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Case Report

Olfactory Groove Anaplasic Meningioma: A Rare Histopathological Entity

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Abstract

Intracranial meningiomas continue to challenge our best clinical efforts to eliminate them once discovered and deemed appropriate for treatment. Malignant meningiomas constitute 10% to 15% of all meningiomas and limited information exists regarding adjuvant treatment. The external whole brain irradiation is recommended. Traditional chemotherapy has proven ineffective; thus, new chemotherapeutic agents and new methods of delivery should be developed. Immunotherapy may be considered for patients with malignant meningiomas when all others previous treatment have failed. We report a case of anaplastic papillary meningioma. A 60-year-old woman presented with a 10-year history of gradually decreasing olfactory function. A magnetic resonance image demonstrated a large Olfactory groove meningioma. The tumor and the infiltrated dura were radically removed. Postoperatively, the patient remained neurologically intact. The treatment was complemented by external whole brain radiation.

Keywords: Papillary meningioma, pathology, radiotherapy, chemotherapy.

INTRODUCTION

Meningioma, one of the most common types of brain tumors in adults, remains a clinical problem yet to be solved by neurologist, neurosurgeons and oncologists. Meningiomas constitute 15% to 20% of all primary brain tumor and 10% to 15% of allmeningioma are considered malignant [1, 2]. These tumors will recur after standard therapies of surgical excision, radiation therapy, radiosurgical techniques, and chemotherapy [1, 3-6]. We report a case of papillary meningioma, considered be analastic, and the literature is reviewed.

CASE

This 60-year-old woman presented with a 10-year history of gradually decreasing olfactory function. The patient had 1.5 years previously been diagnosed with stress, and as a result, reduced her work capacity from full time to part time. This stress particularly influenced the patient's ability for complex processing of tasks at hand, and the patient had to take one subtask at a time. During the previous 6 months, the patient had suffered from intermittent headaches but no history of nausea or vomiting. Our case begins as the patient was admitted to the emergency department with a first-time generalised seizure. Objective examination, including neurological examination, was normal, except for slight suspicion of dysarthria, which resolved. A gadolinium-enhanced cerebral MRI revealed a large extra-axial mass on the anterior skull base with extension to the left frontal lobe, with homogeneous contrast uptake and significant perilesional oedema. The largest diameter of the mass was 75mm. The olfactory function was preoperatively tested with the extended version of the Sniffin' Sticks7 (Burghart Messtechnik, Wedel, Germany) with a threshold score (T) of 2, discrimination score (D) of 7 and identification score (I) of 11. A total TDI score of 20—indicating severe hyposmia. No other causes of olfactory dysfunction were found. Based on the primarily left-sided basofrontal location, the absence of paranasal sinus infiltration and that the patient had intact but diminished olfactory function, a left-sided frontal craniotomy was performed to salvage the olfactory apparatus. Perioperatively, the right olfactory tract was found stretche around the basolateral side of the tumour and could not be preserved. The right olfactory tract was left intact. The tumour was completely resected, and the dural attachment coagulated, corresponding to a Simpson grade II resection (Fig 2). Postoperatively, the patient remained neurologically intact. There were no complications and the patient was discharged at one week. The histopathologic exam showed malignant meningioma with vascular channels surrounded by

neoplastic meningothelial cells with a papilliferous aspect and highly cellular areas with mitoses (Fig 3). The patient was referred to radiotherapy unit to receive complementary treatment with external whole brain radiation.

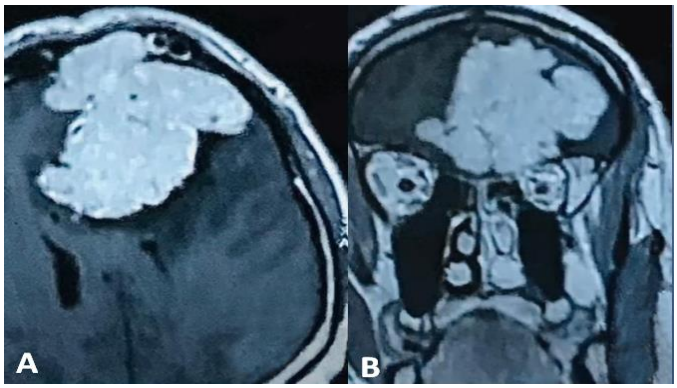

Figure 1: MRI in transversal (A) and and coronal (B) view showed a large extra-axial mass on the anterior skull base with extension to the leftt frontal lobe, with homogeneous contrast uptake and significant perilesional oedema

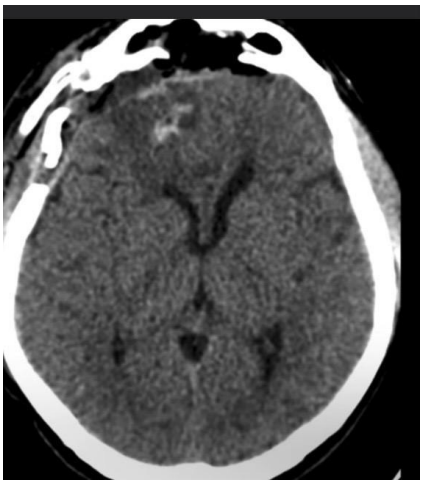

Figure 2: Postoperative CT scan in transversal view showed a total tumor removal

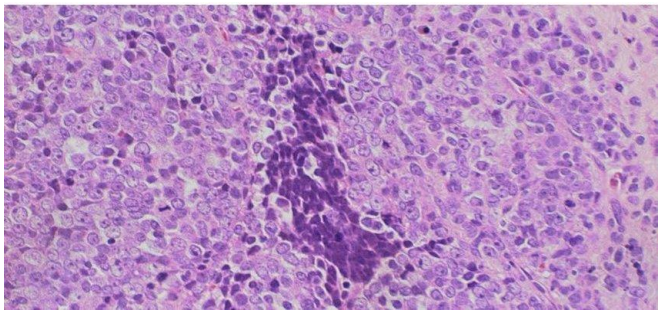

Figure 3: 1H-MRS (d, TE= 68 ms) demonstrates Lac and Lip (the arrow indicates the overlapping of Lac and Lip). The HE section (e, magnification, ×200) reveals high cell density and micronecrosis (arrows)

DISCUSSION

Malignant meningiomas represent 10% to 15% of all meningiomas [1]. The peak incidence of atypical and malignant meningioma was in the seventh and sixth decades, respectively [7]. These malignant meningiomas are defined by several criteria including: 1) invasion of adjacent brain parenchyma or skull; 2) numerous mitoses (> 5/high-powered field); 3) Elevated proliferative index (>3%) as assessed by either 5-bromodeoxyuridine or KI-67 staining; 4) necrosis; 5) Increased cellularity; 6) Nuclear pleomorphism; and 7) metastasis [1, 5, 8-12].

The cell origin of the meningiomas is the arachnoid cap cell, which has a slow rate of cell division. Although tumors originating from the meninges are typically benign, they occasionally behave in an aggressive fashion and carry a much poorer prognosis than do benign meningiomas [1, 3-6]. Tumorigenesis must be the result of exogenous or

endogenous factors acting alone or together. Exogenous factors include trauma, viral infection, and prior brain irradiation. Endogenous stimulation can occur through the action of hormones or growth factors. There was a clear tendency toward a progressively higher frequency of malignant meningiomas among recurrent tumor [13]. Malignant meningiomas were tumors that had undergone reoperation and had originally been either benign or aggressive meningioma. This suggests a likelihood that any benign tumor that recurs will be malignant [1, 6]. Whereas benign meningiomas tend to show preponderance in females, atypical and malignant meningiomas have a male preponderance [12, 14].

Pathology

Cushing coined the term "meningioma" to describe this tumor attached to the meninges. Since that time, a number of histopathologic schemes have been presented. Currently the WHO-II classifications of meningiomas are the most used. This histological classification system divides tumors of meningothelial cells into four groups: classic, atypical, papillary and anaplastic. These classifications can be complemented with the Helsinki grading system [15]. This grading system assigns points from 0-3 for six pathological features: loss of architecture, increased cellularity, nuclear pleomorphism, mitotic figures, focal necrosis, and brain infiltration. The sum of these points is then used to describe the benign, atypical, anaplastic and sarcomatous forms of meningiomas. The proliferative potential of tumors can be quantitated, using bromodeoxyuridine, KI-67, MIB-1 and proliferating cell nuclear antigen (PCNA) labeling index, and this information helps in predicting the clinical behavior of tumors and the need for treatment [1, 12]. Palma et al.6 defined the differencesbetween atypical (42 cases) and malignant (29 cases) meningiomas by the World Health Organization and studied the influence on prognostic: survival at 5 and 10 years was obtained in 95% and 79%, respectively, of patients with atypical meningioma and in 64.3% and 34.5% of patients with malignant meningioma; recurrence-free survival was 11.9 years in patients with atypical meningioma and 2 years with malignant meningioma; the authors concluded that radical extirpation and histological findings were significantly related to prolonged survival.

Radiological Features

Servo *et al*., [16] and Younis *et al*., [12] determined that CT cannot reliably distinguish malignant meningiomas from benign ones. There are, however, some CT or MRI trends that point in favor of malignant meningioma:

1. The absence of visible calcium aggregates12;
2. "Mushrooming" or the presence of a prominent pannus of tumor extending well away from the globoid mass [7, 10, 12];
3. Nonhomogeneous enhancement [10];
4. Necrosis [10]; and
5. Presence of indistinct tumor margins [7, 12, 16].

If angiography is performed, arteriovenous shunting is a feature that suggests malignancy1. Marked peritumoral edema, osteolysis, intrinsic cystlike areas and tumor density have a controversial radiological feature in relation to malignancy [12, 16]. Elster *et al*., [17] could not detect any significant difference on either T1-weighted or T2-weighted studies which allowed differentiation of malignant from benign meningiomas.

Surgery

Surgery remains an important part of treatment of malignant meningiomas. Over the past several years, advances in surgical technique and a revisiting of surgical sive approaches to brain tumors. Despite the grosstotal tumor ressection, the survival of malignant meningiomas without adjuvant therapy is less than 2 years [1, 5, 8-11, 18]. In patients with malignant meningiomas treated with surgery and adjuvant therapy (either radiation alone or radiation plus chemotherapy), median survival time was 5 years and the degree of tumor resection did not predict recurrence14. Chen & Liu4 reported that a recorrence rate with a median follow-up of 3 years after surgery was 44% for atypical or anaplastic meningioma and 6% for benign meningiomas. Younis *et al*., [12] reported that recurrence and survival time was shorter in patients with malignant meningiomas who had received partial resection on first presentation of tumor than in those who underwent total resection; in their series the patient's prognoses did not improve as a result of either chemotherapy or radiotherapy. Dziuk *et al.,* [13] reported a disease free/progression free survival at 5 years was 39% following total resection versus 0% after subtotal resection (p=0.001) in patient with malignant meningioma; they stated that complete surgical resection is crucial for longterm control. Malignant meningiomas located at the parasellar region and the posterior fossa a conservative removal of tumor followed by irradiation is advocated in preference to a radical operation that may cause neurological injury without being curative [1].

Radiation Therapy

The value of fracionated external beam radiation therapy or steriotactic radiosurgery in improving tumor control and survival for patients with subtotally resected, recurrent and malignant meningiomas is confirmed [1, 2, 5, 8, 10, 11, 19, 20]. Goldsmith *et al*., [21] showed that the 5-year progression-free survival rate after subtotal resection and radiation therapy was 89% for benign meningiomas and only 48% for malignant meningiomas; an improved progression-free survival rate was related with a younger age and treatment after 1980 with innovative

technologies, none of these variables affected the progression-free survival rate in the patients with malignant meningioma. Milosevic *et al*., [22] reported 59 patients with atypical or malignant meningiomas treated during the period of 1966 and 1990; the 5-year overall and cause-specific survivals after surgery and radiotherapy were 28 and 34%, respectively; age less than 58 years, treatment after the year 1975 and a radiation dose greater than or equal to 50 GY were associated with improved cause-specific survival; the authors recommended that all patients with atypical or malignant meningiomas receive radiation therapy immediately after surgery. Rodriguez *et al*., [18] analyzed 35 patients with malignant meningioma, of whom 15 (43%) were treated with surgery alone, 12 (34%) with surgery plus radiotherapy, six (17%) with surgery plus radiotherapy and chemotherapy, and two (6%) with no further treatment: the 3-year recurrence probability was 27% for patients who received adjuvant radiotherapy versus 69% for patients treated solely with surgical resection; the 5-year survival rate was estimated at 64%. Sixty-seven patients experienced recurrence at 5 years. According Dziuk *et al*., [13] adjuvant irradiation following initial resection increased the 5-years disease free survival rates from 15% to 80% (p=0.002); when administered for recurrent lesions, adjuvant radiotherapy improved the 2-years disease free survival from 50% to 89% (p=0.015), but had no impact on 5-years disease free survival; the authors concluded that extent of resection, adjuvant radiotherapy, and recurrence status are independent prognostic factors. Kondziolka *et al*., [19] treated 50 patients with meningiomas using the 201-source cobalt-60 gamma knife with a follow-up of 30-month; the authors concluded that radiosurgery was an effective primary treatment alternative for patients with advanced age, medical condition, or high-risk tumor location. Interstitial brachytherapy has been used to the treatment of malignant meningiomas after standard therapies have failed [23, 24]. Rogano *et al*., [23] reported their experience with iodine-125 sources placed at open operation for 22 patients with recurrent or malignant meningiomas obtained a 96 weeks of the median time to tumor progression and a 124 weeks of the median survival from the time of implantation; 38% of their patients had complications related to therapy; the authors conclude that interstitial brachytherapy remains an option for tumor volumes, shapes, and locations not amenable to stereotactic techniques.

Chemotherapy

The role of cytotoxic chemotherapy in the management of recurrent or malignant meningiomas has not fully investigated. The value of adjuvant chemo- anatomy have prompted more agres- therapy is not clear giving the confounding and probably more beneficial effects of adjuvant radiotherapy also administered to these patients14. Wilson1 reported 11 cases with recurrent malignant meningiomas treated with surgery, radiotherapy and chemotherapy using ciclophosphamida, doxorubicin and vincristine with 73% progressed at 1 year and 100% at 2 years. Stewart *et al*., [25] use intra-arterial cis-platinum and intravenous doxorubicin for inoperable recurrent meningiomas with a response in one case and tumor control in the other case. Chamberlain14 reported the use of cyclophosphamide, adriamycin and vincristine after external beam radiation therapy for up to six cycles of treatment for 14 cases of malignant meningiomas. Patients who had gross total resection only received three cycles of chemotherapy; the median time to tumor progression was 4.6 years and the median survival was 5.3 years; the author concluded that these combined modality therapies is associated with acceptable toxicity and a modest improvement in survival when compared to patients treated with surgery alone. Younis *et al*., [12] reported 10 patients with aggressive meningeal tumors who received intra-arterial or intravenous cis-platinum, intravenous dacarbazine and intravenous doxorubicin; clinical improvement or radiologic tumor response related to chemotherapy could not be observed. Rodriguez *et al*., [18] treated six patients with malignant meningioma with surgery plus radiotherapy and chemotherapy with free of recurrence at a median follow-up period of 4 years. The initial encouraging results on long-term oral therapy of 14 cases of unresectable meningiomas with the antiprogesterone mifepristone (RU486) remain to be proven in a larger cohort of patients [26].

Immunotherapy

Immunotherapy may be considered for malignant meningiomas when all others previous treatment have failed. The most effective immunotherapy appears to be administration of interferon-alpha, which is relatively non-toxic and easily tolerated [27, 28]. More studies are needed to better define the roles of these agents in the treatment of malignant meningiomas.

CONCLUSION

An aggressive treatment approach with radical surgery and postoperative radiotherapy is warranted in patients with these tumors. Traditional chemotherapy has proved innefective and the role of adjuvant immunotherapy, brachytherapy or radiosurgery are unknow. Because malignant meningiomas are uncommon tumors, a cooperative group study would be required to assess covariants.

Conflict of Interest: None

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