Meningioma Following Radium Therapy

Case Report

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A case of an intracranial meningioma that developed at the site of a cutaneous vascular malformation treated by radium was recently observed. This unusual sequence of events aroused considerable speculation concerning the pathogenesis of the tumor.

Case Report

A 30-year-old right-handed woman entered the hospital on June 20, 1966, complaining of numbness of the right hand and forearm. This symptom had developed abruptly 2 days previously. Subsequently she experienced a throbbing left frontal headache together with inability to speak on several occasions. Difficulty in writing due to clumsiness in holding a pen had also been noted. Some time during the spring of that year consciousness had been lost for a brief period.

The patient had been born with an extensive vascular nevus affecting the left frontoparietal scalp and two smaller ones located over the right forehead and dorsum of the right foot. These were effectively treated at the age of 2 with radium, a fact verified by Dr. Joel Schweig who administered the treatment.

Examination. A right facial flattening, and sensory deficit involving the right arm comprised the pertinent neurological abnormalities. Position sense and stereognosis were defective in the right hand and pinprick and touch perception impaired below the elbow. Above the left ear, an area of scalp measuring 7 × 7 cm was atrophic and devoid of hair. An exostosis was evident within this area.

X-rays of the skull revealed a thinned vault on the left side (Fig. 1). Electroencephalography demonstrated theta and random delta activity in the left temporal and parietal areas. A left carotid arteriogram disclosed evidence of a tumor, most probably meningioma, in the suprasylvian region. The middle cerebral vessels were depressed, stretched, and displaced away from the inner table of the skull; a uniform stain appeared in the post-arterial phases (Fig. 2).

Operation. Craniotomy on June 29, 1966, confirmed the presence of a meningioma. A large flap was reflected well beyond the anticipated limits of the tumor so as not to jeopardize the viability of the overlying atrophic scalp. The tumor, somewhat larger than a golf ball, weighed 37 gm. It had grown through dura and eroded the overlying bone; its blood supply was derived largely from meningeal vessels. The tumor and the involved bone were completely removed. The wound healed uneventfully.

Microscopic Examination. The tumor showed the characteristic features of a meningioma, including uniform cells arranged
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Fig. 2. Left carotid arteriograms. Left: Anteroposterior view showing middle cerebral vessels depressed and posterior branches displaced away from inner table of skull. Right: Lateral view showing tumor stain still visible during venous phase.

in sheets, parallel rows, and whorls, and psammoma bodies (Fig. 3).

Postoperative Course. Following operation the patient exhibited a mild expressive aphasia, which rapidly cleared. The sensory deficit gradually lessened and, at the time of the last examination on May 16, 1967, was no longer evident.

Discussion

The interest in this case stems from a consideration of the relationship between the congenital vascular malformation of the scalp, its treatment by irradiation, and the subsequent occurrence of a meningioma directly beneath the site of the original cutaneous lesion. Admittedly, the two lesions may be unrelated, their proximity being purely fortuitous. While this interpretation may be correct, the course of events in this case arouses skepticism and invites speculation. The combination of a cutaneous nevus and intracranial tumor is reminiscent of a number of congenital disorders characterized by lesions affecting both skin and nervous system, such as tuberous sclerosis, von Recklinghausen’s neurofibromatosis, neurocutaneous melanosis, and the Sturge-Weber syndrome. Much has been written concerning these dysgenetic disorders and the pleomorphic nature of their manifestations. Cutaneous vascular nevi are one of the components of the Sturge-Weber syndrome and also occur commonly in association with pigmented nevi in neurofibromatosis. Our case, however, does not fulfill the criteria for the diagnosis of any of these disorders. An abortive form of neurofibromatosis might be considered, although the temporal relationship of the lesions and failure of other stigmata of von Recklinghausen’s disease to appear in the interim are strong evidence to the contrary.

Fig. 3. Section of tumor displaying whorls and psammoma bodies. H. & E., ×400.
Finally, the question arises concerning the role of irradiation in the development of the meningioma. There are a number of reports suggesting that sarcomas may occur as a consequence of radiation therapy. The appearance of such tumors following therapeutic irradiation has been observed within brain tissue, in the meninges, and in conjunction with chromophobe adenomas of the pituitary. We are aware of only two papers dealing with meningiomas induced by a radioactive substance or developing after x-ray treatment. Kyle, et al., reported a case of sphenoid wing meningioma in a young adult following thorium dioxide ventriculography 24 years previously. Examination of the neoplasm disclosed “a large number of cells containing pleomorphic hyperchromatic nuclei.” The tumor recurred, and the tissue removed at a second operation was malignant. The occurrence of a meningioma of low-grade malignancy in a 5-year-old child who had been subjected to radiotherapy of the scalp (2000 r) was described by Horányi. To the best of our knowledge there is no unequivocal report of a truly benign meningioma induced by irradiation, nor have we observed this phenomenon previously. For this reason we are reluctant to assume that in the case we have reported, radium was the responsible factor. Nevertheless, the evidence is suggestive and hence this case has been recorded to raise the question of a causal relationship.

Summary

We have reported a case in which a meningioma appeared 28 years after radium therapy for a vascular malformation of the scalp. The tumor developed directly beneath the site of treatment of the cutaneous lesion.

References

3. Rosomoff, H. L. Personal communication.