

Management of sinonasal hemangiopericytomas*

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SUMMARY

The purpose of the present study is to report four cases of sinonasal hemangiopericytoma (HP) diagnosed and treated in our department between 1987 and 1998. The pretreatment findings and the treatment are described and discussed in the light of the literature. HP are unusual vascular tumors, featuring pericytes distributed around normal vascular channels. Two of these four cases were located in the nasal cavity and the other two were located in the maxillary sinus. Inside the nasal cavity, HP presented as a protruding reddish-gray mass with marked bleeding on contact. Electron microscopy and immunohistochemical techniques are essential for diagnosis and to distinguish HP from other sarcomatous tumors. Preoperative assessment included routine CT, MRI, arteriography and selective embolization. These tumors must be treated surgically with complete excision. An endonasal approach was performed in two cases of intranasal HP, while a combined external-endonasal approach was required for the other two cases of HP.

Key-words: hemangiopericytoma, sinonasal, management, radiology, surgery

INTRODUCTION

Hemangiopericytomas (HP) are uncommon mesenchymal tumors, representing about 1% of all vascular tumors (Batsakis, 1979). They are derived from extracapillary cells called pericytes, which surround normal vascular channels in this tumor. Less than one-third of HP occur in the head and neck and only 5% are located in the nasal cavities and paranasal sinuses (Batsakis, 1979). They are initially indolent, but gradually invade the surrounding tissues. Some forms of HP can be considered to be malignant. Late recurrences have been reported in almost one-half of the patients, metastases occur in up to 10% of the cases and the mortality is as high as 50%, after five to twenty years (O'Brien and Brasfield, 1965; Lund, 1996). Wide local excision and long-term follow up are therefore essential.

CASE REPORTS

Case 1

A 43-year-old black African male presented to our department in 1991 with chronic right-sided nasal obstruction. Nasal endoscopy showed a large reddish-gray mass obstructing all of the



Figure 1. Coronal CT showing a very large soft-tissue density mass with internal calcifications, entirely obstructing the right maxillary sinus, right ethmoid sinuses and right nasal fossa. Bony structures are deformed or eroded.

right nasal cavity. HP was diagnosed on biopsy. Preoperative computed tomography (CT) of the nose and paranasal sinuses

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Figure 2. Axial T1-weighted MRI, showing an iso-intense mass, located in the right anterior and posterior sinus.



Figure 3. Axial T2-weighted MRI, showing the same mass, with low signal intensity.

showed that the right maxillary sinus, right nasal cavity, and right ethmoid and sphenoid sinuses were totally filled with a thick soft-tissue density mass with internal calcifications (Figure 1). The walls of the maxillary sinus were also deformed and sometimes eroded, together with the roots of teeth 17 and 18. Carotid arteriography showed that the tumor blood supply was derived from the sphenopalatine and alveolo-antral branches of the right maxillary artery. The right maxillary, right facial and right ascending pharyngeal arteries were embolized. The patient was treated via a midfacial degloving approach. The anterior wall of the right maxillary sinus, the nasal process of the right maxilla and the right nasal bone were removed, provi-

ding large access to the maxillary and ethmoid sinuses. All these bony structures were sacrificed because of the extent of tumor invasion. The tumor and the floor and the periosteum of the posterior and external walls of the maxillary sinus were dissected. Teeth 17 and 18 were removed. Complete sphenothmoidectomy was also performed at the same time. The cribriform plate and ethmoidal roof were fortunately not invaded by the tumor. No bleeding or complications were observed. One year later, the right maxilla and right nasal bone were reconstructed by plastic surgery.

The patient remained in good health for 7 years. His physician referred him to us at the beginning of 1998 after detection of a tumor in his right nasal cavity. Endoscopic exploration showed recurrence of a reddish-gray mass in his right ethmoidal cavity. CT visualized a thick-walled spherical mass, suggestive of mucocele. On Magnetic Resonance Imaging (MRI), this mass presented an intermediate signal intensity on T1-weighted images (Figure 2), a relatively low signal intensity on T2-weighted images (Figure 3) and marked enhancement after intravenous injection of gadolinium chelate. In contrast with CT, the MRI findings were suggestive of local recurrence. The patient underwent strict endonasal surgery under endoscopic guidance. The tumor was easily separated from the surrounding healthy bony walls and was resected *en bloc*. The pathologist diagnosed a recurrence of HP.

Case 2

A 20-year-old Tunisian female was referred to our department in 1997 with a vascular tumor in her left maxillary sinus. This tumor was revealed following heavy bleeding after extraction of teeth 27 and 28, requiring control by a malar flap. Biopsy of the mouth revealed a diagnosis of HP. Clinical examination revealed a red mass inside the mouth, eroding the anterior wall of the maxillary sinus and protruding from the left dento-alveolar crest. Nasal endoscopy did not reveal any abnormality apart from intranasal bowing of the lateral nasal wall into her left inferior meatus. CT visualized complete opacification of the left antrum, with convex walls, but no bone infiltration, suggestive of an extensive, but benign tumor.

No tumor was observed in the other sinuses and nasal cavities. On MRI, the tumor had an intermediate signal intensity on T1-weighted and T2-weighted images, and was strongly enhanced after intravenous injection of Gadolinium chelate. Arteriography showed unipedicular hypervascularization of the tumor, derived from the left facial artery. The left facial and left maxillary arteries were simultaneously embolized.

The tumor was resected via a sublabial approach. A bone flap (including the anterior wall of the maxillary sinus and the nasal process of the maxilla) was raised and replaced at the end of the procedure. The tumor appeared to be encapsulated and was fairly easily removed from the walls of the antrum. Bleeding was minimal. Middle meatotomy was performed at the same time to check the lateral nasal wall and to allow postoperative endoscopic monitoring inside the cavity.

Twenty-four months after surgery, the patient was free of disease on the basis of endoscopic and CT examination.

Case 3

A 72-year-old Caucasian female was referred to the emergency ward with epistaxis occurring during biopsy of an intranasal tumor. She had previously complained of chronic and continuous left-sided nasal obstruction. Biopsy indicated a diagnosis of HP. After 48 hours of endonasal gauze packing, nasal endoscopy showed a bleeding mass in the left nasal cavity, protruding between the middle turbinate and nasal septum. CT showed a contrast-enhanced soft-tissue density mass, located in the left nasal cavity and nasopharynx and non-contrast-enhanced secretions filling the left (anterior and posterior) ethmoid cells and left maxillary sinus. Carotid arteriography showed a marked tumor blush with early draining veins, allowing selective embolization of the left maxillary artery with fairly satisfactory post-embolization control.

The tumor was resected via a strict endonasal approach under endoscopic guidance. The tumor had a pedicle on the nasal septum, 1 cm wide, 5 mm below the skull base. The pedicle was cauterized with a bipolar unit and the tumor (including the pedicle attached to the nasal septum) was then removed *en bloc*. The perpendicular plate of the ethmoid bone and part of the front wall of the sphenoid sinus were slightly eroded by the tumor. These areas were removed to obtain clear resection margins. Frozen sections were tumor-free. Sphenoid and posterior ethmoid sinuses were filled with thick mucus. No postoperative complications were observed and the patient was disease-free at 24-months follow up (endoscopic examination).

Case 4

Early in 1998, a 65-year-old Caucasian male mason presented with epistaxis. Clinical examination revealed an apparently hemorrhagic polypoid intranasal mass. This patient had experienced several episodes of epistaxis eleven years before, for which he was treated by cauterization of the vascular area of the nasal septum. He subsequently underwent septoplasty because of complete obstruction of the left nasal cavity by septal deviation. Another episode of epistaxis had occurred one month later, at which time a hypervascular polyp was observed in the left nasal cavity. Histological examination revealed a pseudo-inflammatory polyp with infiltration by lymphocytes and histiocytes and congested vessels.

The patient presented no bleeding or nasal obstruction during the intervening 11-year period, and then returned with spontaneous bleeding. General anesthesia was required for biopsy because of the hypervascular appearance of the tumor. Biopsy caused heavy bleeding and required 48 hours of bilateral endonasal gauze packing. CT revealed a homogeneous soft-tissue density mass entirely filling the left nasal cavity, extending into the left ethmoid cells and nasopharynx, with non-contrast-enhanced secretions in the left maxillary, frontal and sphenoid sinuses. The nasal septum and lateral nasal wall were deformed, but not eroded.

Biopsy revealed a diagnosis of HP (proliferative cells and mitoses) and review of the paraffin-embedded material obtained from the polyp in 1987 revealed early features of HP. Arteriography indicated an extensive bilateral tumor blood supply,

which was much more abundant on the left side. The blood supply was derived from the left anterior and posterior ethmoidal arteries, the right posterior ethmoidal artery and both sphenopalatine arteries, all of which were embolized.

Surgery was performed via a strict endonasal approach, under endoscopic guidance. The tumor was divided into several pieces until bleeding decreased. This tumor also presented a pedicle attached to the nasal septum below the cribriform plate, which was fortunately not invaded. This pedicle was cauterized with a bipolar unit. Tumor remnants were removed together with a large piece of nasal septum. Resection margins and frozen sections were tumor-free. The postoperative course was uneventful. One year later, no local recurrence was detected on endoscopic examination.

DISCUSSION

HP were first described by Stout and Murray in 1942. Their study included 691 cases of vascular tumors (mostly glomus tumors), in which they isolated (with silver reticulin stain) 9 tumors composed of distinctly extravascular cells, corresponding to capillary pericytes, first described by Zimmermann 20 years previously. This new entity, derived from pericytes, distinct from glomus tumors, was subsequently called HP. The real incidence of HP is unknown, as this entity has only been relatively recently identified and because of the small number of reported cases. HP seem to represent about 1% of all vascular tumors (Batsakis, 1979, 1983), but this percentage may be as high as 10% in children. Approximately 15 to 25% of all HP occur in the head and neck (Batsakis, 1979) and only 5% in the nose and paranasal sinuses (Bertrand et al., 1984; Batsakis and Rice, 1981). These tumors are usually derived from the musculoskeletal system, the skin of the limbs and trunk and the retroperitoneal area. In the head and neck, they are usually derived from soft tissues of the scalp, face or neck (Batsakis et al., 1983; Bertrand et al., 1984). The incidence of HP in the nasal cavity would be twice that observed in the paranasal sinuses. Sphenoid and ethmoid sinuses are involved four times more frequently than maxillary sinuses (Batsakis, 1979).

Reports of case series indicate an equal sex distribution. Two of our patients were Caucasians, one was North African and one black African, confirming the absence of any ethnic predisposition. HP have been reported at all ages, from birth to old age, but there appears to be a slight incidence peak in the 5th and 6th decades of life (Gudrun, 1979). The patients in our study belonged to various age-groups. The etiology of HP remains unknown. A history of trauma has raised the possibility of stimulation of proliferation of pericytes following damage to the capillary network. Long-term steroid therapy and hypertension have also been reported as predisposing factors, but have not been formally demonstrated (Walike and Bailey, 1971) and none of our patients presented any of these factors.

Bleeding (spontaneous or induced) and nasal obstruction are the main complaints in HP of the nose and paranasal sinuses. Pain can occur when lesions are sufficiently large to invade surrounding tissues or when they are confined in unyielding spaces such as the paranasal sinuses (Reiners et al., 1990). However, none of

our patients complained of pain, even the fourth patient with a very large tumor. The usual clinical course therefore consists of a gradually expanding, solitary and indolent mass (Bremond et al., 1975; Delsupehe et al., 1992). Macroscopically, these tumors are described as being soft and tan-colored, but our cases were reddish-gray, granular, raspberry-like, rubbery-thickened masses, which bleed easily and heavily on contact. This appearance is so specific that our last case immediately suggested the diagnosis of HP. Biopsies should be performed carefully in the hospital, because of the risk of bleeding. Preoperative assessment must include CT of the nose and paranasal sinuses with millimetric axial and coronal slices and injection of iodinated contrast agent. This tumor has a soft-tissue density, while sinus secretions show a lower (water) density and markedly desiccated mucus secretions are denser than soft tissue. These density differences are often sufficient to distinguish between tumor and surrounding inflammation. The tumor is contrast-enhanced, while secretions are not. Unfortunately, inflamed mucosa is also contrast-enhanced, which may be mistaken on CT for tumor margins. However, the presence of such signs indicates bone erosion and anatomic abnormalities, constituting surgical risks (Mosesson and Som, 1995; Klossek et al., 1993).

MRI usually visualizes a solid mass with an isointense signal on T1-weighted images, an iso- or low signal intensity on T2-weighted images and enhancement after gadolinium chelate injection. Any entrapped (or accompanying) secretions are not contrast-enhanced. However, tumor, inflammation and normal mucosa are enhanced and tumor size is consequently often overestimated on postcontrast images. It is therefore recommended to evaluate all paranasal sinus tumors by unenhanced, T2- and T1-weighted MR images (Mosesson et al., 1995; Klossek et al., 1993). Contrast enhancement is nevertheless helpful to distinguish mucocele from tumor, i.e. intense contrast-enhancement of tumor versus peripheral contrast-enhancement of mucocele. Gadolinium enhancement is also the best method to demonstrate the extent and nature of dural (or intracranial) components of the lesion eroding the skull base from below.

Conventional angiography played an essential role. It demonstrated the tumor blood supply and allowed preoperative embolization of vascular lesions. Preoperative embolization appeared to be useful for surgical resection of the tumor by decreasing intraoperative bleeding. However, as the real value of preoperative angiography with embolization in the management of HP has not yet been demonstrated, it cannot be recommended as the gold standard.

Some authors have reported intratumoral embolization of various vascular tumors, but we do not have any experience with this technique (Piérot et al., 1994; George et al., 1994; Casasco et al., 1994).

Histological examination of all four tumors, even on biopsy, revealed the typical morphological features of HP: sheets or randomly scattered ovoid or spindle-shaped cells with indistinct cytoplasm and large nuclei, distributed around normal vascular channels. HP tumor cells do not present any typical features; there only specific feature is their concentric extravascular site (Enzinger and Weiss, 1988). Histochemical and immunohisto-

chemical techniques can be useful for diagnosis by identifying reticulin, collagen fibers, smooth muscle and endothelial cells. Tumor cells are strictly confined to the extracapillary area. Although pathologists have not reached a consensus concerning well-defined grading criteria, all of our four lesions were benign according to Enzinger's criteria (Enzinger and Weiss, 1988). All four were well-differentiated and circumscribed, with neither invasion, nor necrosis or hemorrhage and not less than 4 mitoses per 10 High power field. According to most authors, the presence of mitotic figures is suggestive of malignancy and some authors have recommended DNA flow cytometry to define the prognosis (El-Naggar et al., 1992). However, some lesions have developed documented metastases, despite a benign histological appearance (El-Naggar et al., 1992). The differential diagnosis between HP and other neoplasms with prominent vascular patterns may raise considerable difficulties, but HP can generally be recognized by their uniform cellularity and vascular pattern and by the dense reticulin meshwork surrounding individual tumor cells (Enzinger and Weiss, 1988). The most important of the many tumors that can resemble HP are fibrous histiocytoma, synovial sarcoma and mesenchymal chondrosarcoma. Many other benign and malignant tumors must also be distinguished from HP: hemangiomas and glomus tumors (El-Naggar, 1992).

Various metastasis rates have been reported (35% to 57%), but the most likely percentage is 5 to 10% (De Campora et al., 1983). Retroperitoneal and deep extremity lesions, such as those in the head and neck, appear to have a higher propensity to metastasize. The most frequent sites of metastases are lungs, bones and liver (Millman et al., 1994; McMaster et al., 1975). However, no metastasis were observed in any of our cases. Chest x-ray and abdominal ultrasound were performed in all cases. Recurrence rates between 25 and 40% (McMaster et al., 1975) have been reported in the literature. Wide local excision is still considered to be the gold-standard treatment (Batsakis, 1979; O'Brien and Brasfield, 1965), but delayed reconstructive surgery may also be necessary. The type of surgical approach has yet to be defined: extranasal, endonasal or a combined approach. In HP of the nasal sinuses, an extranasal procedure should be preferred to entirely remove the tumor (Maniglia and Phillips, 1995; Weisman, 1995; Catalano and Chandranath, 1995; McGuirt, 1995) and the walls of the cavity (or periosteum) whenever they appear to be invaded. Despite wide excision, a recurrence was observed in one case seven years later. A combined procedure should therefore be preferred in these very large neoplasms. When the tumor is exclusively intranasal and especially when it is attached to the nasal septum, it can be removed together with a large piece of septum, via an endonasal approach under endoscopic guidance (Rice, 1995). This procedure may require rapid division of the tumor to control bleeding, which can remain abundant despite preoperative embolization. Once most of the tumor has been removed, bleeding becomes minimal and can then be controlled by bipolar cautery. The tumor can be entirely removed with safe resection margins. However, endonasal resection of HP must be limited to strictly intranasal lesions such as those reported here in cases 3 and 4. This kind of endonasal approach

must be carefully discussed on the basis of the radiological assessment (CT, MRI, angiography) providing optimal visualization of the site, size, extent of the tumor. The surgical team's experience in endoscopically-guided endonasal surgery should also be considered. However, in cases with maxillary or ethmoidofrontal sinus involvement, an external approach is mandatory. Moreover, the small number and short follow-up of the cases presented here, and the lack of convincing information available in the literature, prevent us from drawing any conclusions concerning the role of endonasal surgery for HP. However, in the case of an upper septal attachment of HP, the external approach would not appear to ensure better control of tumor resection than an endonasal approach. This latter technique, although associated with lower morbidity, obviously requires specific equipment and well trained surgeons.

Radiation therapy is not used, primarily because of the radioresistant nature of the tumor despite its large vascular component (Enzinger and Smith, 1976). The morbidity of high-dose irradiation alone is thought to exceed the morbidity of treatment comprising lower dose irradiation. There is also an increased risk of ocular (or optic) nerve complications whenever high-dose irradiation is used alone (Parsons et al., 1995). It can be used for advanced (and/or unresectable) lesions or in patients refusing surgery or ineligible for surgery for medical reasons or following incomplete resection. None of our patients were treated by radiotherapy.

CONCLUSION

HP are uncommon vascular tumors, exceptionally located in the nasal cavity and paranasal sinuses. They are slowly growing, indolent, reddish-gray masses, which bleed easily and heavily after any trauma. Our four cases were benign according to generally accepted clinical and pathological criteria. CT, MRI and angiography allowed preoperative assessment of the tumor, revealing extension and the tumor blood supply. Angiography then allowed preoperative highly-selective embolization. Tumor excision must be as wide as possible, sometimes requiring subsequent plastic surgery. An endonasal approach under endoscopic guidance can be proposed in the case of intranasal HP. Intrasinus HP usually require a combined extranasal approach. Life-long follow-up is recommended in the literature.

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