Coexistence of arteriovenous malformation with nonfunctioning pituitary adenoma

Sir,

Concomitant brain pathologies have been reported in the literature.^[1,2] Cerebral aneurysms concomitant with pituitary adenomas have been previously reported.^[2,3] However, arteriovenous malformations (AVM) concomitant with pituitary adenomas are very rare.^[4-6] We present a patient, who has been successfully treated with gamma-knife radiosurgery (GKR) for AVM concomitant with nonfunctioning pituitary adenoma. We also discuss the case with the current literature. To our knowledge, this is the first patient who has been successfully treated with GKR for AVM concomitant with pituitary adenoma.

A 28-year-old female, who had cerebral AVM concomitant with pituitary adenoma, was referred to our GKR unit in 2006. At the time of referral, she had 5-time-embolized AVM in the left fronto-parietal lobe and residual pituitary adenoma from the surgery in 2001. Her neurological and ophthalmological examinations were intact. Her hormone levels were unremarkable. The pituitary adenoma was located in close proximity to the left internal carotid artery [Figure 1a]. GKR was planned for both pathologies. The doses of radiation for the pituitary adenoma and the AVM nidus were 18 Gy defined to the 50% isodose line and 22 Gy defined to the 50% isodose line, respectively [Figures 1a and 2a]. The patient was followed-up with brain magnetic resonance imaging (MRI) every 3 months in the 1st year and every 6 months in the following 2 years. At the end of the 3rd year, both brain MRI and brain digital subtraction angiography were performed. The pituitary adenoma had no progression while a residual AVM nidus was still present [Figures 1b and 2b]. We planned a second-stage GKR for the residual AVM. The dose of radiation for the AVM nidus was 20 Gy defined to the 50% isodose. The AVM nidus became obliterated 4.5 years after the second-stage GKR [Figure 2c]. The patient's pituitary hormone levels were unremarkable at the last follow-up. She had no new seizures under anti-epileptic drug regimens during the course of the treatment.

Cerebral vascular lesions may co-exist with brain tumors.^[1,2] Prevalence of pituitary adenomas is 16.7%, whereas prevalence of AVMs is 18 in 100 000 adults.^[7,8] Incidence of cerebral aneurysms in patients with brain tumors is 0.2–0.7% while the incidence of AVMs in the same patient population is only 0.1%.^[9-12] Cerebral aneurysms were reported in 0.04–7.4% of patients with pituitary adenoma.^[3,5,6] Since the first case reported by Licata *et al.* in 1986, only four cases have been reported to have AVM concomitant with pituitary adenoma [Table 1].^[4,9,13,14]

Meta-analyses depicted the high success rates of GKR in both AVMs and pituitary adenomas, particularly when the surgery is not the best choice of treatment for patients with complicated pathologies. It has been reported that 42.3–89% of nonfunctioning pituitary adenomas responded to GKR, functioning adenomas responded to GKR in different ranges based on the hormone (s)



Figure 1: Residual pituitary adenoma is located in close proximity to the left internal carotid artery (a) follow-up magnetic resonance imaging 3 years after the gamma-knife radiosurgery (b)



Figure 2: Residual arteriovenous malformation is apparent in 2006 (a), nidus size diminished at the 3rd-year follow-up (b). Nidus has been obliterated 7.5 years after the first gamma-knife radiosurgery (c)

Authors	Age/sex	Signs and symptoms	AVM location	Treatment	Follow-up
Licata <i>et al.</i> ^[9]	25/male	Headache, drowsiness	Right medial temporal lobe	Surgery (partial removal of the adenoma and total resection of AVM)	No deficit, 1-year
Furuya <i>et al.</i> ^[4]	41/female	Headache	Ventral pons	Total resection of the adenoma, AVM disappeared spontaneously after hemorrhage	Right internal auditory dysfunction, 3 months
Cossu <i>et al.</i> * ^[13]	N/A	N/A	N/A (dural AVM supplied by posterior branches of the middle meningeal artery and drained by transverse sinus)	N/A	N/A
Xu <i>et al.</i> ^[14]	41/male	Headache, visual disturbances	Right anterior middle frontal lobe	Partial removal of the adenoma and total resection of AVM, radiotherapy for the residual adenoma	No deficit, 6 years
Present case	28/female	Asymptomatic (previously had been diagnosed after seizure)	Left fronto-parietal lobe	Gamma-knife radiosurgery for both pathologies, 2 nd -stage gamma-knife radiosurgery to the residual AVM bed in the 3 rd year	No deficit, 7.5 years

	Table	1:	AVM	concomitant	with	pituitary	adenoma	cases	in	the	literature
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*only abstract could be retrieved from Pubmed (PMID: 8160556). AVM: Arteriovenous malformation, N/A: Not applicable

secreted by the ademonas (prolactinoma = 17.4-50%; acromegaly = 36.9-82%; Cushing's disease = 27.9-54%).^[15] On the other hand, complete obliteration rate of cerebral AVMs is 96% (range = 0-100%) with microsurgery, 38% (range = 0-75%) with stereotactic radiosurgery, and 13% (range = 0-94%) with embolization.^[16] Even though success rate of GKR in AVMs after embolization is lower than that of GKR alone (41% vs. 59%), complication rates of both approaches are somehow similar (hemorrhage = 7.3% vs. 5.6%; permanent neurological deficit = 3.3% vs. 3.4%).^[17]

Previously, Licata et al. and Xu et al. reported the successful resection of both pituitary adenoma and AVM.^[9,14] Their patients had had no previous treatments for pituitary adenoma and AVM. Furuya et al., reported a patient with spontaneous disappearance of AVM after hemorrhage.^[4] They detected the AVM before the pituitary surgery. Then, they operated the patient using transsphenoidal route, after which left hemiparesis, right deafness, and nystagmus developed. They detected hematoma in the previous location of the dural AVM.^[4] Remote hemorrhage of AVM after deep brain stimulation surgery has been reported in the literature.^[18] In a meta-analysis about residual nonfunctioning pituitary adenomas, regrowth was detected in 12% patients with undetectable amount of residual adenoma while the incidence rises to 46% in detectable amounts of residual tumor.^[19] Our patient had been operated for pituitary adenoma before she was referred to us. Her residual lesion was in close proximity to the left internal carotid artery. As she had been previously operated, the adenoma was suspected to have a fibrotic capsule. Her AVM had been previously embolized for 5 times. Due to multiple previous invasive

attempts for both pathologies, and risk of hemorrhage during and/or after the pituitary surgery we planned GKR for both pathologies. The pituitary adenoma had no progression, and the AVM diminished in size after 3 years of follow-up. Our patient received a second-stage GKR for the AVM bed. She had no new neurologic deficits and/or other complications in follow-ups. The AVM nodule obliterated 7.5 years after the first GKR.

In conclusion, AVM concomitant with pituitary adenoma is a very rare condition. Treatment should be tailored to the patient, based on the location of AVM and the type of adenoma, previous treatment approaches, and related co-morbidities. To our knowledge, this is the first case of AVM concomitant with nonfunctioning pituitary adenoma both treated successfully with GKR.

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