



Review article

Feeding artery aneurysms associated with large meningiomas: case report and review of the literature[☆]Kyriakos Papadimitriou^a, Alda Rocca^a, Vincent Dunet^b, Roy Thomas Daniel^{a,*}^a Department of Clinical Neurosciences, Service of Neurosurgery Lausanne University Hospital and University of Lausanne, Switzerland^b Department of Diagnostic and Interventional Radiology, Lausanne University Hospital and University of Lausanne, Switzerland

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ABSTRACT

Though brain tumors and intracranial aneurysms co-exist, the occurrence of feeding artery aneurysms with meningiomas are unusual. We describe here a large falcine meningioma that was associated with three feeding artery aneurysms of the anterior circulation. These aneurysms can be treated either by pre-operative endovascular therapy or during the meningioma surgery. The management strategy for these patients will depend on the clinical presentation, morphology and localization of these aneurysms with respect to the tumor.

1. Introduction

Co-existence of brain tumors with intracranial aneurysms is rare, first reported by Arieti in 1944 and occurs with an incidence of 0.3–1% [1, 2], though this association is now being increasingly recognized. The tumors that are known to have this association are meningiomas, pituitary adenomas, and malignant gliomas [1, 2,3, 4, 5]. Of these, meningiomas are more commonly associated with the formation of an intracranial aneurysm [3]. Though coincidental associations are known between these tumors and aneurysm, the true incidence of flow related aneurysms in the presence of a vascular tumor is likely to be very low and not well established.

2. Case presentation

A 55-year-old female patient with no known prior medical problems, presented with one episode of a generalized tonic-clonic seizure. There were no neurological deficits. A magnetic resonance imaging (MRI) revealed a large, highly vascular, extra-axial tumor arising from the anterior part of the falx cerebri. The superior sagittal sinus was patent, and the differential diagnosis considered was meningioma, hemangiopericytoma and solitary fibrous dysplasia (Figure 1 A,B,C). Despite the presence of a large tumor, there was dilated superficial subarachnoid

spaces possibly due to frontal lobe atrophy. On magnetic resonance angiography (MRA), three feeding artery aneurysms were detected in relation to the left pericallosal artery (5 × 4 mm; dysplastic), the left anterior communicating artery (4 × 5 mm saccular) and the right frontopolar artery (5 × 5 mm fusiform) (Figure 1F). In view of the large tumor size, we performed a pre-operative embolization of all three aneurysms (Figure 1 D,E). The tumor was resected through an interhemispheric approach, without any complications. The pathological report showed a meningothelial meningioma OMS grade I. She had a normal neurological examination at discharge from hospital. The neuropsychological examination also was normal. At the follow up visit at three months, she had no neurological symptoms or deficits. The angio MRI did not show any residual aneurysms and demonstrated the remodeling of the dysplastic feeding arteries (Figure 1 G,H).

Informed consent was obtained from the individual participant included in the study.

3. Discussion

The co-existence of intracranial aneurysms and brain tumors are reported to be as low as 0.3% in a series of 23,876 brain tumor patients [4]. Recent advances and availability of imaging techniques has led to an increase in detection of intracranial aneurysms of up to 5% in patients

[☆] This case hasn't been published or presented in any journal or conference.

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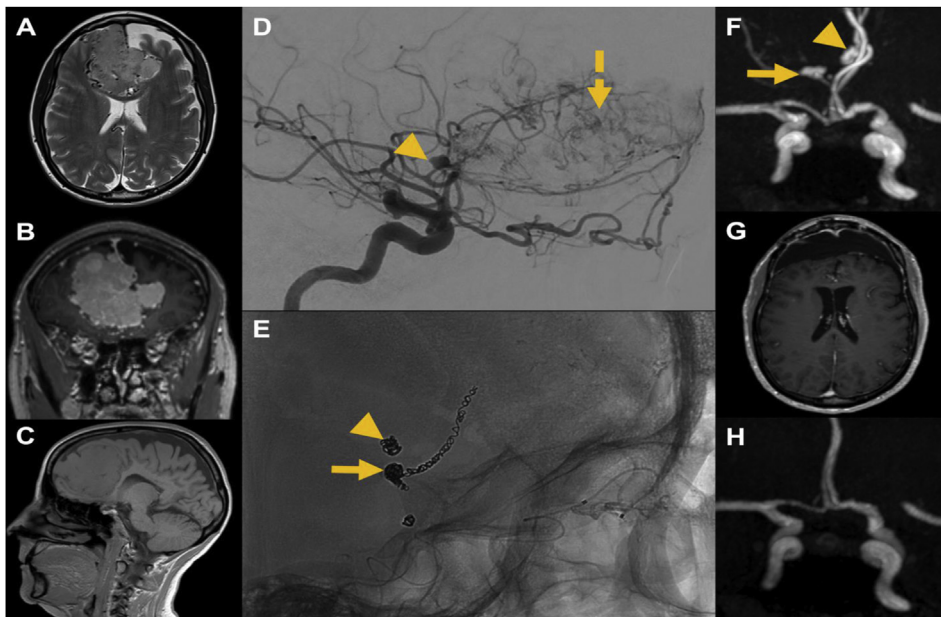


Figure 1. Preoperative and follow-up imaging. The pre-operative brain MRI (A–C) showed a large meningioma arising from the anterior part of the falx cerebri. The tumor was extending on to both sides of the falx with significant brain deformation. The tumor had a marked contrast uptake and was associated with multiple dilated and tortuous vessels at its periphery. The TOF (time-of-flight) images showed three high flow aneurysms related to the feeding arteries namely the left callosal-marginal artery (arrow head), right fronto-polar artery (arrow) and anterior communicating artery (F). The digital subtraction angiography (DSA) showed the presence of dysplastic feeding arteries to the tumor (D, dashed arrow) in addition to the aneurysms. Embolization of the three aneurysms and one of the feeding arteries was performed (E). The MRI contrast enhanced images, 3 months after surgery demonstrates the tumor excision (G). The angiographic sequences (H) showed no residual aneurysms and demonstrated the remodeling of the dysplastic arteries.

diagnosed with brain tumors [6]. However, the association of aneurysms and meningiomas seems to be higher [5, 6,7]. The exact pathologic mechanism underlying the coexistence of these two pathologies remains unclear. Their formation is postulated to be the result of a complex interaction of flow-related factors, hormonal influence and genetic predisposition. Therefore, several mechanisms of aneurysm formation associated with meningiomas have been proposed [2]. Tumor volume appears to have an effect on the formation of intracranial aneurysms. Increase of blood pressure proportionally with intracranial pressure in order to maintain a constant cerebral perfusion pressure, may lead to an increase of hemodynamic stress around growing meningiomas.

Hemodynamic stress induces remodeling and degeneration of internal elastic lamina [7].

Moreover, an increase in directional blood flow in response to angiogenic factors to meet blood supply of highly vascular meningiomas might ensue in abnormal stress on major feeding vessels and is suggested as another possible source of hemodynamic stress [4,7, 8]. These hypotheses can be supported by the report of Tachikawa et al on an anterior ethmoidal artery aneurysm, associated with an olfactory groove meningioma, in a 51 year old male [8]. Following resection of tumor, a post-operative angiography demonstrated the complete disappearance of the aneurysm.

Table 1. Reported cases of meningiomas with flow related aneurysms.

Author/Year	Age/Sex	Meningioma Location	Aneurysm Location	Treatment	Outcome
Arseni 1973 [14]	37/M	Olfactory groove	AcomA	No pre-op embolisation Tumor resection	Patient alive
Kandel 1986 [9]	7/F	Left fronto-temporale	Left MCA	Tumor and aneurysm resection	Patient asymptomatic
O'Neil 1995 [20]	82/F	Convexity meningioma	Middle meningeal artery	Pre-operative embolization and tumor resection	NA
Tancioni 1998 [22]	48/F	Left fronto-temporal	Left MCA	Pre-operative embolization and tumor resection	Improvement of preoperative symptoms
Donelc 1998 [11]	50/M	<i>Tuberculum sellae</i>	Calcified AComA	No pre-op embolisation Tumor resection	Patient was discharge home with improved visual acuity
Ongino 1999 [21]	70/F	<i>Tuberculum sellae</i>	Ruptured AComA	Aneurysm clipping and tumor resection	Patient was discharge home
Lama 2000 [18]	69/F	Right pterional	Right middle meningeal artery	Pre-operative embolization and tumor resection	NA
Tachikawa 2002 [8]	51/F	Olfactory groove	Anterior ethmoidal	No pre op embolization Tumor resection	Spontaneous resolution of aneurysm
Javadpour 2004 [17]	61/F	Suprasellar	AComA	Pre-operative embolization and tumor resection	Patient was discharge home
Fischer 2008 [3]	44/M	Left Sphenoid wing	ICA	Preoperative embolization/ clipping and tumor resection	CNIII palsy
Maekawa 2009 [19]	72/F	Left frontal convexity	Left middle meningeal artery	Pre-operative aneurysm embolization and tumor resection	Satisfactory postoperative course
Dumitrescu 2011 [15]	64/M	<i>Tuberculum sellae</i>	AComA	Aneurysm clipping and tumor resection	Complete anosmia
Zhong 2013 [6]	49/M	Falx/planum	ACA	Aneurysm clipping and tumor resection	Patient alive

In 1998, Kandel et al introduced another hypothesis in a case report of a saccular aneurysm of the middle cerebral artery that was enclosed in a frontotemporal meningioma [9]. Microscopic examination demonstrated that the tumor was attached to the arterial adventitia, and therefore the authors ascribed the development of the aneurysm to the damage to the arterial wall. However, this hypothesis cannot explain the formation of aneurysms not directly localized near tumors.

An endocrinological-mediated mechanism has been proposed in some papers, which presented a possible influence of estrogens in the development of both meningiomas and aneurysms [10, 11]. Other studies focused on the genetic aspects have shown that some gene mutations were common for both pathologies, such as 1p36.2-p34, 11q13 and 17p13.1 [12,13].

Based on the hypothesis that aneurysm formation is, at least partially, due to hemodynamic stress on the feeding arteries of a meningioma, we performed a review of the English literature including only cases of aneurysms localized selectively on a feeding artery of a meningioma (Table 1). A total of 13 cases were identified [3, 6, 8, 9,14, 15, 16, 17, 18, 19, 20, 21, 22]. Eight (61.5%) patients were females. The most common localization was the anterior circulation (61.5%), particularly the anterior communicating artery (38.5%). Three cases of aneurysm on the external carotid artery circulation (middle meningeal artery) were also documented. Only one of these cases presented with a subarachnoid hemorrhage (SAH) related to the rupture of a feeding artery aneurysm [21].

In patients with coexisting intracerebral aneurysms and meningiomas, the lesion that is symptomatic should be treated at first [5]. When subarachnoid hemorrhage is the primary presentation, the aneurysm should be treated before tumor surgery. However, in cases of an unruptured aneurysm, the options of treatment either by pre-tumor excision embolization or by direct clipping during tumor surgery, both exist [4, 6, 7].

In the review of the literature of these 13 cases, 6 patients were treated by pre-operative embolization of the aneurysm, followed by surgical resection of the meningioma (Table 1). Concomitant treatment of the tumor and the aneurysm was achieved in 4 patients. Three patients did not have any treatment for the aneurysms and follow up imaging showed resolution or stability of the aneurysms. All authors have emphasized the importance of MRA and/or DSA in patients with large meningiomas, in order to avoid catastrophic intra-operative surprises. Notably, all these case reports dealt with large sized meningiomas with marked vascularity. This gives credence to the hypothesis of hemodynamic stress as a causative factor for these aneurysms in relation to a feeding artery. Our patient had multiple feeding artery aneurysms in addition to other areas of dysplastic dilatation of the feeding arteries. The pre-operative treatment of the aneurysms allowed a safe dissection of the tumor capsule. The residual dysplasias of the vessels had subsided on follow up imaging. The formation of these dysplastic aneurysms and feeding artery aneurysms are most likely to be due to the hypervascular meningioma, which had behaved like an arteriovenous shunt. This had also interestingly induced a lobar atrophy akin to the “steal syndrome” seen in arteriovenous malformations. The fact that the elimination of the meningioma allowed for reversal of the vasculopathy in the feeding arteries, lends further credence to the etiopathogenesis of the vascular malformations in this patient.

4. Conclusion

The coexistence of large hypervascular meningiomas and feeding arteries aneurysms though rare, needs to be considered in the pre-operative radiological evaluation of these patients. The mechanism of formation of such aneurysms is likely to be related to the hemodynamic

stresses on these feeding arteries caused by the increased blood flow to the meningioma. The treatment strategy should be tailored depending on the clinical presentation and anatomical relation of the aneurysms to the tumor.

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The authors declare no conflict of interest.

Additional information

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References

- [1] A. Hashiguchi, M. Morioka, H. Ichimura, C. Mimata, J. Kuratsu, Glioblastoma with an intratumoral feeding-artery aneurysm, *Clin. Neurol. Neurosurg.* 109 (2007) 302–304.
- [2] J.-S.L. Jen-Lung Chen, Edgar D. Su, Ming-Tsung Chuang, c Hsing-Hong Chen, Coexistence of anterior communicating artery aneurysm and tuberculum sellae meningioma, *Formosan Journal of Surgery* 48 (2015) 226–229.
- [3] B.R. Fischer, S. Palkovic, M. Holling, T. Niederstadt, A. Jeibmann, H. Wassmann, Coexistence of cerebral aneurysm and meningioma—pure accident? *Clin. Neurol. Neurosurg.* 111 (2009) 647–654.
- [4] H.W. Pia, S. Obrador, J.G. Martin, Association of brain tumours and arterial intracranial aneurysms, *Acta Neurochir.* 27 (1972) 189–204.
- [5] N. Takeda, M. Nishihara, S. Yamanishi, K. Kidoguchi, K. Hashimoto, Strategy for patients with co-existence of meningioma and intracerebral aneurysm, especially unruptured aneurysm (-seven cases and review of the literature-), *J. Clin. Neurosci. : offl. J. Neurosurgical Soc. Australasia* 45 (2017) 236–242.
- [6] Z. Zhong, Y. Sun, D. Lin, Q. Sun, L. Bian, Surgical treatment of brain tumor coexisted with intracranial aneurysm—case series and review of the literature, *Neurosurg. Rev.* 36 (2013) 645–656, discussion 656.
- [7] Y.H. Kim, Y.J. Lee, J.H. Han, S. Ahn, J. Lee, J.H. Kim, B.S. Choi, J.S. Bang, C.Y. Kim, G. Hwang, O.K. Kwon, C.W. Oh, Association of intracranial aneurysms and meningiomas: a case-control study, *J. Neurosurg.* 123 (2015) 357–361.
- [8] T. Tachikawa, J. Adachi, R. Nishikawa, M. Matsutani, An anterior ethmoidal artery aneurysm associated with an olfactory groove meningioma. Case illustration, *J. Neurosurg.* 97 (2002) 1479.
- [9] E. Kandel, I. Ludkovskaya, N. Dobjansky, Aneurysm inside meningioma, Case report. *Acta neurochirurgica* 81 (1986) 72–76.
- [10] C.G. Harrod, H.H. Batjer, B.R. Bendok, Deficiencies in estrogen-mediated regulation of cerebrovascular homeostasis may contribute to an increased risk of cerebral aneurysm pathogenesis and rupture in menopausal and postmenopausal women, *Med. Hypotheses* 66 (2006) 736–756.
- [11] S. Pravdenkova, O. Al-Mefty, J. Sawyer, M. Husain, Progesterone and estrogen receptors: opposing prognostic indicators in meningiomas, *J. Neurosurg.* 105 (2006) 163–173.
- [12] B.V. Nahed, M. Bydon, A.K. Ozturk, K. Bilguvar, F. Bayrakli, M. Gunel, Genetics of intracranial aneurysms, *Neurosurgery* 60 (2007) 213–225, discussion 225–216.
- [13] M. Simon, J.P. Bostrom, C. Hartmann, Molecular genetics of meningiomas: from basic research to potential clinical applications, *Neurosurgery* 60 (2007) 787–798, discussion 787–798.
- [14] C. Arseni, M. Maretsis, Meningioma associated with intracranial aneurysm or cerebral thrombosis, *Neurochirurgia* 16 (1973) 131–137.
- [15] M.R.G. Bogdan Constantin Dumitrescu, Giant tuberculum sellae meningioma with unruptured anterior communicating artery aneurysm encased, Case report and review of the literature *Romanian Neurosurgery* 4 (2011) 533–540.
- [16] V.V. Dolenc, R. Pregelj, S. Slokan, M. Skrbec, Anterior communicating artery aneurysm associated with tuberculum sellae meningioma—case report, *Neurol. Med.-Chir.* 38 (1998) 485–488.

- [17] M. Javadpour, A.D. Khan, M.D. Jenkinson, P.M. Foy, H.C. Nahser, Cerebral aneurysm associated with an intracranial tumour: staged endovascular and surgical treatment in two cases, *Br. J. Neurosurg.* 18 (2004) 280–284.
- [18] M. Lama, C. Mottolise, Middle meningeal artery aneurysm associated with meningioma, *J. Neurosurg. Sci.* 44 (2000) 39–41.
- [19] H. Maekawa, M. Tanaka, H. Hadeishi, Middle meningeal artery aneurysm associated with meningioma, *Acta Neurochir.* 151 (2009) 1167–1168.
- [20] O.R. O’Neill, S.L. Barnwell, D.J. Silver, Middle meningeal artery aneurysm associated with meningioma: case report, *Neurosurgery* 36 (1995) 396–398.
- [21] M. Ogino, M. Nakatsukasa, T. Nakagawa, I. Murase, Ruptured anterior communicating artery aneurysm encased in a tuberculum sellae meningioma. Case report, *J. Neurosurg.* 91 (1999) 871–874.
- [22] F. Tancioni, M.G. Egitto, F. Tartara, Aneurysm occurring within a meningioma: case report, *Br. J. Neurosurg.* 12 (1998) 588–591.