Suprasellar osteogenic sarcoma following radiation for pituitary adenoma

Case report

ABDUL R.C. AMINE, M.D., AND OSCAR SUGAR, M.D.

Department of Neurosurgery, Abraham Lincoln School of Medicine of the University of Illinois College of Medicine, Chicago, Illinois

The authors report a case in which sellar and suprasellar osteogenic sarcoma developed 10 years after radiation therapy for pituitary adenoma. The literature is reviewed and the rarity of this condition discussed.

**KEY WORDS** • osteogenic sarcoma • pituitary adenoma • radiation effects

The development of malignant neoplastic lesions in the sellar and suprasellar region following betatron radiation for benign pituitary chromophobe adenoma is very rare. We are presenting a case of suprasellar osteogenic sarcoma that developed 10 years after the discovery and surgical and radiotherapeutic treatment of a pituitary tumor in a 16-year-old girl.

**Case Report**

This girl was 16 years old when first admitted to the University of Illinois Hospital in May, 1964, because of progressive loss of vision over a 2-year period. She was known to have delayed secondary sexual development and had not yet started her menstrual cycle. She had also had intermittent severe headaches. The patient's own past and family history were noncontributory.

*First Admission.* The physical examination was within normal limits except for failure of the patient to develop secondary sexual characteristics normal for her age. Neurological examination revealed normal walking ability, with slight difficulty in tandem gait. The olfactory nerves were intact. Extraocular movements were normal, with some difficulty in fixation of the right eye. The right eye was totally blind, and there was a temporal hemianoptic field defect in the left eye. The pupils reacted to light only on the left. The other cranial nerves and the remainder of the neurological examination were essentially within normal limits. Anteroposterior skull x-ray films revealed an enlarged sella turcica with erosion of its floor. A right brachial angiogram revealed displacement of the carotid siphon, presumably secondary to pituitary tumor.

*First Operation.* A right frontal craniotomy was performed with subtotal removal of a chromophobe adenoma (Fig. 1). Post-
operatively the patient did well. Betatron radiation therapy was administered on a 5 day/wk basis with the first six treatments consisting of 250 rads/day followed by 18 treatments of 200 rads/day, through a 5-cm field centered on the pituitary region. The patient was given a total of 5100 rads in 33 days. She was maintained on daily cortisone and thyroid preparations, and was followed in the outpatient clinic until lost to follow-up after 1968.

In April, 1974, at the age of 26, the patient was admitted to a local hospital because of stupor and difficulty in arousal; her condition had been deteriorating for the previous 2 months. On examination she was found to be lethargic; she responded to pain by withdrawal and some slurred verbal response. Lumbar puncture was considered abnormal, with an opening pressure of 340 mm H2O.

Second Admission. She was transferred to the University of Illinois hospital on April 30. On examination she responded to painful stimuli by appropriate defensive movements and mumbling. She had a markedly stiff neck; her ocular movements were intact, the pupils were equal but neither reacted to light. Funduscopic examination revealed severe bilateral optic atrophy. The deep tendon reflexes were hypoactive but symmetrical. The remainder of the neurological examination that could be done was normal, as was the general physical examination, except for obesity. Blood pressure, pulse, respiration, and temperature were normal. Spinal puncture was done to rule out subarachnoid hemorrhage and meningitis; it showed an opening pressure of 270 mm H2O. The fluid was slightly xanthochromic but clear; there were 8/cu ml red blood cells and 62/cu ml white blood cells, with 34% polymorphonuclear cells and 66% lymphocytes. Protein content was 290 mg% and glucose 70 mg%. The plain skull films did not reveal any changes from those taken in 1968. Right brachial and left carotid arteriograms were performed. The lateral view showed a huge sellar and suprasellar tumor that was found. The consistency of the tumor material was gritty and semisolid, somewhat like that of a meningioma. A large portion of the intrasellar mass was removed. The tumor mass extended into the brain, and no attempt was made to remove all of the tumor.

Histological Examination. Histologically the suprasellar mass was extremely cellular (Fig. 2); the cells showed striking pleomorphism with numerous bizarre giant cell forms and many mitotic figures. In some areas the cells appeared more regular with large well-defined eosinophilic cytoplasm and fairly uniform vesicular nuclei. There was no evidence of osseous tissue deposition. The intrasellar mass was represented by a malignant-appearing spindle-cell parenchyma that formed osteoid tissue or bone (Fig. 3). The cells were extremely pleomorphic, spindle-shaped, oval or polyhedral, with dark hyper-
Specimen from the suprasellar portion of the tumor showing striking pleomorphism with numerous bizarre giant cell forms and mitotic figures. There is no evidence of osseous tissue formation in this portion. H & E, × 256.

Postoperative Course. The patient received cobalt radiation treatment consisting of 3000 rads, one treatment of 400 rads, and the remaining 13 of 200 rads/day within 3 weeks, in the hope that this malignant osteogenic sarcoma might respond to radiation, but she died about 5 weeks after surgery.
Suprasellar osteogenic sarcoma following radiation

Discussion

The incidence of primary intracranial sarcoma does not exceed 1.2% in large series; it is usually found in infants and small children.

The development of malignant sarcoma in the brain following radiation to the brain has been recorded, particularly in the meninges over the site of a glial tumor. It is much less common than primary sarcoma. The average interval between the time of the initial radiation and the development of sarcoma is from 5 to 10 years, but it may range from 2 to 20 years.

Waltz and Brownell reported three cases of sarcoma following irradiation to pituitary adenoma, and found 10 other cases in the literature. Meredith, et al. reported a case of osteogenic sarcoma arising in the temporal region of the skull 6 years after irradiation of a chromophobe pituitary adenoma.

Osteogenic sarcoma of the skull secondary to irradiation is very rare. It is also rare as a primary tumor of the skull, particularly in patients who do not have Paget’s disease.

We have not been able to find reports of a similar case of osteogenic sarcoma in the suprasellar region following irradiation to a benign pituitary adenoma.

References


Address reprint requests to: Abdul R. C. Amine, M.D., Department of Neurosurgery, University of Illinois Hospital, 912 South Wood Street, Chicago, Illinois 60612.