# SURGICAL STRATEGIES FOR RECURRENT CRANIOFACIAL MENINGIOMAS

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**Received,** March 18, 2005.
**Accepted,** October 27, 2005.

**OBJECTIVE:** Recurrent cranial base meningiomas are among the most difficult tumors to treat surgically. Although they are histologically benign, these tumors often invade through the cranial base into the infratemporal and pterygopalatine fossae. We reviewed our experience with these tumors to describe the natural history of these lesions as well as provide a possible treatment paradigm.

**METHODS:** Between 2000 and 2004, seven patients with meningiomas recurring through the cranial base into facial structures were treated at the University of Utah. Five patients were treated with transcranial approaches only, and two were treated with a combination of transcranial and transfacial approaches.

**RESULTS:** The average age of our patients (6 women, 1 man) was 55 years. The original site of tumor was the sphenoid wing in four patients, the middle fossa in two patients, and the left frontal region in one patient. The average interval between the most recent tumor resection and recurrence into the face was 9.9 years. The mean number of resections a patient underwent before invasion into the face was two. All but one patient had adjunctive therapy (including either radiation or chemotherapy) before recurrence into the face.

**CONCLUSION:** Meningiomas that recur into facial structures present a unique treatment challenge. These lesions have a high rate of recurrence once they have invaded through the cranial base. Although combined approaches may be necessary to achieve a gross total resection, these lesions can often be reached using standard transcranial techniques.

**KEY WORDS:** Cranial base tumor, Craniofacial, Meningioma, Neoplasm

*Neurosurgery 58:874-880, 2006* DOI: 10.1227/01.NEU.0000209914.24576.DC www.neurosurgery-online.com

The management of meningiomas of the cranial base is controversial because of morbidity associated with aggressive resection and the high rate of recurrence with subtotal excision. Patients harboring these tumors often tolerate their large size well because of their slow progression. Even with extensive invasion into dura and bone, the presenting symptoms are often quite mild (4, 16, 17). It is precisely this invasion into dura and bone that makes complete resection impossible in many cases. Although new cranial base techniques have resulted in improved outcomes, the recurrence rate for these tumors remains high (1, 2, 7, 11, 33–36). Even after complete resection, the recurrence rate ranges from 7 to 10% at 5 years and 20 to 22% at 10 years (5, 15, 28). Incomplete resection carries significantly higher recurrence rates ranging from 26 to 37% at 5 years and 55 to 74% at 10 years (4).

Cranial base meningiomas, particularly those arising from the medial sphenoid wing, often recur into facial structures, including the infratemporal fossa, pterygopalatine fossa, orbits, and paranasal sinuses (6, 13, 16, 17, 22, 25, 32, 36, 38). Once they have recurred facially, they become a unique surgical challenge and require craniofacial expertise. Surgical management of these tumors often demands combined transcranial and transfacial approaches involving both neurosurgeons and otorhinolaryngologists (19, 20, 23, 26). Many of these techniques require significant disfiguring incisions and osteotomies. Various techniques have been described for resection of these tumors, but gross total resection remains the goal because incomplete resection will lead to further recurrence.

We describe our experience with surgical management of recurrent craniofacial meningiomas. Using different techniques to access the craniofacial skeleton, we have achieved gross total resections in seven patients with recurrent craniofacial meningiomas. Importantly, we have been able to avoid undesirable facial incisions and osteotomies that provide good exposure but are often cosmetically unacceptable. The utility of our paradigm to approaching these tumors and patient outcomes are discussed in terms of the clinical and radiological features of the tumors as well as the operative management, complications, and outcomes.

## METHODS

### Patient Population

Between 2000 and 2004, seven patients with recurrent craniofacial meningiomas were treated surgically at the University of Utah. Demographic data, clinical signs and symptoms, original tumor pathology, location, and imaging data were collected. All patients were treated with an approach involving a transcranial or a transfacial component singly or in combination. Pre- and postoperative imaging was used to assess the extent of surgical resection. Treatment-associated complications were also catalogued. Patient outcomes were based on postoperative assessments.

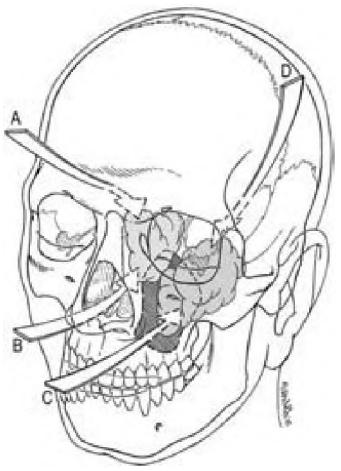


**FIGURE 1.** *Various routes of entry to meningiomas that have invaded through the cranial base into facial structures.* A, *transbasal approach, which can be used to access much of the midline face. "Blind spot" inferior to orbit may be visualized with an endoscope through intracranial exposure.* B, *transnasal approach.* C, *transmaxillary approach.* D, *frontotemporal (pterional) approach. Transcranial approaches can often be used to resect these tumors without undesirable facial incisions.*

### Treatment Paradigm

The surgical approach was chosen depending on the extent of invasion of the tumor into the face and its intracranial component. For tumors that were accessible from an intracranial approach, either a frontotemporal or a transbasal approach was selected. The frontotemporal approach has the advantage of familiarity and can be used to explore the middle fossa, the pterygopalatine fossa, the sella turcica, and the cavernous sinus. The transbasal approach has the advantages of accessing tumors involving the frontal sinus anteriorly, the ethmoid and sphenoid sinus posteriorly, and as low as the cervical–clival junction inferiorly. Decompression of the orbital roofs can be achieved from the transbasal approach. Even when the intracranial tumor has extended inferiorly into the nasal cavity and laterally into the maxillary sinus, complete resection can often be achieved from above (26). *Figure 1* shows the various routes of access to these tumors that have invaded into the pterygopalatine and infratemporal fossa.

When tumor removal left a large cranio-orbito-maxillary defect at the cranial base, reconstruction was performed with a vascularized flap (free flap or pedicled myocutaneous flap). Smaller defects were repaired with tensor fascia lata and fat autografts. These types of reconstructions are necessary to prevent a cerebrospinal fluid fistula (8, 21, 30).

## RESULTS

### Patient Data

Basic patient demographic data are summarized in *Table 1*. Six women and one man, with a mean age of 55 years (range 30–68 yr), were treated. Patients had undergone an average of two resections before recurrence into the face (range 1–4). An average of 9.9 years (range 3–26 yr) passed between the initial resection and recurrence into the face. According to previous operative records, all patients were believed to have had a gross total resection of their tumors at first operation. Of the three patients with multiple resections, the average length of

**TABLE 1. Demographic data of patients with meningiomas that have recurred into facial structures[a]**

| Patient no. | Sex/Age (yr) | Location of original tumor | No. procedures before recurrence | Interval (yr) | Site of recurrence into face | Adjuvant therapy before recurrence |
|---|---|---|---|---|---|---|
| 1 | M/54 | Left sphenoid wing | 1 | 9 | Infratemporal fossa orbit | Radiation, hydroxyurea |
| 2 | F/61 | Right middle fossa | 1 | 3 | Infratemporal fossa, maxillary sinus | Radiation |
| 3 | F/66 | Left sphenoid wing | 4 | 26 | Infratemporal fossa orbit | Hydroxyurea, tamoxifen[b] |
| 4 | F/68 | Left frontal | 3 | 10 | Orbit, sphenoid/ethmoid sinus | Radiation |
| 5 | F/45 | Right middle fossa | 1 | 6 | Pterygopalatine fossa | Radiation |
| 6 | F/30 | Left sphenoid wing | 2 | 5 | Pterygopalatine fossa, infratemporal fossa | Radiation |
| 7 | F/58 | Left sphenoid wing | 1 | 10 | Infratemporal fossa, maxillary sinus | None |

[a] Sphenoid wing origin refers to tumor arising from the dura along the sphenoid wing in the frontal and temporal fossae, whereas middle fossa origin refers to a tumor arising from the temporal lobe dura along the middle fossa floor or lateral temporal fossa

[b] This patient received radiation for retinoblastoma at age 18 months.

time between the previous resection and the recurrence into the face was 8.3 years. Four patients were treated with a solely transcranial approach, two patients required a transcranial as well as a transmaxillary approach, and one patient required a transbasal approach. Three patients required a vascularized free flap to achieve an adequate closure and to prevent a cerebrospinal fluid leak. The remaining four patients had tensor fascia lata autograft placed to provide a dural closure.

Six of seven patients had received adjuvant therapy including either chemotherapy or fractionated radiation before their facial recurrence. None of them received any adjuvant therapy after their last resection to treat the facial recurrence. The final pathology of all patients was benign meningioma (World Health Organization grade 1). Immediate postoperative imaging showed a gross total resection in all seven patients. At surgery, each of these patients was believed to have a gross total resection. Six of the seven patients had already received radiation before the recurrence and were not considered candidates for further radiation therapy. The last patient (patient 7) (*Table 1*) did not receive radiation before or after the resection of her recurrence because gross total resection was achieved. We decided to reserve radiotherapy to use if her tumor recurred. Complications (each occurring in 1 patient) included necrosis of a free flap resulting in a persistent cerebrospinal fluid leak necessitating a ventriculoperitoneal shunt, unilateral visual loss, wound dehiscence requiring a surgical revision, and a myocardial infarction. After their resections, four patients were discharged to home, two were discharged to an inpatient rehabilitation facility, and one was discharged to an extended care facility. All patients discharged to inpatient rehabilitation or care facilities were ultimately discharged to home.

### Illustrative Case 1: Patient 5

A 45-year-old woman had a right middle fossa meningioma that recurred into the pterygopalatine fossa 6 years after her original resection (*Fig. 2*). A transcranial approach (frontotemporal craniotomy) was used for reresection. A significant amount of bone was removed, including the floor of the middle fossa, exposing the infratemporal fossa (*Fig. 3*). A tensor fascia lata graft was used to repair the large cranial base dural defect. At surgery, it was believed that a gross total resection was achieved, and no residual tumor was left behind. Three years after the reresection, her follow-up magnetic resonance imaging scans reveal no evidence of recurrent tumor, and the patient is doing well.

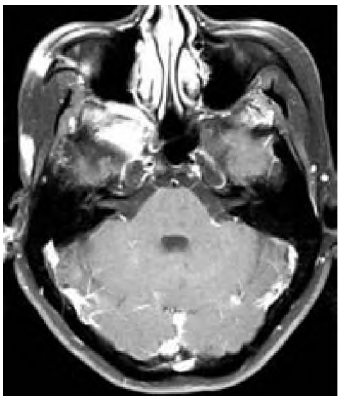

**FIGURE 2.** *Preoperative magnetic resonance imaging showing a meningioma that originally was located in right middle fossa and invaded through cranial base into pterygopalatine fossa.*

### Illustrative Case 2: Patient 2

A 61-year-old woman had a right middle fossa tumor that recurred into the infratemporal fossa and maxillary sinus 3 years after her original resection of a right middle fossa meningioma (*Fig. 4*). A combined approach (including a transfacial approach) was required to resect the extensive recurrence into the face (*Fig. 5*). The patient required a radial forearm free flap for reconstruction. Despite the aggressive resection (believed to be a gross total resection), the patient's tumor recurred within 14 months after surgery. Her most recent magnetic resonance imaging shows a tumor recurrence. She has declined further treatment.

**FIGURE 3.** *Postoperative magnetic resonance imaging showing resection with fat and tensor fascia lata used to repair cranial base defect.*

### Illustrative Case 3: Patient 6

A 30-year-old woman originally presented with a left-sided sphenoid wing meningioma. She underwent resection but experienced recurrence of her disease 1 year later into the left middle fossa. She underwent resection of this recurrence and presented almost 4 years later with another recurrence of tumor into the infratemporal and pterygopalatine fossae as well as the maxillary sinus (*Fig. 6*). Combined transcranial and transfacial approaches were used to resect this tumor. She underwent a frontotemporal craniotomy for resection of the intracranial portion of the tumor. The transfacial exposure was undertaken with the surgeons from the head and neck–otolaryngology department for resection of the maxillary and pterygopalatine extensions of tumor. The patient required a radial forearm free flap for reconstruction of the large cranial base defect after the resection (*Fig. 7*). A gross total resection was achieved. At 1 year follow-up, no evidence of recurrent tumor was observed.

## DISCUSSION

Convexity, parasagittal, and sphenoid ridge locations account for 75% of intracranial meningiomas (31). Although the

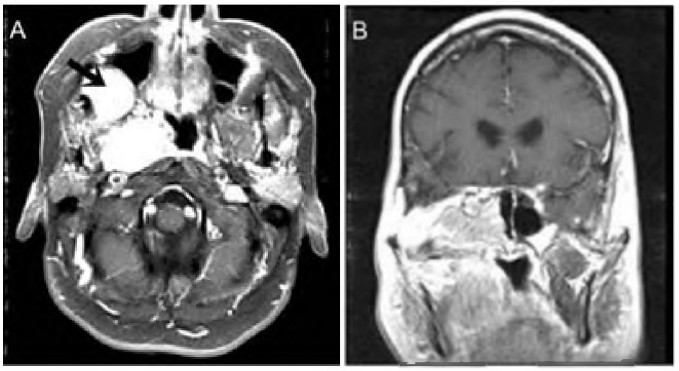


**FIGURE 4.** *Preoperative axial* (A) *and coronal* (B) *magnetic resonance imaging showing a meningioma originally located in right middle fossa that has invaded into infratemporal fossa and maxillary sinus (indicated by* arrow*).*

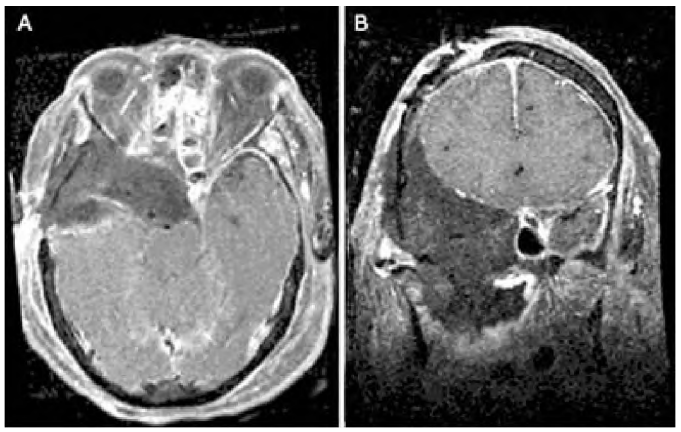


**FIGURE 5.** *Postoperative axial (A) and coronal (B) magnetic resonance imaging showing extensive resection with a radial forearm free flap used to reconstruct cranial base. The involved segment of the internal carotid was removed with neanastomosis after resection.*

**FIGURE 6.** *Preoperative magnetic resonance imaging showing a meningioma that originated from left sphenoid wing. This tumor recurred 4 years after initial resection and invaded into infratemporal and pterygopalatine fossae as well as maxillary sinus (indicated by* arrow*).*

first two sites are amenable to gross total resection and presumably a lower recurrence rate, sphenoid wing meningiomas may recur into facial structures, making their treatment more difficult. Although gross total resection should always be the primary goal of surgery, this may be tempered if tumor involves the cavernous sinus. In such cases, decompression of optic structures while avoiding damage to the cranial nerves traversing through the cavernous sinus may be the goal. Injury in this location may result in disabling gaze palsies. As a result of avoiding injury, tumor may be left behind, resulting in recurrence extending into the face. Recurrent meningiomas in such locations present a treatment challenge. Reresection often involves dissection of significant amounts of scar or irradiated tissue. When these tumors recur into facial structures, they will involve structures that may be unfamiliar to neurosurgeons. Nevertheless, resection should be possible with a combination of transcranial and transfacial approaches.

## Recurrence of Meningiomas into Facial Bones and Soft Tissue

The natural history of meningiomas suggests that recurrence is common. Ten-year recurrence rates are as high as 22% with a gross total resection and as high as 74% with a subtotal resection (4, 5, 28). These slow-growing tumors often present with benign symptoms such as headache but can also be very disfiguring, with facial invasion. The senior authors have found that recurrent meningiomas gained access to the face either by growing through existing foramen (such as the superior orbital fissure, foramen rotundum, or foramen ovale) or by direct extension through the floor of the middle fossa to the infratemporal fossa and facial bones. The tumor often results in hyperostosis, necessitating complete removal of the involved bone. In their review of their experience with craniofacial meningiomas, Gabibov et al. (17) indicated that most of the expansive lesions they had treated were the result of recurrences after "nonradical" operations. They found that most of their 54 patients had been operated on by ophthalmologists for orbital meningiomas or neurosurgeons for sphenoid wing meningiomas and had incomplete resections. Of note, 100% of the patients who were operated on for recurrent craniofacial meningiomas had subsequent recurrences between 6 months and 5 years postoperatively. This speaks to the importance of achieving a gross total resection whenever possible. Maroon et al. (27) found that sphenoid wing meningiomas with orbital extension have a recurrence rate of 35 to 50%. They stressed the importance of early and aggressive surgery with attention to sensitive structures including the orbit, cavernous sinus, and neurovascular structures in the medial sphenoid wing. They also indicated that hyperostotic bone is part of the disease process and should be resected. In their review of 15 sphenoid wing meningiomas with an average follow-up of 40 months (range 3–97 mo), Honeybul et al. (19) found that two patients demonstrated local recurrence after resection. Seven of their patients who were resected had residual disease that was not considered to be progressing. They recommended that these patients receive regular radiographic follow-up to evaluate for resection. Our experience suggests that it may take several years before these tumors recur (an average of >9 yr). In our limited number of patients, four patients originally presented with sphenoid wing meningiomas, and none of the patients who had recurrent facial meningiomas had a gross total resection originally.

**FIGURE 7.** *Postoperative imaging showing resection of tumor shown in Figure 6 after combined transcranial and transfacial approaches were used. A radial forearm free flap was used for cranial base reconstruction.*

## Choosing Surgical Approaches for Resection

Craniofacial access is well described for many lesions of the cranial base, benign and malignant, including juvenile nasal

angiofibroma, meningioma, adenoid cystic carcinoma, sarcoma, and squamous cell carcinoma. Treatment of craniofacial lesions including recurrent meningiomas is highly variable. Many surgeons advocate en bloc resection of these tumors, particularly if they are malignant, but less attention is given to functional and esthetic considerations as a consequence (9, 14, 22). Although this approach may provide excellent surgical access as well as an avenue for radical resection, it may be cosmetically unacceptable when combined with facial incisions and osteotomies. Several authors have classified various access routes, but the existing literature regarding these techniques is inconsistent. For example, Neil-Dwyer et al. (29) divided surgical approaches into anterior and lateral, with anterior approaches involving the cranium, midface, and mandible. They also described "midface" approaches to include transfacial and transoral techniques. The approaches to the anterior cranial base described by George et al. (18) include descriptions of transfacial, transbasal, and fronto-orbital ridge deposition, which were combined with a standard pterional craniotomy in some cases. In their extensive review of craniofacial access, they encountered seven meningiomas (of 43 benign tumors of the face), which were treated with the assistance of a classification scheme based on the anteroposterior axis and the vertical axis of the cranial base. They favored a transfacial approach for lesions that involved the infratemporal and pterygopalatine fossae. This involved a paralateronasal facial incision. Gabibov and Tcherekayev (16) offered a two-stage procedure: the transcranial stage included a bifrontal craniotomy with resection of the orbital and ethmoidal roofs for intracranial resection, and the transfacial approach involved a paralateronasal facial incision for access to the maxilla and nasal cavity. The variability in the literature regarding these approaches makes selecting the appropriate method to treat the challenging recurrent facial meningioma even more difficult. Although many of these techniques were described originally for malignant tumors originating from the sinuses or orbits, they are clearly applicable to recurrent craniofacial meningiomas, even through irradiated and previously operated tissues.

We and others have described techniques of accessing the facial skeleton that avoid disfiguring incisions yet provide enough exposure to perform a gross total resection (3, 10, 23, 24, 26, 37). With use of the transbasal approach, it is possible to gain access to the entire anterior cranial base, the nasal cavity, and the majority of the maxillary sinuses (26). Blind spots underneath the orbits are limiting in this approach, but increased visualization is facilitated by the use of endoscopy. An angled endoscope may be inserted through the intracranial exposure to visualize the inferior aspects of the orbits. More lateral extensions that are unobtainable from this approach may require a transmaxillary approach, as has been described previously (12, 26). It is important in these cases to resect all involved hyperostotic bone, which will form a nidus for recurrence if left in situ. Thus, if the recurrence is primarily midline, the tumor may be removed with a standard transbasal approach. Attention must be paid to closure and the necessity of dural closure (with appropriate substitute) and use of vascular pericranial flap, as described in previous publications (10).

For more lateral recurrences, including infratemporal fossa, pterygoid plates, pterygopalatine fossa, and posterior maxillary sinus, a decision must be made whether the tumor will be entirely accessible via the transcranial route. On the basis of our experience, the tumor up to the posterior maxillary wall (including the pterygopalatine fossa) will be accessible through the transcranial route. A transfacial approach may be necessary to resect the anterior and inferior portion of the tumor. In the cases presented here, we have chosen a transmaxillary route to the anterior aspect of the tumor if there is clear extension to the maxilla.

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## Acknowledgment

We thank Kristin Kraus for her superb editorial assistance in preparing this paper.

# COMMENTS

Rao et al. describe a series of patients with meningiomas that have grown into the face after initial treatment. Similar to many difficult neurosurgical problems, this one is better prevented than treated once it has occurred. Most of the authors' cases arose in patients whose initial tumor was located in the sphenoid wing or floor of the middle fossa and was thought to have been completely resected. This underscores the importance of removal of all abnormal bone at the initial operation as well as of any tumor that has grown through the temporal bone into the medial surface of the temporalis muscle. Cushing (1) and Cushing and Eisenhardt (2), citing other publications as early as 1910, recognized that the hyperostotic sphenoid wing adjacent to a meningioma frequently contains tumor cells. Pieper et al. (3) found this to be true in 25 out of 26 cases with hyperostotic bone identified preoperatively and noted the floor of the middle fossa as a location poorly evaluated for hyperostosis using standard axial computed tomographic imaging. Stereotactic guidance using computed tomographic images (rather than magnetic resonance imaging scans) can be very helpful in evaluating the extent of hyperostosis in these cases, as well as in gauging the progress of the necessary (though tedious) bony removal.

Surgery to resect facial recurrences that have extended through air sinuses and tissue planes as far as the thyroid cartilage can be surprisingly easy because the tumor molds itself into existing spaces without obliterating the sinus mucosa and shells out readily. An aggressive removal of these tumors is often mandated by threats to vision, but, in some elderly patients, palliative operations, with preservation of the nasal airway and reducing epistaxis as the goals, can be a reasonable alternative.

In our experience, meningioma recurrence into the face has sometimes been associated with a more aggressive histological grade than seen at the initial operation, especially after previous irradiation. Even in the absence of effective adjuvant treatment, an extensive resection can sometimes extend survival for years in this situation with reasonable quality of life.

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3. Pieper DR, Al-Mefty O, Hanada Y, Büchner D: Hyperostosis associated with meningioma of the cranial base: Secondary changes or tumor invasion. **Neurosurgery** 44:742–746, 1999.

Despite being benign tumors, cranial base meningiomas are surgically challenging. They also have a high rate of recurrence (even with a gross total resection). In this article, Rao et al. describe their experience with recurrent craniofacial meningiomas in seven patients. They appropriately emphasize that a gross total resection should be the goal of surgery, and they achieved a gross total resection in all of their patients. Rao et al. address recurrent meningiomas that extend out of the anterior or middle fossa. Although surgical treatment can be daunting because many of the current approaches have high morbidities with poor cosmesis, the novel approaches described by the authors allow for aggressive resections with lower morbidities and minimal disfigurement.

Although extensive surgical resection is a good initial step, the propensity of these tumors for continued recurrence is a concern. In the second illustrative case described, the tumor recurred at approximately 1 year out, despite a gross total resection. Harris et al. (1) examined the utility of stereotactic radiosurgery after surgery in patients with aggressive meningiomas and advocated radiation, especially if patients had residual tumor. For improved tumor control,

they also suggested that the radiation be introduced early in the postoperative period, rather than waiting for radiographic signs of progression (1). In the described series of aggressive recurrent craniofacial meningiomas, adjuvant stereotactic radiosurgery should be considered after surgery to reduce the risk of recurrence.

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The authors should be commended for trying to put some order into the exceedingly challenging issue of the surgical treatment of recurrent anterior cranial base meningiomas that transgress the cranial base and invade the facial structures. One of the challenges they highlight is the disfiguring nature of most of the surgical approaches that have been reported thus far. They recommend a treatment paradigm in which most of these tumors are approached through the anterior transcranial transnasal exposure. For meningiomas invading through the cranial base behind the posterior wall of the maxillary sinus and lateral to it, a transfacial approach may be additionally necessary.

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Surgical management of tumor recurrences is not very satisfying. When recurrences involve the cranial base and are "benign," the challenge is greater and the operative judgment more conflicted because expectations with benign tumors are probably greater than justifiable. The surgery is often a tedious, bleeding-obscured ordeal compounded by lack of normal tissue planes and landmarks to guide the surgeon. This surgical challenge is typified by the small group of patients with recurrent craniofacial tumors described in this article. This is not really a controversial area: nearly all patients had received adjuvant therapy (with varying degrees of success), so the treatment options at recurrence were limited. Nevertheless, the reasonably favorable results with aggressive resection provide some comfort for surgeons seeking justification in this no-win situation. The small number of patients makes generalizations about surgical approaches little more than anecdotal; however, the principle of aggressive surgery using standard surgical approaches is sound. Long-term results would be of interest to confirm the benefit of surgery in this complex group of patients.

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