Multiple meningiomas

TO THE EDITOR: We read with great interest the article by Tsermoulas et al.6 (Tsermoulas G, Turel MK, Wilcox JT, et al: Management of multiple meningiomas. J Neurosurg [epub ahead of print July 21, 2017. DOI: 10.3171/2017.2.JNS162608]) regarding the management of multiple meningiomas. The authors performed a retrospective observational study and described their comprehensive management strategy, providing details on epidemiology, decision making, and treatment options. The concept of multiple meningiomas was first proposed by Cushing and Eisenhardt in 1938,2 and researchers began to focus on this field thereafter. The character of multifocality, however, contributes to the complexity and challenge of the management of these cases. Tsermoulas and colleagues should be commended, because their study helps to address these issues by presenting comprehensive evidence.

We agree with the management principles of this study in general as well as with the authors’ point that patient- and tumor-specific factors should be considered in the decision-making process. With respect to radiotherapy, we believe that the potential benefits of this treatment should be balanced against the risks, because meningiomas can be both induced and treated by radiation. As described by the authors, in 26 (20%) of the 133 patients included in the study—with 110 (25%) of the total of 448 meningiomas—the tumors were considered to be associated with prior radiation at baseline, and a total of 27 patients with 55 meningiomas underwent radiotherapy modalities (in the form of Gamma Knife stereotactic radiosurgery [SRS] or fractionated radiation therapy).6 Radiation-induced meningiomas have been reported to develop decades after the original radiotherapy in a substantial proportion of surviving patients who were treated with brain radiotherapy in childhood.13,5 According to Strojan et al.,4 the latency period between radiation exposure and development of radiation-induced meningioma can range from 2 to 63 years. Given that the potential risks of radiotherapy as well as the finding that metachronous meningiomas were more common in the patients with radiation-induced tumors than in the sporadic meningioma group (62% vs 16%, p < 0.001),6 the treatment strategy should be chosen with caution, and longer follow-up is necessary for patients with multiple meningiomas associated with prior irradiation.

We note that 79 of the patients in this study were not treated at initial presentation and were followed instead. Of these 79, 31 (39%) required treatment at a later stage. Are there any differences between this group and the other 48 patients? More details may need to be explored in order to detect the clinical and radiological features that could predict a greater likelihood of the need for additional treatment. Patients who have a higher risk of requiring treatment of multiple meningiomas over their lifetime may need more vigorous surveillance.

A related issue that we would like to mention is that there is an interesting phenomenon among patients with multiple meningiomas called “mother-son tumor,” which was reported by Tian et al.5 Although Tsermoulas and colleagues did not provide the precise sizes of the tumors in their case series, they provided information about 67 patients with signs or symptoms attributed to one of their tumors, always the largest one. In our clinical practice, the type of multiple meningioma case that we encounter most often is a large meningioma in the presence of one or more small ones. The exact mechanism for this phenomenon is yet to be understood. Further studies are warranted to confirm and elaborate on this finding.

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Disclosures
The authors report no conflict of interest.

Response
We thank Dr. Wang and colleagues for their comments. The management of multiple meningiomas can be challenging, and the strategy we proposed was to treat the symptomatic tumors or the ones likely to become symptomatic. The rest should be observed for a period of time tailored to the individual patient. This is the same philosophy behind the management of single meningiomas as well.

We agree that it may seem paradoxical to treat radiation-induced meningiomas with radiotherapy. In our cohort, 22 of 26 patients with radiation-induced meningiomas had treatment; 16 were treated with surgery, 5 with Gamma Knife SRS, and 1 with a combination of surgery and SRS. Our treatment strategy for the radiation-induced meningiomas is resection when feasible. We consider SRS in tumors of appropriate size with high surgical risk, because the dose of radiation to the brain is clinically inconsequential and the control rate is satisfactory.

None of the patients with radiation-induced meningiomas had fractionated radiotherapy, because of the potential toxicity of re-irradiation and the fact that the long-term control rate of fractionated radiotherapy as primary treatment for meningiomas is not always satisfactory.

Regarding the 79 patients who were not treated at presentation, 31 had surgery and/or radiotherapy during surveillance and 48 did not require treatment. We did not compare the 2 groups because our study was not powered to examine possible differences in their baseline characteristics, but we accept that it is valuable to be able to predict which patients will need treatment and therefore we analyzed the relevant data. We found that the patients who had treatment during observation were more likely to have radiation-induced meningiomas (55% vs 9%, p = 0.003), be younger (52 vs 62 years, p = 0.007), and be male (48% vs 14%, p = 0.03). Radiation-induced meningiomas are more likely to be treated during observation because they are more aggressive than sporadic meningiomas. The age difference is self-explanatory, as meningiomas have more time to grow and become symptomatic in young patients and the growth rate is lower in older patients. There was no statistically significant difference in the number of meningiomas at presentation (p = 0.9) or the proportion of patients with large and medium-sized tumors in the 2 groups (p = 0.7 and p = 0.2, respectively).

Wang et al. mention the “mother-son tumor” phenomenon and report that the cases of multiple meningiomas that they encounter most often are large tumors with one or more small ones, implying a common origin from a single large meningioma. In our study, however, more than two-thirds of the patients (68%) did not have a tumor with a maximum cross-sectional diameter greater than 4 cm.

Finally, we agree that tumorigenesis of multiple meningiomas is not yet well understood and it is currently the subject of laboratory research in our center.

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References

Endoscopic approach for skull base malignancies: data sparsity

TO THE EDITOR: We were interested to read the article authored by Kim and colleagues (Kim YH, Jeon C, Se YB, et al: Clinical outcomes of an endoscopic transclival and transpetrosal approach for primary skull base malignancies involving the clivus. J Neurosurg [epub ahead of print June 2, 2017. DOI: 10.3171/2016.12.JNS161920]). The authors’ purpose was to retrospectively review the clinical outcomes of patients who underwent an endoscopic endonasal approach to treat primary malignancies involving the clivus and to analyze prognostic factors for gross-total resection (GTR). As one of the important results, they found that the tumor laterality was a significant predictor of GTR in both univariable (OR 6.25, 95% CI 1.51–25.86) and multivariable (OR 41.16, 95% CI 1.12–1512.65) models, which is questionable. It has been reported that a large measure of association such as an OR with a substantially wide CI is yielded in studies with sparse data.

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