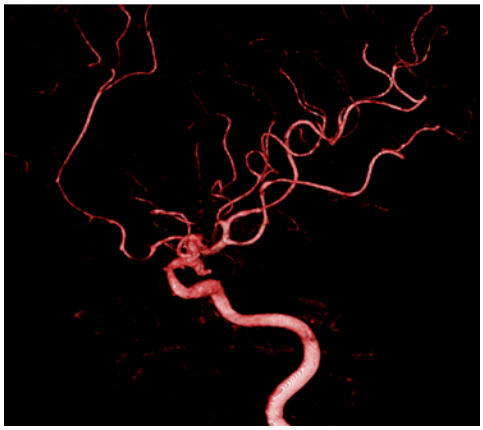

Figure 6: 3D DSA reconstructed image of Left ICA frontal projection shows multiple aneurysms in the left ICA.

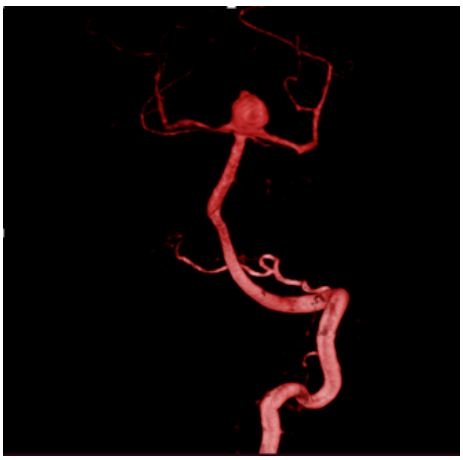

Figure 7: 3D DSA reconstructed image of Left vertebral artery frontal projection shows large basilar top aneurysm.

5. Abbreviations

1. SAH-Sub arachnoid Hemorrhage
2. DSA- Digital Subtraction Angiography
3. MCA- Middle cerebral artery
4. ACA- Anterior cerebral artery
5. CAD- Coronary Artery Disease
6. IC- Intracranial
7. GCS- Glascow Coma Scale
8. CVS- Cardio-Vascular System
9. RS- Respiratory System
10. AML- Anterior Mitral Leaflet
11. AR- Aortic Regurgitation
12. TR- Tricuspid Regurgitation
13. MVP- Mitral Valve Prolapse
14. PHT- Pulmonary Hypertension
15. PE- Pulmonary Embolism
16. ADPKD- Autosomal Dominant polycystic kidney Disease

6. Declarations

6.1. Ethics approval and consent to participate

Ethical approval is not obtained, since it's a retrospective case report study. Written consent had been obtained from the participant for participation.

7. Source of Funding

None.

8. Conflict of Interest

None.

References

1. Rajagopal N, Yamada Y, Balaji A, Kawase T, Kato Y. Rare cases of multiple unruptured intracranial aneurysms: Illustrative report and review on management options. *Interdiscip Neurosurg*. 2019;18:100466. doi:10.1016/j.inat.2019.04.011.
2. Knap D, Partyka R, Zbroszczyk M. Tętniaki mózgu - współczesne metody leczenia wewnątrznaczyniowego. *Pol Prz Neurol*. 2010;6(1):22–6.
3. Meyers PM, Schumacher HC, Higashida RT. Reporting standards for endovascular repair of saccular intracranial cerebral aneurysms. *J Neurointerv Surg*. 2010;2(4):312–35.
4. Caranci F, Briganti F, Cirillo L, Leonardi M, Muto M. Epidemiology and genetics of intracranial aneurysms. *Eur J Radiol*. 2013;82(10):1598–605.
5. Poppenberg KE, Jiang K, Tso MK, Snyder KV, Siddiqui AH, Kolega J, et al. Epigenetic landscapes suggest that genetic risk for intracranial aneurysm operates on the endothelium. *BMC Med Genomics*. 2019;12(1):149. doi:10.1186/s12920-019-0591-7.
6. Brown RD, Broderick JP. Unruptured intracranial aneurysms: epidemiology, natural history, management options, and familial screening. . *The Lancet Neurol*. 2014;13(4):393–404.
7. Sanchis IM, Shukoor S, Irazabal MV, Madsen CD, Chebib FT, Hogan MC, et al. Presymptomatic Screening for Intracranial Aneurysms in Patients with Autosomal Dominant Polycystic Kidney Disease. *Clin J Am Soc Nephrol*. 2019;14(8):1151–60.
8. Brown RD, Torner J. Unruptured intracranial aneurysms: Some questions answered, many questions remain. Re: Pelz D. CURES and the dilemma of unruptured intracranial aneurysms. *Can J Neuro Sci*. 2011;38(2):191–3.
9. Macdonald RL, Schweizer TA. Spontaneous subarachnoid haemorrhage. *Lancet*. 2017;389(10069):655–66.
10. Bromberg JE, Rinkel GJ, Algra A, Greebe P, Van Duyn C, Hasan D, et al. Subarachnoid haemorrhage in first and second degree relatives of patients with subarachnoid haemorrhage. *BMJ*. 1995;311(7000):288–9.
11. of Unruptured Intracranial Aneurysms Investigators IS. International Study of Unruptured Intracranial Aneurysms Investigators: Unruptured intracranial aneurysms–risk of rupture and risks of surgical intervention. *N Engl J Med*. 1998;339(24):1725–33.
12. Trivedi RA, Watts C, Kirkpatrick PJ. Multiple cerebral aneurysms and the Diamond-Blackfan syndromeJournal of Neurology. *Neurosurg Psychiatry*. 2002;72(5):678–9.
13. Murrone D, Romanelli B, Ierardi A. Surgical case series of multiple aneurysms: A single-centre experience of 16 years. *Int J Surg Case Rep*. 2017;42:5737947.

Author biography

AL Periyakappan, Associate Professor

K Mohamed Rafi, Senior Resident https://orcid.org/0000-0002-8225-9387

Cite this article: Periyakappan AL, Mohamed Rafi K. Fourteen intracranial aneurysms in a single patient—the largest number ever reported in the literature. *IP J Nutr Metab Health Sci* 2023;6(4):194-198.