

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr



Case Report

Endovascular NBCA treatment of a ruptured arteriovenous malformation with venous pseudoaneurysm in a young child [†]

Federico De Lucia, MD^{a,*}, Thomas Bonnet, MD^b, Stephanie Elens, MD^b

^a Radiology, Université Libre de Bruxelles, Brussels, Belgium ^b Department of Radiology, Hôpital Erasme, Brussels, Belgium

ARTICLE INFO

Article history: Received 9 July 2023 Revised 6 August 2023 Accepted 8 August 2023

Keywords:

Arterio-venous malformation Venous pseudoaneurysm N-butyl cyanoacrylate Neuroradiology Interventional radiology

ABSTRACT

We reported imaging findings of arterio-venous malformation complicated by hemorrhage and venous pseudoaneurysm in a young child consulting for headache and emesis: to our knowledge venous pseudoaneurysm in association with ruptured arteriovenous malformation is a rare complication reported in the literature. We present the indications for endovascular treatment, especially with NBCA (N-butyl cyanoacrylate).

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Arteriovenous malformations (AVMs) are high flow lesions formed by feeding arteries, a cluster of vessels (the nidus) and drainage veins without interposition of capillaries. The incidence of cerebral AVMs in childhood is expected to be 1 per 100,000 children [1]. The treatment consists in microsurgery, endovascular embolization, radiosurgery, and the combination of these techniques.

Case report

A 6-year-old male patient consulted for headache and emesis in the past 5 days, after à first medical imaging in another institution, he was addressed in Erasme Hospital.

Vital signs, biological results, and family history were not relevant.

Physical and neurological examination were within normal limits.

^{*} Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

^{*} Corresponding author.

E-mail address: federicodelucia507@gmail.com (F. De Lucia).

https://doi.org/10.1016/j.radcr.2023.08.049

^{1930-0433/© 2023} The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 – Brain enhanced CT in arterial phase: distal parietal branch of left sylvian artery (arrow) and nearby aneurismal vessel dilatation (arrowhead).

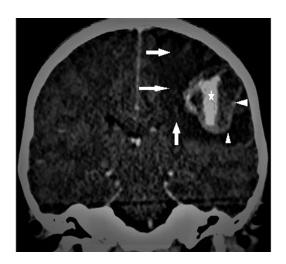


Fig. 2 – Brain enhanced CT in venous phase: perilesional oedema (arrows), peripheral enhancement (arrowheads) and an intralesional aneurysm (star).

The only known antecedent is an acute right otitis media complicated by transient facial paralysis.

Brain-CT with contrast (Figs. 1 and 2) showed an intraparenchymal left rolandic hemorrhagic lesion of about 3 cm, perilesional oedema, peripheral enhancement, an intralesional aneurysm, and possible cortical venous drainage.

A digital subtraction angiography was performed (Fig. 3). It discovered a left parietal arterio-venous shunt with cortical drainage and venous pseudoaneurysm. After selective catheterization of the left sylvian afferent branch, a trans arterial embolization with glue (1 ml Glubran + Lipiodol ultra-fluide [33%]) was performed to obtain complete occlusion of the arteriovenous shunt.



Fig. 3 – Angiogram showed venous pseudoaneurysm (arrow) in proximity of arteriovenous shunt and a small nidus (arrowhead).

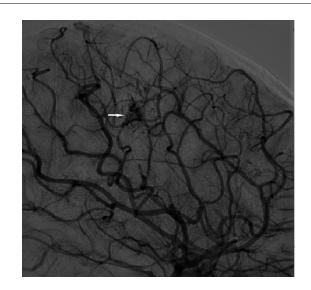


Fig. 4 – Forty-day control DSA, complete AVM obliteration and small scar residue (arrow). AVM, arteriovenous malformations; DSA, digital subtraction angiography.

The last control by digital subtraction angiography after 40 days of admission, showed complete occlusion of the AVM and a small scar residue (Fig. 4).

No symptoms or clinical signs were reported in the last records.

Discussion

The natural history of cerebral AVMs is not benign and the risk factor for hemorrhage are: prior hemorrhage, deep location, exclusively deep venous drainage and associated aneurysms [2]. Children have a higher risk of bleeding and rebleeding [1,3].

The ARUBA (a randomized trial of unruptured brain arteriovenous malformations) study showed that medical management alone is superior to interventional treatment for the prevention of death or symptomatic stroke in adults patients with an uncomplicated cerebral AVM [4] but this strategy is discussed especially for the pediatric population.

Microsurgery is the treatment of choice however is limited by the localization of the lesion in the deep and eloquent regions, for which endovascular embolization is the best primary treatment [5].

The choice of endovascular treatment depends on the number and size of feeder vessels, the size of the nidus and the team's experience. The aim is the complete embolization of feeder vessel and nidus.

Embolization materials including NBCA ethylene vinyl alcohol (ONYX: EVOH COPOLYMER), dehydrated alcohol and coils [5].

The levels of evidence in the literature remain low for endovascular approaches, studies with the use of HONIX shows good effectiveness of the method [6,7].

In this case for the deep location of AMV, the endovascular NBCA treatment has been performed.

NBCA allows a decrease in operating time and gives less imaging artifacts for MRI follow-ups and possible gamma knife treatment planning.

NBCA polymerizes in contact with an anionic environment.

It is mixed with ethiodol and tantalum to transport it, to delay the polymerization and make it radiopaque, the catheter flushes is performed with dextrose 5% (D5W) in water [8].

The major risks of NBCA are: the catheter adherence to the vessel, glue migration and too distal or too proximal embolization [8]. But the complications reported are low, Li et al. [9] observed after AVMs embolization with NBCA alone a severe morbidity of 0.7% and a mortality of 0.5%.

PH, temperatures, biological catalysts, glue formulation, blood flow and lysed red blood cells influenced polymerization rate of NBCA but differences between in vitro and in vivo are reported [10]. For this reason, a training simulator or adapted preparation formulas do not exist currently.

We observed a venous pseudoaneurysm secondary to the rupture of the AVM. The nidus was not clearly visible in the first radiological investigations due to compression by hemorrhage and may re-emerge later with hemorrhage resorption.

Conclusions

We report a case of successful NBCA AVM embolization in a 6-year-old child, guidelines for the use of this technique and tailored glue preparation formulas remain to be defined.

Patient consent

Written informed consent was obtained from the patient's parent.

REFERENCES

- Menovsky T, van Overbeeke JJ. Cerebral arteriovenous malformations in childhood: state of the art with special reference to treatment. Eur J Pediatr 1997;156(10):741–6. doi:10.1007/s004310050703.
- [2] Gross BA, Du R. Natural history of cerebral arteriovenous malformations: a meta-analysis. J Neurosurg 2013;118(2):437–43 Epub 2012 Nov 30. doi:10.3171/2012.10.JNS121280.
- [3] Qureshi AM, Muthusami P, Krings T, Amirabadi A, Radovanovic I, Dirks P, et al. Clinical and angioarchitectural features of hemorrhagic brain arterio-venous malformations in adults and children: contrasts and implications on outcome. Neurosurgery 2021;89(4):645–52. doi:10.1093/neuros/nyab251.
- [4] Mohr JP, Overbey JR, Hartmann A, Kummer RV, Al-Shahi Salman R, Kim H, et al. ARUBA co-investigators. Medical management with interventional therapy versus medical management alone for unruptured brain arteriovenous malformations (ARUBA): final follow-up of a multicentre, non-blinded, randomised controlled trial. Lancet Neurol 2020;19(7):573–81. doi:10.1016/S1474-4422(20)30181-2.5.
- [5] Millar C, Bissonnette B, Humphreys RP. Cerebral arteriovenous malformations in children. Can J Anaesth 1994;41(4):321–31. doi:10.1007/BF03009913.
- [6] Baharvahdat H, Blanc R, Fahed R, Smajda S, Ciccio G, Desilles JP, et al. Endovascular treatment for low-grade (Spetzler-Martin I-II) brain arteriovenous malformations. AJNR Am J Neuroradiol 2019;40(4):668–72 Epub 2019 Feb 21. doi:10.3174/ajnr.A5988.
- [7] Mounayer C, Hammami N, Piotin M, Spelle L, Benndorf G, Kessler I, et al. Nidal embolization of brain arteriovenous malformations using Onyx in 94 patients. AJNR Am J Neuroradiol 2007;28(3):518–23.
- [8] Vaidya S, Tozer KR, Chen J. An overview of embolic agents. Semin Intervent Radiol 2008;25(3):204–15. doi:10.1055/s-0028-1085930.
- [9] Li TL, Fang B, He XY, Duan CZ, Wang QJ, Zhao QP, et al. Complication analysis of 469 brain arteriovenous malformations treated with N-butyl cyanoacrylate. Interv Neuroradiol 2005;11(2):141–8. doi:10.1177/159101990501100204.
- [10] Wang BH, Boulton M, Lee DH, Pelz DM, Lownie SP. A systematic characterization of the factors influencing polymerization and dynamic behavior of n-butyl cyanoacrylate. J NeuroInterven Surg 2018;10:150–5.