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Two-stage surgical resection of an atypical teratoid rhabdoid tumor occupying the infratentorial and supratentorial compartment in children under two years: Report of two cases



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ABSTRACT

INTRODUCTION: Atypical teratoid rhabdoid tumors are highly malignant neoplasms that present in young children and can grow to a large size. Maximal safe surgical resection is a mainstay of treatment.

PRESENTATION OF CASES: Two cases of children under the age of two with large tumors involving the supratentorial and infratentorial compartments are presented. A two-staged operative approach combining a standard suboccipital approach to the fourth ventricle followed by an infratentorial, supracerebellar approach was utilized for resection.

DISCUSSION: Maximal safe surgical resection of large tumors in young children is challenging. A staged approach is presented that affords maximal tumor resection while minimizing perioperative morbidity. **CONCLUSION:** A staged operative approach appears safe and efficacious when resecting large tumors from both the infratentorial and supratentorial compartments in children less than two years of age.

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1. Introduction

Atypical teratoid rhabdoid tumors (ATRT) are rare, highly malignant embryonal tumors of the central nervous system (CNS) primarily occurring in young children with an overall age-adjusted incidence of 0.07 per 100,000 [1]. While they represent only 1.6% of all brain and CNS tumors diagnosed in the pediatric population, ATRTs represent 10.1% of tumors diagnosed in patients less than 1 year of age [1]. Although the overall survival remains poor, with most patients living less than 1 year, aggressive multi-modality therapy has improved survival [1–4]. Extent of surgical resection has been identified as a significant prognostic factor in the treatment of these aggressive tumors [2,4–6]. However, maximal safe resection of large tumors in very young children can be challenging due to technical factors, blood loss, and prolonged anesthesia.

Two cases of large ATRTs occupying both the infratentorial and supratentorial compartments in children less than two years of age are presented.

2. Presentation of cases

2.1. Case 1

An 18 month-old girl presents to an outside hospital with a history of vomiting and acute onset altered mental status. On exam, she was lethargic and unresponsive, but withdrew to painful stimuli. A computed tomography (CT) was obtained demonstrating a large posterior fossa mass with hemorrhage and hydrocephalus (Fig. 1). She was then intubated and transferred to Children's of Alabama (COA).

On arrival, she was a Glasgow Coma Scale (GCS) 4 with a right gaze preference, reactive pupils, absent left corneal reflex, and absent cough and gag reflex. An external ventricular drain (EVD) was placed at bedside and the patient taken emergently to the operating room (OR).

Patient was positioned prone on the operating table for a suboccipital craniotomy for hematoma evacuation and tumor debulking. A skin and fascial incision was performed, exposing the occiput, posterior ring of C1, and the top of the C2 lamina. A high-speed drill was then used to perform the occipital craniotomy and C1 laminectomy. Dura and arachnoid were then opened and tumor immediately encountered (Fig. 2A). Tumor resection proceeded rostrally through the fourth ventricle and into the third ventricle (Fig. 2B). Blood loss at that time was approximately 150 cc; the patient began to experience episodes of hypotension and the decision was made to stop the operation. Dura was closed using an expansile patch and bone was replaced. The wound was

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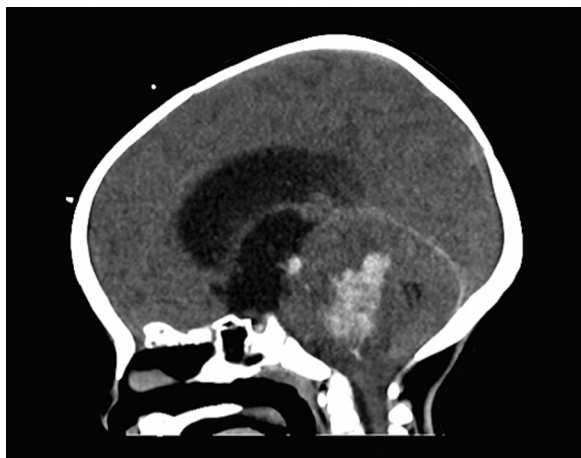


Fig. 1. Sagittal CT head demonstrating a large, hemorrhagic posterior fossa mass filling the fourth ventricle with associated hydrocephalus and brainstem compression.

closed and she was transferred to the intensive care unit (ICU) in stable condition. Postoperative magnetic resonance imaging (MRI) demonstrated subtotal resection of a very large infratentorial tumor with extension through the tentorial incisura into the supratentorial compartment. Histopathologic examination of tissue from this procedure revealed atypical teratoid rhabdoid tumor (ATRT).

The patient made an excellent recovery and was extubated on postoperative day 5. Given the large remaining tumor burden, plans were made for a repeat craniotomy for completion of the posterior fossa tumor resection combined with a supracerebellar, infratentorial approach to residual tumor mass. Incision was opened, but this time the bone removal was extended superiorly exposing the transverse sinus and confluence of sinuses. The dura was again opened and additional tumor removed from the fourth ventricle and cerebellopontine angle (CPA). Attention was then turned to the supracerebellar surface. After coagulating and dividing bridging veins between the tentorium and cerebellum, the space was opened revealing extensive amounts of tumor. Resection continued rostrally, dissecting tumor from the deep venous structures, skeletonizing the vein of Galen, and eventually exposing the atrium of the lateral ventricle. Tumor resection continued until pia and choroidal fissure were identified. Navigation was then used to confirm that all accessible tumor had been removed. Once again, an expansile duraplasty was performed with replacement of the craniotomy flap. Patient was extubated at the end of the procedure and returned to the ICU at her neurologic baseline. Postoperative MRI demonstrated near complete resection of residual tumor.

The patient's post-operative course was significant for hydrocephalus, treated with ventriculoperitoneal shunting. She made a full recovery, and underwent ablative chemotherapy with autologous stem cell rescue followed by radiation.

2.2. Case 2

A 22-month old male presents to an outside hospital with gait instability, nausea, and vomiting. On exam he was neurologically intact except for an ataxic gait. A CT was obtained demonstrating a large posterior fossa mass and hydrocephalus. He was transferred to COA. On initial evaluation, he was alert and interactive but developed lethargy, bradycardia and hypertension over the subsequent 24 h. An EVD was placed at bedside with clinical improvement. A MRI was obtained the following day revealing a large posterior fossa mass with extension through the tentorial incisura into the supratentorial compartment with involvement of the tectum (Fig. 3A). Plans were made for a two-staged surgical resection.

For the first stage, patient was positioned prone on the operating table and incision, dissection, and bone removal was similar to that previously described. Tumor was evident between the cerebellar tonsils upon opening of the dura and arachnoid. Tumor resection proceeded rostrally to the level of the aqueduct. At this point the tectum was noted to be engulfed in tumor. During microdissection of tumor off the dorsal mesencephalon, bleeding was encountered from a traversing vein. Additionally, the patient experienced a bout of hyperthermia coincident with an intraoperative blood transfusion and the decision was made to stop the operation. Hyperthermia resolved with acetaminophen and ice packs. Patient was closed with an expansile duraplasty with replacement of the craniotomy flap. Temperature normalized by the conclusion of the case and patient awoke at his neurologic baseline. Postoperative MRI demonstrated a large amount of residual tumor in the quadrigeminal cistern and extending superiorly through the tentorial incisura with near complete resection of the fourth ventricular aspect (Fig. 3B). Histopathologic examination again revealed ATRT.

As planned, the patient was brought back to the operating room on postoperative day 6 for a redo craniotomy for a supracerebellar, infratentorial approach. The supracerebellar space was entered as previously described (Fig. 4A and B). Tumor was apparent encasing the deep venous structures. Using microsurgical dissection, soft tumor was removed from the venous complex. After sacrifice of the precentral cerebellar vein to improve access to the midline, the resection was carried down to and stopping short of the tectum. At this point, near-complete resection had been accomplished and the decision was made to stop. The case was closed as previously described. Patient awoke moving all extremities and returned to the ICU. As the anesthetic effects cleared, he was noted to have right cranial nerve VI and VII palsies in addition to truncal ataxia and

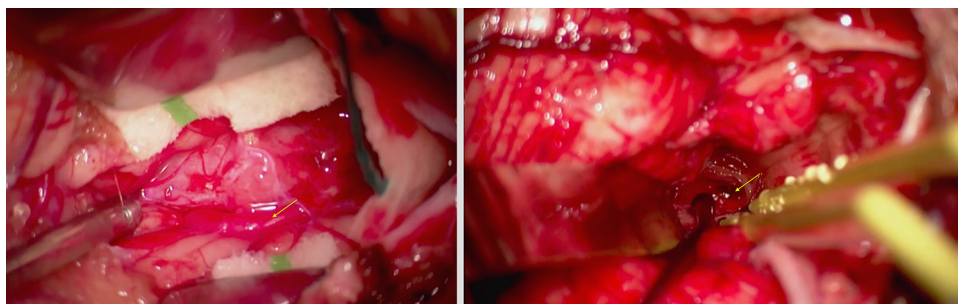


Fig. 2. Intraoperative photographs from Case 1—suboccipital, infracerebellar approach for tumor resection and hematoma evacuation. (A) Tumor extruding from the fourth ventricle upon opening of the dura and arachnoid (Arrow notes left PICA). (B) Following tumor resection and hematoma evacuation, a rostrally oriented view of the fourth ventricle and aqueduct (Arrow at cerebral aqueduct).

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