Intracranial Meningiomas Associated with Glial Tumours:
A Review Based on 54 Selected Literature Cases from the Literature
and 3 Additional Personal Cases

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Summary
The simultaneous occurrence of meningioma and glioma is extremely rare. Three new cases and 54 adequately described in the literature are analyzed. Clinical diagnosis may be difficult due to discrepancy between clinical and radiological findings. Unexpected clinical deterioration following removal of a tumour and relapse simulating recurrence may occur. The introduction of CT technology does not seem to have offered the expected contribution to the early diagnosis of these coincidental lesions, at least before the introduction of the newer generation scanners or MRI. While removal of both tumours in one session yielded the best results, surgery for the sole glioma appeared to be associated with an unacceptably high mortality. Although several aetiopathogenetic hypotheses have been suggested for explaining this curious association, coincidental meningioma and glioma are most likely to be different primary brain tumours occurring randomly in the same individual.

Keywords: Glioma; meningioma; coincidental intracranial tumours.

Introduction
The simultaneous occurrence of different primary brain tumours is a rare event which poses obvious problems of diagnosis and management.

The exclusion of cases occurring in patients with stigmata of “systematic hamartoblastomatosis” such as von Recklinghausen’s disease, von Hippel Lindau’s disease, tuberous sclerosis as well as “satellite tumours” (that is little discontinuously growing neoplasms possibly stimulated by a larger tumoural lesion, whether or not histologically related), “collision tumours” and gliosarcomas, makes multiple histologically different primary brain tumours occurring in the same individual rather uncommon.

In this group of multiple brain tumours, the association of meningioma and glioma represents the most common variant.

A review of the literature disclosed 111 cases of coincidental intracranial glioma and meningioma, to which we add three additional cases which we observed recently. The rarity of these occurrences and the problems in the diagnosis and management of this pathological condition, prompted the present report.

Case Material
We carefully reviewed the literature and found 111 cases reported from 1936 to date, but considered for the purpose of the present study only 54 of these instances. We excluded from the present review 23 cases whose clinical details were incomplete.

Table 1. Sex and Age in 54 Cases from the Literature and in the Three Present Cases

<table>
<thead>
<tr>
<th>Decade</th>
<th>1930s</th>
<th>1940s</th>
<th>1950s</th>
<th>1960s</th>
<th>1970s</th>
<th>1980s</th>
<th>1990s</th>
</tr>
</thead>
<tbody>
<tr>
<td>No.</td>
<td>8</td>
<td>15</td>
<td>20</td>
<td>30</td>
<td>11</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

Note: The table is not provided in the original text but is represented graphically.
Table 2. Localization of Adjacent Tumours in 18 Patients

<table>
<thead>
<tr>
<th></th>
<th>Meningiomas</th>
<th>Gliomas</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Hemispheric</strong></td>
<td>—</td>
<td>16.5, 14, 17, 19, 28, 42, 46, 48, 64, 70, 71, 75, 81</td>
</tr>
<tr>
<td><strong>Convexity</strong></td>
<td>16, 14, 17, 19, 29, 48, 51, 57, 71, 73, 81</td>
<td>—</td>
</tr>
<tr>
<td><strong>Falk/corpus callosum</strong></td>
<td>176</td>
<td>176</td>
</tr>
<tr>
<td><strong>Parasagittal</strong></td>
<td>176, 142</td>
<td>—</td>
</tr>
<tr>
<td>**Sphenoid wing/</td>
<td>173, 153</td>
<td>—</td>
</tr>
<tr>
<td><strong>basal frontal lobe</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Peritumoral</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Subtentorial</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Cerebello-pontine angle</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Other</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>19*</td>
<td>18</td>
</tr>
</tbody>
</table>

The case reported by Zbrojekiewicz et al. harboured a second contralateral meningioma.

Table 3. Localization of Non-Adjacent Tumours in 39 Patients

<table>
<thead>
<tr>
<th></th>
<th>Meningiomas</th>
<th>Gliomas</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Hemispheric</strong></td>
<td>—</td>
<td>34, 19, 21, 30, 22, 24, 25, 35, 45, 47, 56, 61, 63, 65, 72, 74, 76, 78</td>
</tr>
<tr>
<td><strong>Convexity</strong></td>
<td>17, 13, 19, 22, 24, 42, 50, 56, 57, 67, 72, 73, 79, 81</td>
<td>27, 22</td>
</tr>
<tr>
<td><strong>Falk/corpus callosum</strong></td>
<td>4, 12, 15, 40, 73</td>
<td>53, 11, 16, 49, 66</td>
</tr>
<tr>
<td><strong>Parasagittal</strong></td>
<td>4, 19, 76, Spallone et al.</td>
<td>179</td>
</tr>
<tr>
<td>**Sphenoid wing/</td>
<td>4, 19, 47, 73</td>
<td>—</td>
</tr>
<tr>
<td><strong>basal frontal lobe</strong></td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td><strong>Peritumoral</strong></td>
<td>27, 23</td>
<td>—</td>
</tr>
<tr>
<td><strong>Subtentorial</strong></td>
<td>177, 197</td>
<td>2, 23, 41</td>
</tr>
<tr>
<td><strong>Cerebello-pontine angle</strong></td>
<td>6, 11, 19, 22, 41, 61, Spallone et al.</td>
<td>5, 13, 15, 41</td>
</tr>
<tr>
<td><strong>Other</strong></td>
<td>2, 35, 46</td>
<td>—</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>40*</td>
<td>46**</td>
</tr>
</tbody>
</table>

The patient reported by King and Botton harboured two meningiomas.

** Five patients reported by Blihaye et al. (two), Kirchbaum (three), Madiouek and Shapiro (two) and our case nr. 3 (two) harboured multiple gliomas.

** In cases whose meningeal tumours showed features of histological malignancy, 5, 20, 22, 55, 78; 8 cases in which one of the tumours could have been induced by radiotherapy, 9, 23, 26, 54, 60, 77; 6 cases of demonstrated von Recklinghausen's disease, 20, 26, 44, 30, 77; 4 cases of "mixed" (meningeal and neoplastic tumours, 19, 79, 97, 2 cases

Fig. 1. Contrast enhanced CT scan, showing a hyperdense right parieto-occipital mass with marked odema extending anteriorly (thin black arrows). The scanty visible isodense right temporal lesion (large arrowheads) was not recognized at first

in which one tumour involved the spine, 2, 2 cases in which the development of the meningioma was related to previously occurring operative injury, 2, 3.

The relevant details concerning the 57 thoroughly documented cases are summarized in Tables 1–3. These include also our three recently observed patients, who are briefly reported in detail.

Case 1. A 47-year-old man presented on admission with a one-year history of generalized seizures associated with intellectual impairment. Neurological examination disclosed a left homonymous hemianopia, with bilateral papilloedema. A Computerized Tomography (CT) scan, unfortunately of poor quality, showed a right parieto-occipital mass with marked contrast enhancement (Fig. 1). Right carotid angiography showed caudal displacement of the Sylvian vessels and a fairly blush (Fig. 2A, B). Through a right occipital craniotomy, a convexity meningioma was completely removed. The postoperative course was uneventful and the patient was discharged neurologically intact except for the homonymous hemianopia. 15 months later, the patient developed a left hemiparesis and bilateral papilloedema and was readmitted. A disproportionate right temporo-parietal lesion surrounded by marked odema was demonstrated by CT scan (Fig. 3). Subtotal excision of an infiltrating and deeply located neoplasm of the temporal lobe was performed by a right temporo-parietal craniotomy. The histological diagnosis was grade II astrocytoma. After uncomplicated recovery, the patient was discharged with the sole persistence of the visual field defect present on admission. Appropriate radiation and chemotherapy was prescribed. Six months later, on a "routine" control, a CT scan failed to show any sign of recurrence. Recurrent tumour developed subsequently leading to death 18 months later.

Case 2. A man aged 33 was admitted in January 1985, with a depressive-anxiety syndrome of one-year duration as his chief com-
plaint. An episode of generalized seizure had occurred two months before. Neurological examination showed a right homonymous inferior quadrantanopia and an impairment of mentation. A CT scan performed elsewhere showed a right frontal parasagittal mass, most probably a meningioma. In view of the clinical findings, the patient underwent a second CT scan which disclosed another lesion in the left temporo-occipital region (Fig. 4A, B). Through a right frontal craniotomy and the endotheial parasagittal meningioma was removed as a first step. As a second step, 40 days after the first operation, and left occipital lobectomy was performed and a glioblastoma was partially removed. Radiation therapy on the location of the gial tumour was prescribed. The patient died of recurrence one year later.

Case 3. A 48-year-old man was first seen in May 1986, with a 4-months history of headache and confusion. A contrast CT showed a left deep paraventricular hypodense lesion – possibly a glioma – as well as a retrostellar mass extending to the cavernous sinus, with the CT characteristics of a meningioma (Fig. 5). A stereotactic biopsy of the paraventricular lesion gave evidence of a malignant astrocytoma. The patient refused open surgery and went to Freiburg (Prof. Mundinger), to have intracavitary stereotactic irradiation of the gliomatous lesion. This was performed in June 1986, when 10,000 rads of 1-125 were delivered to within the tumour. This was followed by 4,000 rads whole-brain irradiation. The patient underwent subsequently two full courses of BCNU chemotherapy and enjoyed good health until December 1986 when confusion and headache reappeared. A control CT showed again the possible parasellar meningioma, an area of decreased density in the left parietal region but also demonstrated an oval mass in the left insular region disomogeneously enhancing with contrast (Fig. 6). In January 1987 the patient again underwent stereotactic biopsy of the intracerebral masses in Freiburg. By that time the mass in the left insular region had progressed considerably. Histological examination of this lesion showed the meningioma remarkably similar to the first biopsied lesion. The possible meningioma in the retrostellar region never appeared to be symptomatic, therefore neither direct attack nor biopsy were considered. Despite intracavitary 7,000 rads irradiation of the second mass, no evident improvement was noticed. The patient progressively deteriorated thereafter, and died two months later. Autopsy confirmed the presence of the two discontinuous gliomatous masses and showed the hyperdense lesion to be a fibroblastic meningioma.

Discussion

Incidence: Multiple intracranial tumours represent 4 to 8% of all brain neoplasm. In particular, approximately 8% of the glial tumours may occur simultaneously in more than one location. As to meningeal tumours, the rate of multiple occurrences varies from 1 to 6% in different series. However it is well recognized that the association of meningioma and glioma in the same patient represents a rare event. Extensive search of large autopsy series might perhaps
Fig. 4. Contrast enhanced CT scan; A) right frontal parasagittal meningioma; B) left temporo-occipital glioma

Fig. 5. Contrast enhanced CT scan showing: A) an irregularly ring-shaped hypodense lesion situated in the left posterior paraventricular region; B) hyperdense homogeneously enhancing retrostellar mass extending to the left cavernous sinus

Fig. 6. Contrast enhanced CT scan (performed 7 months later with another scanner; sides are inverted) showing besides the lesions already demonstrated in the previous CT scan, another oval mass in the left temporal region; the retrostellar mass appears to be unchanged
increase the number of cases of association of meningioma and glioma, particularly if one considers that such cases may be discovered incidentally during "routine" autopsy, more often than expected.  

Pathogenesis: The occurrence of two (or more) primary intracranial tumours of diverse germinal origin in the same individual is undoubtedly an intriguing fact. The pathogenesis of which is still a matter of debate. Several hypotheses have been advocated in the past: the Cohnheim's theory on a multifactorial oncogenic role of embryonal rest cell, 

The association of meningioma and glioma is considered purely coincidental by most authors. However, the fact that in one third of the cases reported in the literature the two tumours were adjacent, demands a possibly different explanation. Therefore, the possibility that one of the tumours acts locally as an irritant factor ultimately inducing another histologically unrelated neoplasm has also been suggested. Clinical - radiological findings: Clinical presentation of coincidental meningioma and glioma shows several features which may well explain the commonly observed difficulty in the clinical diagnosis, including: discrepancy between clinical and radiological findings, as well as among the data of the different neuroradiological examinations; incidental discovery of the asymptomatic tumour during surgery for the symptomatic lesion or during "routine" autopsy; unexpected clinical deterioration either previous to or following removal of the asymptomatic neoplasm; relapse of clinical signs at a long interval after removal of one lesion, simulating recurrence.

Conclusion: It has been speculated that the association of multiple different intracranial tumours might represent a "forme fruste" of central neurofibromatosis. But no proof of this hypothesis has been given so far. It also has been suggested that the meningioma itself might have acted as oncogenic stimulus to the development of the glial tumour or "vice versa" in those cases, in which the two tumours are adjacent. Thus, the fact that adjacent location occurs only in about one third of the reported cases, speaks against this assumption. It also cannot be supported by any other findings. Therefore it is most likely, that the simultaneous
occurrence of meningioma and glioma in a single individual, as already stated by several authors, is not related causally but only randomly.

Acknowledgements
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