

Case Report

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Contribution of flow MRI in the therapeutic management of middle face high flow arteriovenous malformation: A case report



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ABSTRACT

Backround: The radiosurgical management of high flow arteriovenous malformations (HFAVM) in the "destructive" stage requires a precise hemodynamic and anatomical assessment.

Patient and methods/case report: We report the case of a 32 years-old patient with a large ulcerated face HFAVM, on which Doppler ultrasound was impossible to perform. We show that, by combining 3D PCA and 2D CINE PC-MRI sequences, magnetic resonance imaging is capable to provide a complete morphometric and velocimetric mapping of the nidus and feeding arteries of the HFAVM. *Conclusion:* Although Doppler ultrasound is the reference examination in the HFAVM, Flow MRI without

contrast agent provides an advantageous alternative to assess vascular pathologies and choose the therapeutic strategy.

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1. Introduction

Initially developed for aortic studies [1], flow MRI without contrast agent [2–5] allows a quantitative and non-invasive exploration of vascular blood flows [6] and of the spinal cerebrospinal fluid flow [7,8]. The recent development and optimization of specific sequences dedicated to the analysis of arterial flows in the collateral branches of the external carotid artery [9,10] opened new perspectives in the exploration of vascular hemodynamics. It is then feasible to characterize both anatomical and hemodynamic features in millimetric vessels – smaller than 5 mm in diameter, whigh precision and a possible use in clinical routine.

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2. Case report

We report the case of a 32-year-old patient suffering from a high flow arterio-venous malformation (HFAVM) of the right middle face that was evolving since childhood (Fig. 1). Having refused a first treatment option a few years before, the patient reviewed his position upon the appearance of daily haemorrhagic episodes that had become hardly controllable by external compressions.

Facing the imminent risk of an uncontrollable cataclismatic hemorrhage, an emergency assessment of the HFAVM vascular territory was conducted, including angioTDM, angioMRI (T1, T2, T2*, Diffusion, Flair, TRICKS) and arteriography. It confirmed the presence of a voluminous facial arterio-venous malformation in a "destructive" phase (Schobinger stage III) [11], supplied by all the collateral branches of the right and left external carotid arteries as well as the right and left ophthalmic arteries. The TRICKS sequences showed a rapid "wash out" of the contrast product with significant venous arterialization. J. Bettoni et al./J Stomatol Oral Maxillofac Surg 120 (2019) 361-365



Fig. 1. Clinical photographs of the face (a) and profiles (b and c).

The initial strategy chosen for the emergency radiosurgical management consisted of the embolization of the principal arteriovenous nidus and the excision of the vascular malformation with immediate reconstruction by micro-anastomosis free flap of latissimus dorsi. We initially opted for an intra-operative surgical control of the ophthalmic arteries rather than an embolization, as we considered at this stage that embolization had a risk of iatrogenic blindness in case of a retrograde embolization of the central arteries of the retina, and that this risk was higher than the risk of intra-operative haemorrhage.

We, however, opted to evaluate the risk of per-operative hemorrhage prior to the surgery, but were restricted by the skin ulceration, which made any ultrasound examination impossible due to the risk of hemorrhage upon the passage of the probe. After having embolized the collateral branches of the right and left external carotid arteries by femoral access, it was decided that the patient would benefit from a flow MRI examination the day before the surgical procedure. The MRI examination was performed on a Philips 3 TESLA ACHIEVA DSTREAM (Best, Netherlands) research MRI, following our original protocol [9,10] developed as part of a research project that aimed at exploring the facial vascularization in physiological and pathological situations (clinical trial No. 2013-A00319-36). Its originality is to associate 3-Dimensional Phase Contrast Angiographic (3D PCA) sequences, along with 2-Dimensional hemodynamic Phase Contrast-MRI (2D MRI-PC) sequences synchronized on the heart rate (Table 1). This combination allows to first identify the arteries of interest and position imaging planes of interest perpendicularly to the vessels, and then quantify the flow field within these planes. The objective of the MRI measurements were to quantify the flow in the main arteries related to the arteriovenous malformation in order to estimate the per-operative hemorrhagic risk.

The analysis of the 2D CINE PC-MRI sequences was performed in post-processing using Flow software developed by Balédent et al. [7] which is based on a semi-automatic segmentation using a flow gradient vector algorithm [12,14]. Data processing revealed the persistence of vascularization of the arteriovenous malformation mainly through the right ophthalmic artery and the left superficial temporal artery with respective flow rates of 371 mL/ min (Fig. 2) and 157 mL/min (Fig. 3).

These flow conditions indicated that there was a very high risk of haemorrhage in the right ophthalmic and left superficial temporal arteries that engaged the vital prognostic of the patient, and that this risk outweighed the functional risk of blindness associated with embolization. The risk-benefit ratio between embolization and surgical control being the reverse to what was initially believed, we postponed the surgery and a second embolization session was performed to control endovascularly

Table 1

Parameters of the 3D PCA and 2D CINE PC-MRI sequences.

	3D PCA sequence 32 head coil	2D CINE-PC sequence 32 head coil	2D CINE-PC sequence 32 head coil
Objectif	Vascular mapping of the high flow malformation	Flow quantification of the right ophtalmic artery	Flow quantification of the left superficial temporal artery
TE/TR (ms)	3/5	13/8	13/8
Spatial resolution (mm ³)	$0.5 \times 0.5 \times 1$	$0.5 \times 0.5 \times 3$	$0.15 \times 0.15 \times 2$
FOV (mm ²)	240×240	120×120	50×50
Encoding velocities (cm/s)	50 and 100	110	60
Number of frames per cardiac cycle	1	16	16



Fig. 2. Hemodynamic analysis of the right ophthalmic artery: a: positioning of the 2D CINE PC-MRI sequence (encoding velocity: 110 cm/s) on the 3D PCA acquisition (encoding velocity: 100 cm/s); b: Semi-automatic segmentation, variation of the vessel flow rate over one cardiac cycle (average flow of 371 mL/min).

the right ophtalmic and left superficial temporal arteries. After having confirmed the absence of post-embolization blindness, the patient underwent extensive surgical excision of the arteriovenous malformation with no hemorrhagic complication and with immediate reconstruction by micro-anastomosis free flap of latissimus dorsi (Fig. 4).

3. Discussion

Arteriovenous malformations are congenital pathologies characterized by the presence of abnormal vessels (arteries, veins and capillaries) forming arteriovenous shunts, also called "nidus", with a precocious and arterialized venous return. Their diagnosis requires a



Fig. 3. Hemodynamic analysis of the left superficial temporal artery: a: Positioning of the 2D CINE PC-MRI sequence (encoding velocity: 50 cm/s) on the 3D PCA acquisition (encoding velocity: 60 cm/s); b: semi-automatic segmentation, variation of the vessel flow rate over one cardiac cycle (average flow of 157 mL/min).



Fig. 4. a: pre-operative and b: intra-operative photographs with resection of arteriovenous malformation and c: after reconstruction by latissimus dorsi free flap.

fine and delicate clinical examination and must be suspected in the presence of any hot pulsatile skin lesion with or without a thrill. Because of the risk of functional and aesthetic sequelae upon surgical management, the International Society for the Vascular Anomalies (ISSVA) recommends to correlate therapeutic indications with the evolution and aggressiveness of the malformation [11-13] and to restrict an interventional attitude in the case of destructive lesions (stage III). The assessment of the arteriovenous malformation thus requires anatomical and hemodynamic explorations of the nidus and of its related vessels, using either Doppler echography (the goldstandard), angioTDM, angioMRI or arteriography. By reporting here a first example in using flow MRI for the management of a HFAVM, we enlarge the diagnostic tools panel for vascular explorations, with a non-invasive and operator-independent examination that combines anatomical and hemodynamic exploration of the main vessels related to the lesion.

The phase contrast sequences presently used were initially developed to explore the external carotid artery collateral branches, in order to help the surgeon choosing the best recipient vessels in head and neck free flaps reconstruction [9,10]. The high-precision sequences were developed to image and quantify blood flow in millimetric vessels. They were validated ex vivo, with a measurement accuracy of 97% [14] and in vivo proving the feasibility of an anatomical [9] and hemodynamic [10] exploration of small caliber vessels with a non-invasive and operator-independent technique.

In the patient case reported, it is remarkable to observe the importance of having both anatomical and hemodynamic data available when managing a large HFAVM. Indeed, the quantification in the patient's ophthalmic artery of a flow rate 14 times greater than the normal values [15], and three times higher than in a normal external carotid [10], forced us to revise our therapeutical approach, relying on a very high risk of hemorrhage. We thus decided to reconsider our position towards the functional risk of blindness associated with embolization, as it became acceptable in regards to the intra-operative haemorrhage risk, which involved the vital prognosis of the patient.

Flow MRI should be conceived as a complement to the standard arteriovenous malformation assessment. It seems particularly recommended in the case of haemorrhagic or voluminous lesions, because the identification of the main vascular axes on the 3D PCA sequences allows the surgeon to better plan lesion removal and identify the main "hot spots" with high haemorrhagic risk.

In the future, it seems conceivable that flow MRI could become the reference imaging technique for the mornitoring of arteriovenous malformations owing to its non-invasive nature and operator-independence. It has the great advantage to provide the possibility of extracting complete 3D morphometric information on the lesional volume from 3D PCA sequences as well as quantitative information on the flow in the main related vessels from the CINE PC-MRI measurements. Technically, it is important to perform several 3D PCA sequences with different encoding speeds in order to choose the appropriate encoding speed for the hemodynamic sequences. Indeed, in case of inappropriate encoding speed, too much aliasing affects the accuracy of the 2D CINE PC-MRI acquisitions, as it quickly exceeds the software capabilities of correction.

4. Conclusion

Phase contrast MRI sequences dedicated to the exploration of infracentimetric vascular structures (typically below 5 mm in diameter) constitute a precise reproducible and non-invasive alternative to Doppler ultrasound in the assessment of vascular malformations. It advantageously provides 3D anatomical and hemodynamic data within the intralesional vessels, which allows to precisely evaluate the risk-benefit ratio of the various therapeutic modalities and optimally prepare complex interventions, thus reducing the risk of vital prognosis. Although its generalization may seem difficult at present, PC-MRI appears as a valuable imaging approach to favor in case of complex vasculatures and when conventional imaging is not possible.

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Disclosure of interest

The authors declare that they have no competing interest.

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References

- [1] Herment A, Lefort M, Kachenoura N, De Cesare A, Taviani V, Graves MJ, et al. Automated estimation of aortic strain from steady-state free-precession and phase contrast PR images. Magn Reson Med 2011;65:986–93. <u>http:// dx.doi.org/10.1002/mm.22678</u>.
- [2] Nayler GL, Firmin DN, Longmore DB. Blood flow imaging by cine magnetic resonance. J Comput Assist Tomogr 1986;10:715–22 [PMID: 3528245].
- [3] Pelc NJ, Herlkens RJ, Shimakawa A, Enzmann DR. Phase contrast cine magnetic resonance imaging. Magn Reson Q 1991;7:229–54.
- [4] Rebergen SA, van der Wall EE, Doombos J, de Roos A. Magnetic resonance measurement of velocity and flow: technique, validation, and cardiovascular applications. Am Heart J 1993;126:1439–56 [PMID: 8249802].
- [5] Pelc NJ, Sommer FG, Li KC, Brosnan TJ, Herlkens RJ, Enzmann DR. Quantitative magnetic resonance flow imaging. Magn Reson Q 1994;10:125–47 [PMID: 7811608].
- [6] Nayak KS, Nielsan JF, Bernstein MA, Markl M, Gatehouse DM, Botnar R, et al. Cardiovascular magnetic resonance phase contrast imaging. J Cardiovasc Magn Reson 2015;17:71. <u>http://dx.doi.org/10.1186/s12968-015-0172-7</u>.
- [7] Balédent O, Henry-Feugeas MC, Idy-Peretti I. Cerebrospinal fluid dynamics and relation with blood flow: a magnetic resonance study with semiautomated cerebrospinal fluid segmentation. Invest Radiol 2001;36:368–77.
- [8] Saliou G, Balédent O, Lehmann P, Paradot G, Gondry-Jouet C, Bouzerar R, et al. Acute CSF changes in the mesencephalon acqueduct after subarachnoid hemorrhage as measured by PC-MRI. J Neuroradiol 2009;36:41–7. <u>http://</u> <u>dx.doi.org/10.1016/j.neurad.2008.07.004</u>.

- [9] Bettoni J, Pagé G, Salsac AV, Constans JM, Testelin S, Devauchelle B, et al. 3T noninjected phase-contrast MRI sequences for mapping of the external carotid branches: In vivo radio-anatomical pilot study for feasibility analysis. J Craniomaxillofac Surg 2018;46:98–106. <u>http://dx.doi.org/10.1016/j.jcms.2017.09.005</u>.
- [10] Bettoni J, Pagé G, Salsac AV, Constans JM, Testelin S, Devauchelle B, et al. Quantitative assessment of the flow distribution in the branches of the external carotid by non-injected flow MRI. Dentomaxillofac Radiol 2018. http://dx.doi.org/10.1259/dmfr.20180153.
- [11] Kohout MP, Hansen M, Pribaz JJ, Milliken JB. Arteriovenous malformations of the head and neck: natural history and management. Plast Reconstr Surg 1998;102:643–54 [PMID: 9727427].
- [12] Xu C, Prince JL. Snakes, shapes, and gradient vector flow. IEEE Trans Image Process 1998;7:359–69. <u>http://dx.doi.org/10.1109/83.661186</u>.
- [13] Wassef M, Blei F, Adams D, Alomari A, Baselga E, Berenstein A, et al. Vascular anomalies classification: recommendations from the international society for the study of vascular anomalies. Pediatrics 2015;136:e203–14. <u>http:// dx.doi.org/10.1542/peds.2014-3673</u>.
- [14] Pagé G, Bettoni J, Salsac AV, Balédent O. Influence of principal component analysis acceleration factor on velocity measurement in 2D and 4D PC-MRI. MAGMA 2018. <u>http://dx.doi.org/10.1007/s10334-018-0673-0</u>.
- [15] Promelle V, Daouk J, Bouzerar R, Jany B, Milazzo S, Balédent O. Ocular blood flow and cerebrospinal pressure in glaucoma. Acta Radiol Open 2016;5. <u>http://</u> <u>dx.doi.org/10.1177/2058460115624275</u> [2058460115624275].