Long Follow-Up of Radiosurgery for Brainstem Cavernoma

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Key words
- Cavernous angioma
- Late complications
- Radiosurgery

Abbreviations and Acronyms
CAB: Cavernous angioma of the brainstem
MRI: Magnetic resonance imaging

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BACKGROUND: **There is a relative paucity of long-term follow-up of treatment of brainstem cavernous angiomas. This has led to uncertainty and a lack of consensus regarding optimum therapy, especially radiosurgery.**

METHODS: **Report of a single case with a 24-year follow-up.**

RESULTS: **This patient was minimally symptomatic before her radiosurgery procedure. She developed an acute complication, from which she incompletely recovered. Two decades later she has become more symptomatic, with new deficits, and magnetic resonance imagings disclose progressive brainstem atrophy.**

CONCLUSIONS: **Long-term follow-up of patients treated with radiosurgery is important.**

INTRODUCTION

In their review, The Management of Brainstem Cavernous Malformations, Drs. Almefty and Spetzler[1] cautioned that the optimum therapy of cavernous angiomas of the brainstem (CABs) is unclear, in part because "the duration of follow-up in these cases [of stereotaxic radiosurgery-treated brainstem cavernomas] is only a few years." The following is a report of a case with long follow-up.

CASE REPORT

In October 1991, a 22-year-old woman abruptly developed diplopia, mild headache, mild decrease of hearing in her right ear, and mild unsteadiness of gait. Her symptoms almost completely cleared in the following 2–3 months. A magnetic resonance imaging (MRI) disclosed a 2 × 2 × 1.2 cm left pontomesencephalic cavernous angioma (Figure 1). At another hospital she was treated with stereotaxic radiosurgery [1555 cGy to the isocenter] in May 1992. Three weeks after treatment she rapidly developed marked sensory loss over her right side from face to leg. An MRI was repeated 4 weeks after her procedure and showed enlargement of the lesion (Figure 2). She was treated for several months with oral steroids. After about 6 months sensory perception partially returned, but she was left with fluctuatingly severe unpleasant distortion of cutaneous sensory stimuli, which became more intense at the time of her menstrual cycle. Periodic MRIs for the next decade showed that the lesion had decreased in size, but persisted. There were no delayed hemorrhages. She was briefly employed as a teacher and raised a family.

Ten years after treatment she became aware that sensory perception was gradually worsening over the right side of her face and body. Twenty years after treatment the unpleasant sensory symptoms seemed to increase in intensity and she became aware of annoying hypersensitivity to sound, especially in the right ear, for which she uses acoustic earplugs. On neurological examination 24 years after treatment she was stable on Romberg testing and tandem gait and was accurate on finger nose. Reflexes were 2+ in both arms, 3+ right knee more than left, and 2–3+ both ankles but with no clonus. Pinprick perception was distorted and unpleasant over right face, trunk, and limbs. Vibratory perception was intact, including both great toes. Right palpebral fissure was slightly larger than the left and sounds were somewhat distorted and unpleasant in the right ear, but cranial nerves otherwise were grossly intact.

Brain MRI scan from 2005 showed an irregular roughly rectangular, quite dark, lesion in the superolateral aspect of the pontomesencephalic junction measuring 1.3 × 1.2 × 0.9 cm (Figure 3). There was no mass effect. Caudal to this there was mild but noticeable atrophy in the left side of the medulla. The brain itself appeared youthfully normal with nothing to suggest additional lesions. Brain MRI scan, with and without enhancement, in the spring of 2015 showed no change in the lesion and no enhancement (Figure 4). The degree of atrophy in the medulla and pons was more pronounced and now there was an area of diffuse mild T_2 brightening measuring approximately 5 by 7 mm caudal to the lesion.

DISCUSSION

CABs are relatively rare, and consensus is lacking regarding several aspects of their care. Recent reviews by Frisher et al[2] and Almefty and Spetzler[1] have specifically addressed this issue. Both reviews assume that the current dominant opinion is that small CABs, which have not bled, should not be treated aggressively with either open surgery or radiosurgery. On the other hand, larger lesions that are superficially situated and those that have bled more than once should be treated with surgical

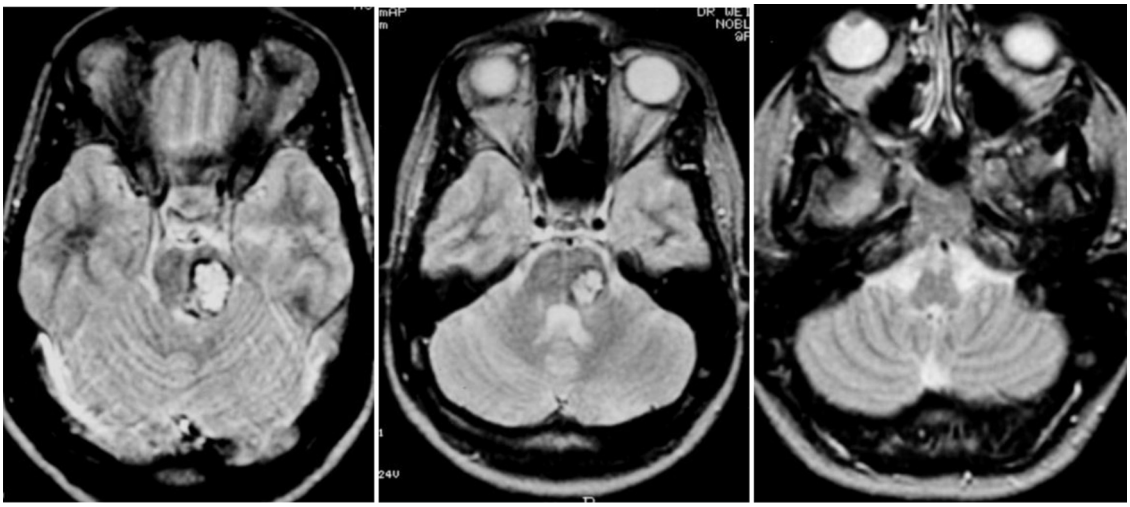

Figure 1. Magnetic resonance imaging scan at presentation. The patient was experiencing only diplopia, mild unsteadiness, and mild hearing loss in 1 ear.

excision. In these cases the divergence of opinion relates chiefly to a definition of which lesions are surgically amenable. Centers with more experience tend to recommend a more aggressive approach and are able to quote good outcomes. In the series by Frisher et al[2] most of their patients achieved good outcomes with overall improvement in Rankin scores, but 10% developed new neurological deficits after surgical resection.

More divergence of opinion relates to the treatment of CABs with radiosurgery, especially smaller lesions that are more deep seated and have bled only once. Almefty and Spetzler[1] stated that the primary objective of treating CABs is to reduce the risk of rebleeding, but the corollary is that the objective also is to prevent the development of new neurological deficits. The risk of rebleeding after radiosurgery of arteriovenous malformations and CABs seems to be the most within the first 2 years after treatment, but symptomatic radiation necrosis has been reported as late as 2 1/2 years after treatment.[3] Based on a literature review, Steiner et al[4] reported an overall complication rate after radiosurgery of cavernous angiomas of 19%, with half of these being permanent. Almefty and Spetzler[1] stated their concern that follow-up in reports of radiosurgery for CABs are usually only a few years, with few long-term reports,

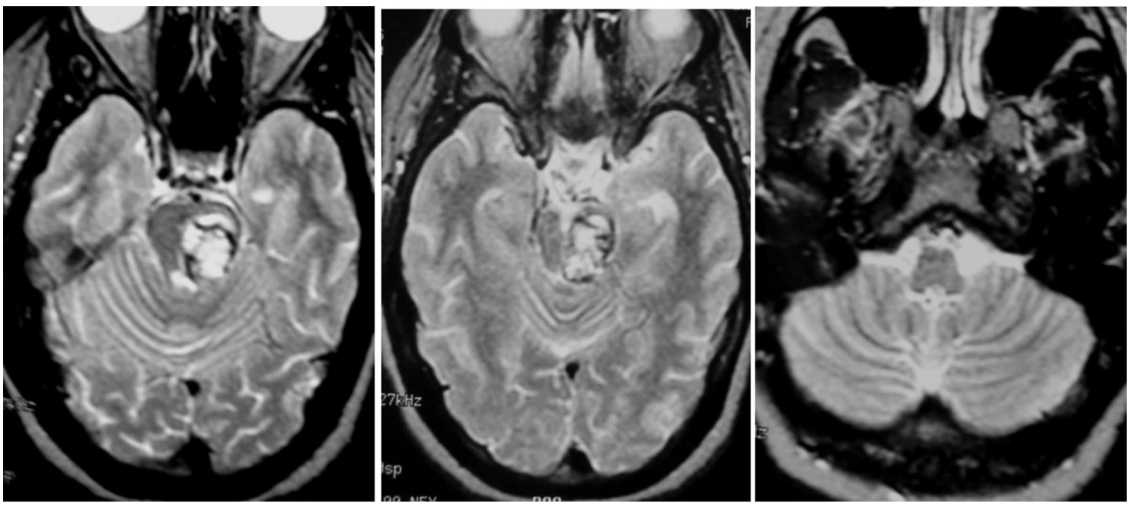

Figure 2. Magnetic resonance imaging scan from July 1992, 1 month after radiosurgery, demonstrating marked enlargement of the cavernous angioma of the brainstem when she was experiencing acute, new neurological deficits after the procedure.