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Clinical Study

Management of central nervous system teratoma

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ABSTRACT

Central nervous system (CNS) teratomas are very rare neoplasms that contain tissues derived from all three germ cell layers (endoderm, mesoderm, and ectoderm). Patients with teratomas usually have a good prognosis. Given the paucity of cases in the literature, we present a retrospective review of 15 CNS teratomas treated over a 25 year period at the University of California, San Francisco. We describe the presentation, location, treatment, and adjuvant therapy for these patients, and highlight three unique cases that emphasize the diverse presentation and treatment of these rare tumors.

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1. Introduction

Central nervous system (CNS) teratomas are very rare neoplasms that include mixed and malignant subtypes and account for approximately 0.5–1% of primary adult intracranial tumors. There is a higher frequency in children (~7%) and in countries such as Japan, Korea, and Taiwan (1.8–5% in adults, up to 15% in children) [1]. Teratomas are a subset of nongerminomatous germ cell tumors (that also include embryonal carcinomas, choriocarcinomas, and yolk sac/endodermal sinus tumors), and are unique in that they are composed of tissues derived from all three germ cell layers (endoderm, ectoderm, and mesoderm). In the CNS, they usually arise in midline locations and are more common in the pineal region, suprasellar cistern, basal ganglia, and thalamus [2], with rare examples in other locations such as the temporal lobe [3], cerebellum [4], lateral ventricles [5], or posterior fossa [6,7]. In the spine, they are more often extramedullary [8], and are more often found at thoracolumbar levels [8,9]. Unlike other germ cell tumors, teratomas usually have a good prognosis, with 5 year survival rates ranging from 87 to 100% for mature teratomas [1,10,11] and 33 to 71% for malignant teratomas [1,11].

Given their rarity, there are relatively few studies of CNS teratomas in the literature. Besides the limited case reports of congenital teratomas [12–18], to our knowledge there are only two large case series, one with 14 pediatric patients with intracranial teratomas [1], and another with 31 patients with intracranial teratomas in South Korea [2]. In this paper, we present a retrospective review of 15 patients with CNS teratomas treated at the University of California, San Francisco, over a 25 year period, from 1982 to 2007.

2. Methods

A retrospective review was performed of all patients undergoing surgery for resection of a teratoma at the University of California, San Francisco (UCSF) between 1982 and 2007. A list of all patients undergoing resection for a brain or spinal tumor was generated from a database containing all operative procedures, and cross-referenced with a pathology database to identify all patients with histopathologically confirmed teratomas. Identifying patient characteristics, including name, diagnosis, and tumor location were prospectively collected in these databases for all consenting patients undergoing neurosurgical evaluation at UCSF; this occurred in accordance with the Committee for Human Research (CHR# H7828-29842-01).

The medical records, radiographic imaging, pathology reports, and operative notes for each of these patients were reviewed. Data collected included patient demographics, preoperative symptoms, surgical approach, tumor location, extent of resection, postoperative symptoms, adjuvant therapy, tumor recurrence, and years of follow-up. Standard pathologic examination (gross and microscopic examination) was previously performed on all tumor spec-

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imens. The diagnosis of teratoma was established by the presence of endodermal, mesodermal, and ectodermal tissues. Immunohistochemical stains were performed if needed to aid in highlighting particular components of the teratoma in a limited sample or to exclude other diagnoses. The typical diagnostic challenge is in excluding components of other germ cell neoplasms (i.e. mixed germ cell tumor), including immature teratoma, yolk sac tumor, germinoma, embryonal carcinoma, and/or choriocarcinoma.

2.1. Illustrative Patient 1

Our first case was a 10-year-old boy with no prior medical history who presented with 2 weeks of headaches, dizziness, emesis, and a generalized tonic-clonic seizure. A ventriculoperitoneal shunt was placed at an outside hospital for treatment of his hydrocephalus (Fig. 1A, B). For definitive treatment of his third ventricular/pineal region mass, a bifrontal craniotomy using a transcallosal approach was performed and a gross total resection was achieved. The patient experienced no postoperative complications and was discharged home neurologically intact 4 days after surgery without any new neurologic deficits.

At the time of surgery, the tumor was noted to be a reddish, irregular semi-solid mass, with areas of hair, calcification, and cyst. Gross examination of the specimen showed a multiloculated cystic mass, composed of skin, hair, hard bony tissue, and filled with tan-gelatinous material to clear serous fluid. A few separate fragments of tan-brown to red-gray tissue were also present. The gross and microscopic findings were compatible with a mature teratoma. No microscopic description was provided in the pathology report, and the slides are no longer available for re-review. Five years later, a new pineal region mass with extension along the right ambient cistern and vermis was identified on follow-up MRI. At this time, the patient had no neurologic symptoms, but slight limitation of upgaze bilaterally with convergence-retraction nystagmus was noted. Serum and cerebrospinal fluid (CSF) markers for beta-human chorionic gonadoptropin (beta-hCG) and alpha-fetoprotein (AFP) were negative, and the CSF cytology was negative. The patient had an MRI-guided biopsy, which showed a low-grade astrocytoma. Because of its rapid growth, the tumor was treated as an anaplastic astrocytoma and the patient received focal radiotherapy to a total of 54 Gy, with good response.

2.2. Illustrative Patient 2

In this case, a teratoma was identified in a more unusual location. A 52-year-old man was known to have an intracranial mass for 10 years, and was followed at another institution without definitive treatment. He then presented with headaches, nausea, vomiting, dizziness, hyponatremia, and a possible seizure. An imaging study showed a 4×3 cm contrast enhancing heterogeneous mass in the left temporal lobe (Fig. 2A–D). At the time of admission, the patient was drowsy but awakened easily to voice, was oriented and briskly followed commands. However, he became increasingly lethargic while awaiting surgery. A craniotomy was performed with subtotal resection of the mass. Postoperatively, the patient's neurologic status deteriorated, and an

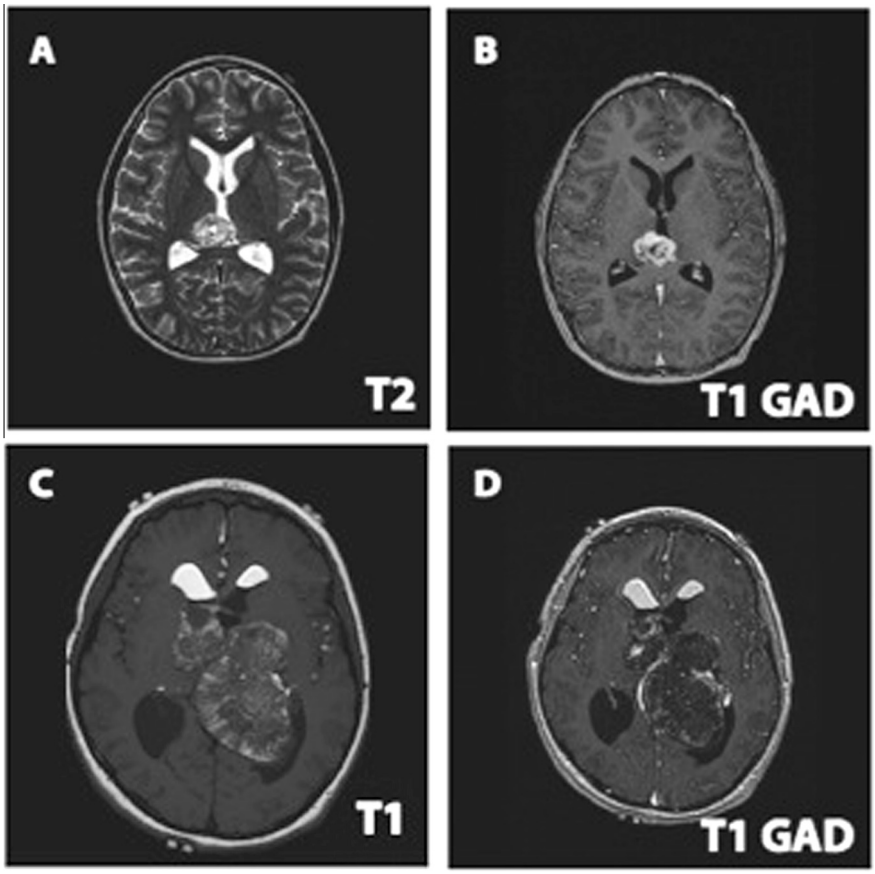

Fig. 1. Teratomas of the pineal region. Axial (A) T2-weighted and (B) T1-weighted with gadolinium (GAD) contrast MRI of a heterogeneous mass of the third ventricular/pineal region (similar in size and location to that described in Illustrative Patient 1, whose images were obtained before 1990 and are no longer available). Axial (C) T1-weighted and (D) T1-weighted with gadolinium contrast MRI showing a multilobular teratoma that is also centered at the third ventricle, but spreads out much further. In this particular case, the teratoma had ruptured into the ventricular system, and the hyper-intense signal in the frontal horns represents dermoid-like material in the ventricles.

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