Journal of Clinical Neuroscience xxx (2017) xxx–xxx

Contents lists available at ScienceDirect

Journal of Clinical Neuroscience

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

Clinical commentary

Management of a complex intracranial arteriovenous malformation with gamma knife radiosurgery: A case report with review of literature

Shyamal C. Bir [a], Subhas K. Konar [c], Devi P. Patra [a], Tanmoy K. Maiti [a], Alireza Minagar [b], Anil Nanda [a,*]

[a] Department of Neurosurgery, LSU Health-Shreveport, 1501 Kings Highway, Shreveport, LA 71130-3932, USA
[b] Department of Neurology, LSU Health-Shreveport, 1501 Kings Highway, Shreveport, LA 71130-3932, USA
[c] Department of Neurosurgery, NIMHANS, Bangalore, India

ARTICLE INFO

Article history:
Received 28 June 2017
Accepted 23 October 2017
Available online xxxx

Keywords:
Microsurgery
Gamma knife radiosurgery
Outcomes and complications
Predictors

ABSTRACT

The risks and benefits of arteriovenous malformation (AVM) treatment should be considered cautiously in each patient since management strategy of it depends on various factors including age of the patient, location and volume of AVM and presence of other vascular abnormalities. Current management options of AVM include observation, endovascular embolization, radiosurgery and microsurgical resection or in combination of any two of the above procedures. Here, we have discussed a case of intracranial AVM with radiation induced early cyst formation, and performed a literature review to determine the optimum treatment of complex intracranial AVM. Standard search strategies were performed in PubMed/Medline using appropriate terms such as "intracranial arteriovenous malformation" radiosurgery, embolization and microsurgical resection as well as medical subject headings. The particular case in this study was retrospectively reviewed. Literature review revealed that the mean marginal radiation dose used by the different authors was 19 Gy, cysts were developed in 3.6% patients, the average time to form cyst was 6.6 years, average volume of cyst was 6.7 ml and maximum cysts were removed by resection. In our case, the cyst was developed 2.5 years after radiosurgery. Radiation induced cyst formation is a delayed complication of AVM management. However, cyst formation in this case was comparatively earlier in our case. Therefore, continuous follow-up after radiosurgery is required for early detection of cyst formation. In addition, the review revealed that embolization before radiosurgery was a poor strategy.

1. Introduction

Natural history of a large complex AVM is uncertain, and radiosurgery is a good treatment option for this type of AVM. Obliteration rate after gamma knife radiosurgery (GKRS) for large cerebral arteriovenous malformations is 28–40% [8]. According to the literature, the incidence of post radiosurgery adverse effect ranges from 9% to 11% [2]. The adverse consequence of radiosurgery directly correlates with duration of follow-up. The adverse effects of radiosurgery are reported as short term long term complications. Long term (>10 years) adverse consequences of AVM treatment such as delayed hemorrhage, necrosis, seizure, arterial stenosis and cyst formation are reported in the literature [1]. The incidence of delayed complications quoted in literature is 3.2–19.8% [17]. The risk of hemorrhage after GKRS depends on larger size of AVM (e.g., Spetzler-Martin grade IV-V) and lower radiation dose [3,10]. Delayed cyst formation after GKRS in large AVM is a rare sequel. Break down of blood brain barrier, high blood flow volume and high radiation doses are the proposed hypothesis for delayed cyst formation [13,18]. We are reporting a case of large complex AVM (i.e., Spetzler-Martin grade V) received GKRS as the treatment after embolization who was presented later with large cyst. We have also reviewed the cases with GKRS induced cysts formation in the patients with AVM in existing literature.

2. Patient and methods

Standard search strategies were performed in PubMed/Medline using appropriate terms such as "intracranial arteriovenous malformation" radiosurgery, embolization and microsurgical resection as well as medical subject headings. The particular search strings used in the PubMed search builder and the number of article titles generated by strings are summarized in Table 1. The particular case in this study was retrospectively reviewed.

A 44-year-old male patient presented with complaints of left sided numbness and weakness at an outside hospital. The patient was hypertensive. Magnetic Resonance Imaging (MRI) of the brain

* Corresponding author.
E-mail address: ananda@lsuhsc.edu (A. Nanda).

https://doi.org/10.1016/j.jocn.2017.10.056
0967-5868/

revealed a right parietal arteriovenous malformations of Spetzler-Martin grade V. Neuro-interventionalist at the outside hospital performed embolization of the AVM. Initial strategy for embolization of AVM was to reduce the nidus size.

2.1. Procedure of embolization

Embolization is performed under general anesthesia in an angiographic unit. Systolic blood pressure was maintained between 100 to 110 mm of Hg. Catheterization was performed with transfemoral approach utilizing the slandered coaxial technique. Heparin was flushed to block the thrombosis in guiding catheter. Thereafter, a DMSO-compatible microcatheter was inserted to navigate the nidus of the AVM with the aid of a guidewire. Onyx was carried out in microcatheter once it has reached to the desired position of AVM nidus. Onyx was injected slowly in the nidus under fluoroscopic guidance. Angiogram was performed by pausing the Onyx injection to assess the AVM obliteration and status of draining veins. When the reflux of onyx reached 1.5–2 cm of microcatheter tip, the catheter was withdrawn [16].

After initial treatment with endovascular embolization, the patient came to our hospital for the second opinion. Four vessels angiogram was performed and showed a 19.5 cm^3. AVM with feeding arteries primarily from the middle cerebral artery and partially from the anterior cerebral artery. The venous drainage was towards the superior sagittal sinus and vein of Galen (Fig. 1A–D). After a discussion about the options, benefits and risks of available treatments, the patient decided to go with gamma knife radiosurgery.

2.2. GKRS procedure

Gamma knife radiosurgery was performed using the Leksell stereotactic unit model "C" (Elekta, Atlanta, Georgia, USA) with the automatic positioning system. The Leksell head frame was applied to the patient's head under local anesthesia and intravenous sedation. MRI the imaging data was then transferred to the Gamma Knife planning computer via the Ethernet. A team of neurosurgeon, neuro-oncologist and physicist were involved in gamma plan. The Leksell Gamma Plan software v.5.34 was used to perform the radiation dose planning. The median marginal dose to the tumor was 16 Gy, the median maximum dose to the tumor was 32 Gy and the median isodose line was 50%. Thirteen shots were given to complete the first radiosurgery procedure. During the second GKR, similar dose of radiation was used.

At three months follow-up after GKRS, the angiogram showed a 14 cm^3 AVM. The second stage of GKRS with the same dose (16 Gy) of radiation was repeated. At two and half year follow-up, the angiogram revealed a reduced size of the AVM 1.5 cm^3 with superficial and deep venous drainage**.** However, a small multi-septate cyst was noted medial to the AVM and patient was under observation. Five weeks later, the patient presented with hemiparesis, and an MR-angiogram was performed. MR and CT angiogram revealed the AVM with nidus and a large cyst (29 cm^3) lateral to it (Fig. 2A–C). Once again, after discussion with the patient, a craniotomy was performed, and a large amount of fluid was collected from the cyst as well as the cyst was fenestrated to the ventricle. A resection of AVM was also performed.

2.3. Microsurgical resection

After induction of anesthesia, the patients were put into Mayfield three-point fixation in a radiolucent frame. A linear skin incision was made in the parietooccipital area and skin flap was reflected. Thereafter, two burr holes were made and a 5 cm bone flap was removed. The dura was opened and then cyst was visual-

Table 1
Data of existing literatures with AVMs and cysts formation after radiosurgery.

Articles	AVM site	AVM volume (cm^3)	S_M grade	AVM Management, Embolization (E), Surgery (S), Radiosurgery (R)	Radiation dose (RD)	Number of cyst	Interval of cyst formation (years)	Cyst managementMarsupialization (M), Conservative (C)
Al Hinai Q et. al. [1]	Vermis, PO and frontal	3.3, 22.2, 0.5	I, IV, II	ESR, ER, SR	19	3/201	12	M,C, M
Matsuo T et al. [10]	P, F, F, T, O	5, 1.2, 13.56, 10.75, 11.45	NR	R, SR, R, R, R	14.4	5/109	NR	NR
Flickinger JC et el.[2]	NR	5.7	NR	R	19	5/102	7	Resection
Pan H-G et al. [11]	F = 3, P = 5, t = 2, o = 5 Thalamus = 5, pons = 1, cerebellum = 1	3.9	NR	E (11), R	20.7	20/1203	7.6	Resection
Shuto T. et al. [13]	F = 6, p = 3, t = 5, o = 2, thalamus = 1, callosum = 1	1.9–36, medin = 6 ml	NR	E = 7, SR = 10, R = 8	20	18/775	3.7	Resection = 2, shrinkage = 6, stable = 4, disappeared = 3, unstable 2, LFU = 1
Izawa M et al. [5]	NR	9.9 ± 3.7	I-II = 3, III-IV = 5	R	21.8	8/237	6.8	
Current	Frontal	19.44	IV	ER	16	1/101	2.5	Resection
Total	–	–	–	–	Mean RD = 19 Gy	60/1645 (3.6%)	1–12 (mean = 6.6 years)	Resection

NR = not reported. P = parietal, T = Temporal, F = frontal, O = occipital.