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Angioblastic meningioma with hepatic metastasis

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SUMMARY Infrequently, intracranial neoplasms metastasize to extracranial sites. In 1963, Glasauer and Yuan reviewed the 88 reported cases of metastatic intracranial tumours of which approximately two-fifths were meningiomas. This report concerns an angioblastic meningioma with a large hepatic metastasis. Cushing's original classification of angioblastic meningiomas and the differential diagnosis between these tumours and the haemangiopericytoma and cerebellar haemangioblastoma are discussed.

Meningiomas comprise approximately 14% of intracranial tumours and are the second most common intracranial tumour of adults (Russell and Rubinstein, 1963). Although it is not unusual for meningiomas to recur locally or even to invade adjacent bone and muscle, metastases outside the cranial vault are rare. In a review of the literature in 1963, Glasauer and Yuan (1963) found only 88 intracranial tumours with extracranial metastases; approximately two-fifths of these tumours were meningiomas. This report discusses a patient with repeated local recurrences of an angioblastic meningioma over a period of 17 years, who at necropsy was found to have a large solitary hepatic metastasis.

CASE REPORT

CLINICAL HISTORY A 76 year old white female was first admitted to The New York Hospital 17 years before death with a six month history of occipital headaches and recent onset of ataxia, dysarthria, and intermittent vomiting. She had dysmetria of the upper extremities. Pneumoventriculography revealed a dilated ventricular system with absence of filling of the aqueduct and fourth ventricle. At operation, a 90 g firm, encapsulated tumour mass, solidly attached to the inferior surface of the tentorium, was resected. Postoperatively, the patient did well, with disappearance of her neurological deficit until 11 years before death when she was readmitted with homonymous hemianopsia and recurrence of the headaches, dysarthria, and dysmetria. Arteriography showed a large mass in the right posterior fossa. At operation, a small nodule of residual tumour was found at the original site. In addition, a 70 g encapsulated mass was firmly adherent to the superior surface of the tentorium. The tumour was excised along with the tentorium and transverse sinus which had been extensively invaded by tumour. After surgery, the hemianopsia and ataxia resolved and once again she did well until three years before death when she developed recurrence of her neurological symptoms. A 125 g tumour was removed from the posterior fossa. Her final admission was preceded again by several months of headaches, ataxia, dysarthria, and dysmetria of the right upper extremity. Brain scan revealed a large posterior fossa tumour. A 125 g tumour mass filled the posterior fossa bilaterally. After this fourth resection of the tumour she did well. However, two days before death, she perforated a duodenal ulcer and developed diffuse peritonitis. Several hours after plication of the ulcer, she had a cardiac arrest and died.

NECROPSY FINDINGS (New York Hospital No. 24357). Examination of the superior surface of the right cerebellar hemisphere revealed a large surgical defect that communicated with the fourth ventricle. The right occipital pole was distorted by haemorrhage and necrosis. There was haemosiderin staining of the right occipital cerebral tissue, the meninges at the base of the brain, and the ependymal lining of the fourth ventricle, aqueduct, and posterior right lateral ventricle. The dura mater in the right posterior fossa was thickened and fibrotic, but no residual tumour could be identified grossly. A discrete 10 cm firm, yellow-white tumour mass was present in the right lobe of the liver (Fig. 1).

Additional findings included a plicated duodenal ulcer, diffuse, purulent peritonitis, and multiple bilateral thromboemboli of the pulmonary arteries.

HISTOLOGICAL FINDINGS The tumour resected 17, 11, three years and finally one month before death was identical with the solitary focus of microscopic tumour found in the dura mater overlying the right cerebellar

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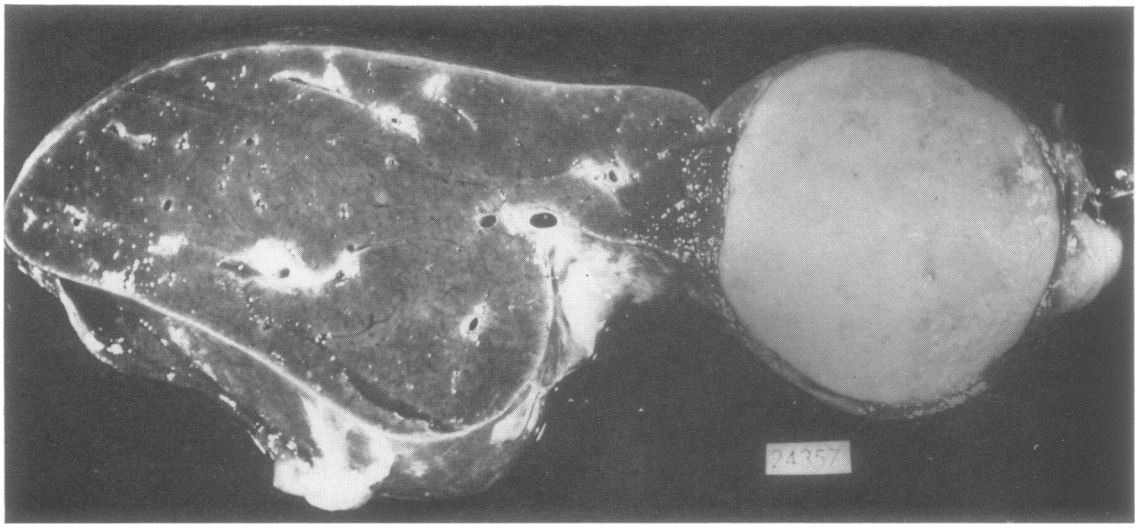

FIG. 1. *Liver with large solitary tumour mass in right lobe.*

hemisphere. The tumour was composed of sheets of polygonal cells with scanty, acidophilic cytoplasm, poorly-defined cell borders, and ovoid vesicular nuclei with prominent nucleoli (Fig. 2). Some nuclei were elongated and occasionally spindle-shaped. Numerous small, endothelial-lined blood vessels were present. Reticulum and fine collagen fibres surrounded the blood vessels and the individual cells (Fig. 3). The solitary hepatic metastasis showed an identical histological appearance (Fig. 4). Extensive sectioning of the lungs failed to reveal metastatic tumour.

Microscopic examination of the right occipital lobe

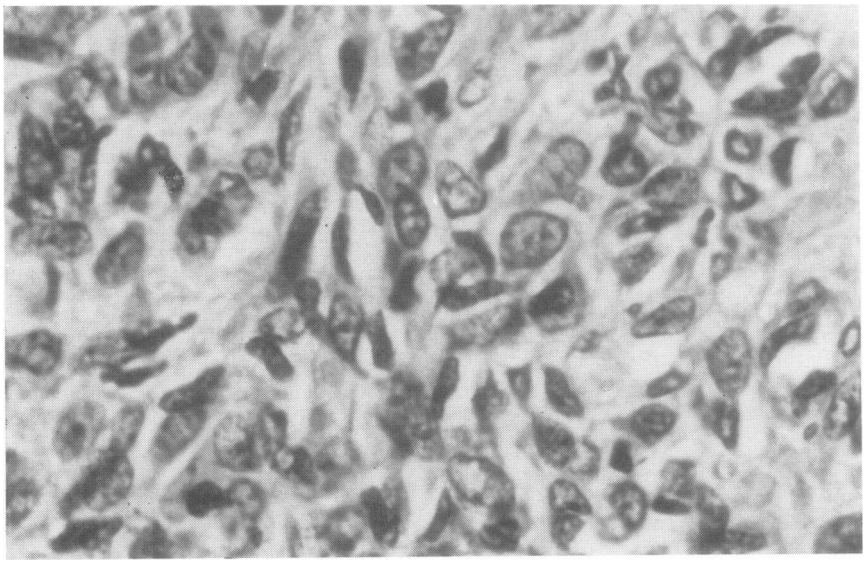

FIG. 2. *Tumour from 1952 biopsy showing the characteristic large oval and elongated nuclei and poorly defined cytoplasmic boundaries of the stromal cells. Small endothelial lined vessels are also present. Haematoxylin and eosin,* $\times 400$.

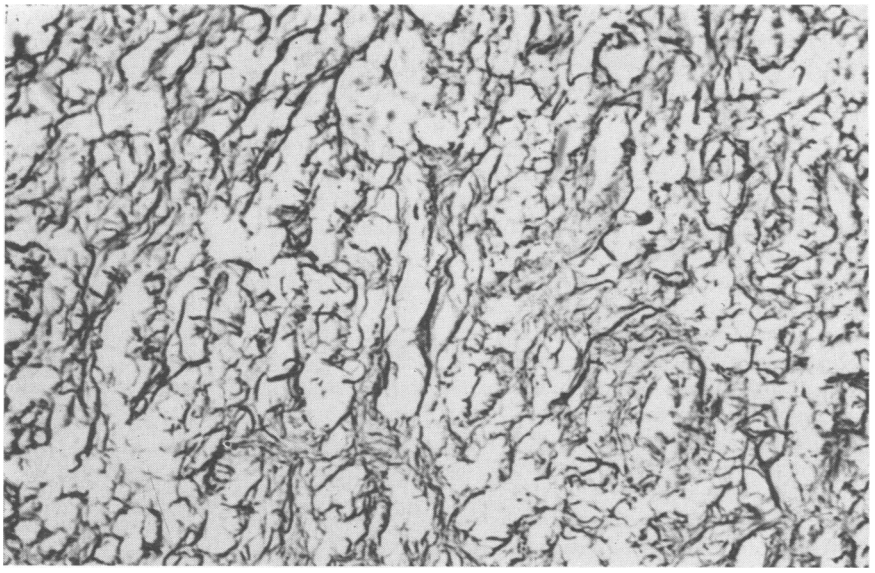

FIG. 3. *A prominent reticulum network is present about blood vessels and individual tumour cells. Biopsy, 1955. Foot-Foot reticulum, ×125.*

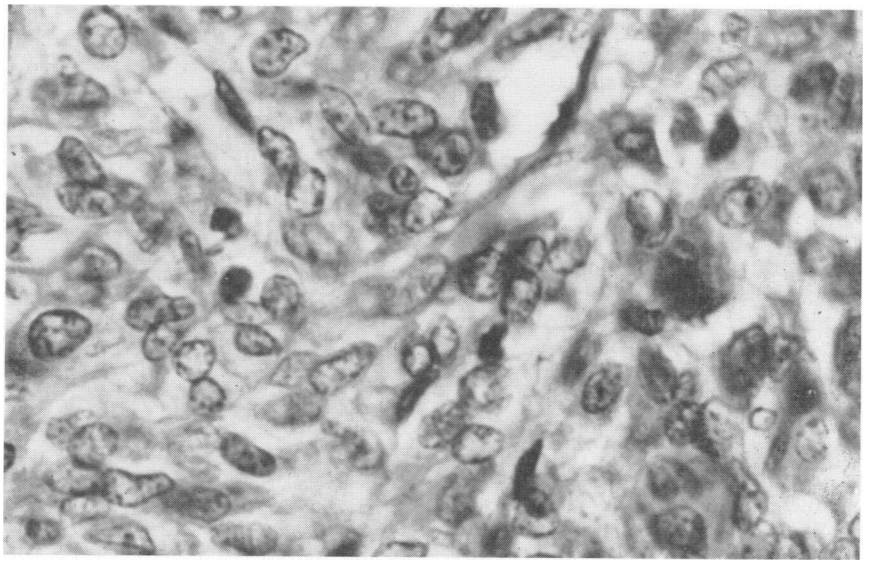

FIG. 4. *Tumour nodule in liver showing identical pattern with intracranial neoplasm with small endothelial lined vessels and characteristic stromal cells. Haematoxylin and eosin, ×400.*

and cerebellar hemispheres showed fibrosis and haemosiderin pigment in the meninges with reactive gliosis and granulation tissue in the underlying brain tissue.

COMMENT

The histological features of the meningioma resected at surgery and that found at necropsy in the dura mater and liver are characteristic of the angioblastic meningioma first described by Bailey, Cushing, and Eisenhardt in 1928. They later divided these tumours into three variants: variant I—angioblastic, variant II—transitional, and variant III—angioblastomatous (Cushing and Eisenhardt, 1938). Our meningioma is most similar to their first variant. In their review of 81 angioblastic meningiomas from the files of the Armed Forces Institute of Pathology, Pitkethly, Hardman, Kempe, and Earle (1970) classified the tumours into Cushing's three variants. They described those similar to variant I as 'hemangiopericytic', and found that these tumours behaved in a more malignant fashion, tended to recur locally, and were responsible for the few metastatic meningiomas in their series.

This type of meningioma has been considered by others to represent either a cerebellar haemangioblastoma or a haemangiopericytoma, thereby making statistical reviews difficult. For example, the metastatic intracranial neoplasm reported by Abbott and Love (1943) has been classified not only as a haemangioblastoma and angioblastic meningioma, but also as a haemangioendothelioma by different authors, and Kruse (1960) did not include it in his extensive review of metastasizing meningiomas.

It is generally accepted that a cerebellar haemangioblastoma may present a microscopic picture identical with an angioblastic meningioma, and they are usually differentiated by location. The fact that this patient's tumour is not a haemangioblastoma, even though occurring in the posterior fossa, is substantiated by its location at the time of operation. It was firmly attached to the tentorium, displaced by the cerebellum, and had no gross or microscopic evidence of continuity with cerebellar tissue. In addition, it was firm and noncystic.

Stout and Murray (1942) first differentiated the haemangiopericytoma from other vascular neoplasms, describing it as a tumour of the pericytes of Zimmerman. Since that time, there have been at least 12 published reports of haemangiopericytomas arising in the meninges (Stout, 1949; Begg and Garret, 1954; Fisher, Davis, and Lemmen, 1958; Kruse, 1961; McDonald and Terry, 1961; Stefanko and Glowacki, 1962), one of which metastasized to the vertebral bodies and femur. Stout (1942), Begg and Garret (1954), and other authors consider Cushing's original angioblastic meningioma to be, in fact, a haemangiopericytoma. Others, including Russell and Rubinstein (1963) and Pitkethly *et al.* (1970), feel the original concept should remain. Substantiating this latter view is the work of Muller and Mealy (1967). They grew numerous explants of a diagnosed meningeal haemangiopericytoma in tissue culture and from each culture the cells formed numerous delicate whorls, typical of those seen in meningioma tissue cultures. Electron microscopic examination has clarified the ultrastructural differences between haemangiopericytomas and haemangioendotheliomas (Ramsey, 1966). Such studies have not yet been reported for angioblastic meningiomas and might help resolve the controversy concerning the classification of these tumours.

In 1960, Kruse published a review of 20 reported cases that he accepted as metastasizing meningioma, adding two additional ones from the 803 cases of meningeal tumours at the Armed Forces Institute of Pathology. There have been 11 additional case reports of metastasizing meningiomas with sufficient detail to include in a statistical review (Abbott and Lowe, 1943; Zülch, Pompeu, and Pinto, 1954; Vlachos and Prose, 1958; Meredith and Belter, 1959; Hoye, Hoar, and Murray, 1960; Hukill and Lowman, 1960; Robertson, 1960; Noto and Gyori, 1961; Strang, Tovi, and Nordenstam, 1964; Gordon and Maloney, 1965; Opsahl, 1965). The present report brings the total number to 34. Of these, there have been five other probable angioblastic meningiomas (Abbott and Lowe, 1945; Zülch *et al.*, 1954; Meredith and Belter, 1959; Hukill and Lowman, 1960; Gordon and Maloney, 1965) with histological appearances similar to those of the present case and with evidence of metastases. Both Gordon and Maloney (1965) and Hukill and Lowman (1960) reported a case of an angioblastic meningioma and Hukill reclassified three previously reported meningeal tumours (Abbott and Lowe, 1943; Zülch *et al.*, 1954; Meredith and Belter, 1959) as angioblastic, as judged from the illustrations and descriptions. We would agree with this interpretation, and these five cases will therefore be considered as metastatic angioblastic meningiomas in this paper.

Thus, in reviewing the 33 reported metastasizing meningiomas, the transitional or meningothelial types occurred 15 times, the fibrosarcomatous and malignant 10, the fibroblastic four, and the angioblastic five. In general, the histological appearance of the meningioma can not be used to predict possible metastasis, since the histologically benign tumours metastasize as frequently as the malignant varieties. However, among angioblastic meningiomas,

Cushing's variant I appears more malignant than the other two types.

The mechanisms of spread outside the central nervous system have been discussed by previous authors, including Gyepes and D'Angio (1966). These authors feel that intracranial tumours metastasize via meningeal lymphatics or vascular channels, especially after surgery with the subsequent breakdown of the 'barrier to metastasize'. Meningeal tumours are ideally situated for lymphatic drainage and it is perhaps unusual that they do not do so with more frequency. It is of interest that approximately two-thirds of the metastasizing meningiomas occurred after operation. The present patient underwent four operations; tumour was found invading the transverse sinus 11 years before death.

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ADDENDUM

Since this manuscript was submitted for publication an additional case of a metastasizing meningioma has been reported. (Shuangshoti, S., Chaturaporn, H., and Netsky, M. (1970). Metastasizing meningioma. *Cancer*, **26,** 832-841).