MULTIPLE PRIMARY INTRACRANIAL TUMORS

CASE REPORT

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The purpose of this report is to record a case presenting a combination of meningioma and glioma (oligodendroglioma), probably existing simultaneously but recognized 17 years apart. This case is presented to call attention to this rare problem which, if unrecognized, can lead to serious errors in management.

The presence of multiple intracranial tumors of the same histologic type is not rare. The more frequent examples include multiple meningiomas, multiple gliomas, and central neurofibromatosis involving two or more nerve roots. Courville in 1936 collected a series of 134 cases from the literature and his own experience. He stated that the incidence of multiple gliomas, in a series of autopsies, was about 1/2 per thousand, and that about 8 per cent of gliomas were multiple. The presence of multiple tumors of neuroectodermal origin, but of different histologic type, also is seen occasionally. A less commonly reported combination consists of tumors both of which arise from the primitive ectodermal layer, one from the neuroectoderm and the other from congenital rests of epithelial cells. An example of this combination is the case reported by Carson and Hellwig presenting a suprasellar adamantinoma and a cystic astrocytoma of the left temporal lobe. Rare instances of multiple intracranial tumors arising from different germinal layers have been recorded.

The histogenesis of the meninges is still undetermined. Although there is evidence for their origin from the neural crest (ectodermal) there is much to indicate that they behave more like mesodermal derivatives. It is uncertain, therefore, whether meningioma should be considered a tumor of ectodermal or mesodermal origin and whether the present case represents simultaneous neoplasia in different germinal layers.

CASE REPORT

M.R. (#153538), a 25-year-old female, was admitted to the Cincinnati General Hospital on Jan. 4, 1941 complaining of blurred vision. Three months previously she experienced sudden onset of occipital headache which recurred with increasing frequency up to admission. Ten days prior to entry she noted blurring of vision in the right eye and later in the left eye. There also was elicited a history suggesting olfactory hallucinations of indeterminate duration.

Examination. General physical findings were normal except for enlargement of the uterus to 4 finger breadths above the symphysis, interpreted as a pregnancy of approximately 2 months. There was bilateral papilledema of 3 diopters with atrophy of the right disc. Perception of light only was present on the right. Right hemihypalgesia, more marked in the leg, was noted, as well as weakness of that leg and ataxia of the right arm and leg.

Lumbar puncture revealed an initial pressure of 375 mm. of water. The fluid was clear and colorless and contained 21 mg. per cent protein.

During the next 2 days the patient became more drowsy and right hemiparesis became more pronounced.

On Jan. 8, 1941, ventriculography was performed (Fig. 1A). Fluid obtained from the left lateral ventricle was xanthochromic while fluid from the right ventricle was colorless.

1st Operation. Left frontal craniotomy was done and a grossly total removal of an intraventricular tumor was achieved. The tumor was a reddish-gray vascular mass blocking the left foramen of Monro and extending backward along the body of the ventricle. Some exploration along the falx was performed but no tumor was found in this region.

Histologic Diagnosis. The tumor removed from the ventricle was interpreted originally as an ependymoma.

Recently the microscopic sections of this tumor have been reviewed by Dr. Abner Wolf, New York City, who wrote “the tumor in the left frontotemporal region, removed 17 years ago, is an oligodendroglioma (Fig. 2) even though it is intraventricular in position. In this region, the oligodendroglioma may arise from the septum pellucidum or from this portion of the ventricular wall and sometimes have an intermixture of ependymal type of tumor. I did not find any such in this tumor, however. The features that I think are typical of oligodendroglioma are the high cellularity, relative uniformity, delicate and rather regular vascular network, small cells with rather regular spherical nuclei and the presence of perinuclear halos in same cells.”

Postoperative course was difficult, requiring several ventricular taps. Two weeks later, roentgen-ray therapy was instituted and a calculated tumor dose of 3840 r was administered through 4 ports.

The patient was discharged on Feb. 27, 1941 and followed in out-patient clinic over the subsequent 17 years. Marked visual loss, associated with optic atrophy in the right eye, persisted although papilledema had
Fig. 1. (A) Ventriculogram performed Jan. 8, 1941. Separation of the lateral ventricles is seen but other views were interpreted to indicate an intraventricular mass on the left. (B) Ventriculogram performed July 11, 1958. Ventricular separation is seen again and other views confirmed marked deformity and compression of the frontal horns.

Fig. 2. Oligodendroglioma removed Jan. 8, 1941. Hematoxylin and eosin, $\times 650$. 
disappeared. She had occasional grand mal seizures which were controlled satisfactorily by anticonvulsant drugs. She also noted occasional headache and disturbance of memory. In general, the patient did very well during this interval.

Readmission, July 1, 1958. During the previous month the patient had exhibited increasing loss of memory, confusion, change in personality, unsteadiness of gait, and several episodes of loss of contact without motor seizures.

Examination. Positive neurological findings were lethargy, partial disorientation, visual deficit in the right eye with optic atrophy, generalized hyperreflexia with bilateral Hoffmann reflex and equivocal plantar responses, and ataxia and tremor of all extremities.

Carotid arteriogram and ventriculogram (Fig. 1B) showed deformities consistent with a large midline tumor.

2nd Operation. Left frontal craniotomy revealed a large tumor extending into the left frontal lobe from the falk, consistent grossly with meningioma. This portion of the tumor was removed and 2 weeks later tumor of similar size was removed from the right frontal region.

Histologic diagnosis of the tumor was meningioma (Fig. 3).

Postoperative course was one of gradual mental clearing and improvement of right hemiparesis which was present after the first removal of meningioma. On her last follow-up visit in January 1960 there was complete neurologic recovery except for residual right optic nerve atrophy.

DISCUSSION

We have been able to find 18 recorded instances in which an intracranial glioma was associated with a meningioma (Table 1). These include 13 case reports, and 5 cases referred to by Russell and Rubinstein. The neuroectodermal tumors represented were as follows: glioblastoma multiforme 9, reported as gliomas or gliogenous tumors 3, astrocytomas 4, oligodendroglioma 1, and spongioblastoma ependymale 1.

In 4 instances the meningioma appeared to be the younger tumor and was in juxtaposition to its neuroectodermal mate. In these cases, the stimulus for development of a meningioma might be considered a reaction from the adjacent neoplasm, since trauma and irritation have been incriminated as etiological factors in the development of meningeal tumors. However, in at least 8 of the reported cases, meningioma was in a distant part of the brain, and in some cases in the hemisphere opposite from the glioma. In this regard, Myerson, discussing a case in which there occurred 6 primary tumors of varying histologic pattern in one cerebral hemisphere, suggested...
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TABLE 1

Reported cases of intracranial glioma associated with meningioma

<table>
<thead>
<tr>
<th>Author</th>
<th>Date</th>
<th>Meningioma</th>
<th>Glioma</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Shapland &amp; Greenfield®</td>
<td>1935</td>
<td>Multiple, with central neuro-</td>
<td>Frontal lobe and cord</td>
</tr>
<tr>
<td></td>
<td></td>
<td>matosis</td>
<td></td>
</tr>
<tr>
<td>2. Cushing &amp; Eisenhardt®</td>
<td>1938</td>
<td>Paratemporal</td>
<td>Glioblastoma, corpus callosum</td>
</tr>
<tr>
<td>3. Myerson®</td>
<td>1942</td>
<td>Two, right hemisphere</td>
<td>Four, right hemisphere</td>
</tr>
<tr>
<td>4. Arieti®</td>
<td>1944</td>
<td>Right frontal, angioblastic</td>
<td>Astrocytoma, right frontal</td>
</tr>
<tr>
<td>5. Arieti®</td>
<td>1944</td>
<td>Left temporal</td>
<td>Astrocytoma, left para-occipital</td>
</tr>
<tr>
<td>6. Kirschbaum®</td>
<td>1945</td>
<td>Intracerebellar</td>
<td>Multiple subependymal glioblastomas</td>
</tr>
<tr>
<td>7. Feiring &amp; Davidoff®</td>
<td>1947</td>
<td>Fissure</td>
<td>Glioblastoma, frontal lobe</td>
</tr>
<tr>
<td>8. Alexander®</td>
<td>1948</td>
<td>Frontal convexity</td>
<td>Spongiosarblastoma ependymal, lateral ventricle</td>
</tr>
<tr>
<td>10. Hoffmann®</td>
<td>1952</td>
<td>Cerebellopontine angle</td>
<td>Astrocytoma, temporoparietal</td>
</tr>
<tr>
<td>11. Fincher®</td>
<td>1954</td>
<td>Left frontoparietal</td>
<td>Astrocytoma, temporal lobe</td>
</tr>
<tr>
<td>12. Fincher®</td>
<td>1954</td>
<td>Right frontoparietal</td>
<td>Glioblastoma, occipital</td>
</tr>
<tr>
<td>13. Austin et al.®</td>
<td>1958</td>
<td>Left temporal</td>
<td>Astrocytoma, right temporal</td>
</tr>
<tr>
<td>11-18. Russell &amp; Rubinstein®</td>
<td>1959</td>
<td>Details not given</td>
<td>Glioblastoma</td>
</tr>
</tbody>
</table>

Several factors might have permitted the patient to harbor this slowly growing neoplasm for many years following removal of the glioma: 1) the intraventricular oligodendroglioma was undoubtedly producing some increase of intracranial tension, as the operator noted that it appeared to block the left foramen of Monro; 2) a portion of the bone flap was removed at the first operation, thus allowing some decompression; 3) meningiomas of the falx characteristically achieve a large size before recognition; 4) seizures, which are often the first sign of such a lesion, were in this case easily attributable to the previous intracranial surgery; 5) the patient had no further pregnancies.

The relationship of pregnancy to the symptomaticatology of meningiomas has been recognized repeatedly. Our patient was pregnant at the time of her first hospitalization, but thereafter had no pregnancies. Symptomatic exacerbation probably is attributable to increase in the volume of circulating blood occurring during pregnancy or to some metabolic alteration that accelerates growth of tumor.

It is noteworthy that of the previously recorded cases of combinations of meningiomas and gliomas in only 2 was the diagnosis made during life. This obviously is a reflection of the diagnostic difficulty encountered in considering the possibility of multiple tumors of different histologic character. In a case reported by Feiring and Davidoff® the second tumor was diagnosed before death but unfortunately was a glioblastoma multiforme and the patient died 2 months after operation. In a case of Gass and Van Wagenen® the tumors were found in juxtaposition at opera-
tion and were removed simultaneously. It is of interest that, in this particular case, the tumors were meningioma and oligodendroglioma.

**SUMMARY**

A case is presented in which a patient had removal of a large bilateral meningioma of the falx 17 years after she had undergone removal of an intraventricular oligodendroglioma. It seems quite likely that the meningioma was present at the time of her initial operation. Although the occurrence of multiple intracranial tumors of different histogenesis is quite rare, this possibility should be kept in mind when symptoms persist following removal of a neoplasm, or when recurrent symptoms are not easily attributable to regrowth of the original tumor.

**REFERENCES**