Operative Technique: Angiomatoid Fibrous Histiocytoma—Unique Case and Management

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Abstract

Objective We describe the first jugular foramen angiomatoid fibrous histiocytoma (AFH) case and the first treatment with preoperative endovascular embolization. AFH is a rare intracranial neoplasm, primarily found in pediatric patient extremities. With an increase in AFH awareness and a well-described genetic profile, intracranial prevalence has also subsequently increased.

Study Design We compare this case to previously reported cases using PubMed/Medline literature search, which was performed using the algorithm “[intracranial” AND “angiomatoid fibrous histiocytoma]” through December 2020 (23 manuscripts with 46 unique cases).

Patient An 8-year-old female presented with failure to thrive and right-sided hearing loss. Work-up revealed an absence of right-sided serviceable hearing and a large jugular foramen mass. Angiogram revealed primary arterial supply from the posterior branch of the ascending pharyngeal artery, which was preoperatively embolized.

Intervention Gross total resection was performed via a translabyrinthine approach.

Conclusion The case presented is unique; the first reported AFH at the jugular foramen and the first reported case utilizing preoperative embolization. Preoperative embolization is a relatively safe technique that can improve the surgeon’s ability to perform a maximally safe resection, which may decrease the need for adjuvant radiation in rare skull base tumors in young patients.

Introduction

Angiomatoid fibrous histiocytoma (AFH) is a rare neoplasm. It primarily occurs in the soft tissues of extremities.1 AFH can rarely occur as a primary lesion in the intracranial space.2 The neoplasm predominantly affects adolescents and young adults with an average age of 14 years at diagnosis.3 Considered a benign neoplasm with low metastatic potential, AFH does exhibit a high recurrence rate4 and a high rate of associated hemorrhage.4–9 Treatment is principally through...
surgical resection. It is thought that there is a spectrum of soft tissue tumors involving the female-expressed transcript-cAMP-response element-binding (FET-CREB) protein fusion that includes AFH.3 The naming convention will change in the next World Health Organization classification system to intracranial mesenchymal tumor, FET-CREB fusion positive.10 There are rare case reports and case series describing the intracranial occurrence of AFH.2–9,11–24

Intracranial AFH is most commonly associated with the meninges, usually located along the cerebral convexities or in the intraventricular space.3 Similar to most intracranial masses, AFHs present with local mass effect or obstructive hydrocephalus.3 Due to their rarity, they are often not considered in the differential diagnosis of extra-axial lesions in young patient populations. The differential diagnosis often includes more common tumors with a similar appearance, including meningioma and solitary fibrous tumor.

In this case, we present the first reported AFH located at the jugular foramen with a unique treatment plan involving preoperative transarterial embolization for devascularization to aid in tumor resection. We also provide a literature review to compare reported cases in the literature in an effort to better elucidate optimal treatment strategies.

**Materials and Methods**

**Clinical Presentation**

An 8-year-old female, with a past medical history of asthma, mild developmental delay, pseudostrabismus, and rare headaches, presented to our emergency department (ED) after approximately 2 weeks of worsening nausea and vomiting with poor oral intake. Two days prior to the presentation, the child had complained about a tingling sensation in her right eye. She was initially taken to an outside hospital’s ED, where a brain magnetic resonance image (MRI) revealed an extra-axial right posterior fossa mass abutting the petrous temporal bone. The patient was transferred to our pediatric intensive care unit (PICU;► Fig. 1).

Upon examination, she had end gaze nystagmus in all directions, right upper and lower facial weakness (complete eye closure with significant effort and nasolabial flattening), and decreased right-sided hearing. She was started on dexamethasone. Skull base computed tomography (CT) was obtained with no obvious evidence of skull involvement or widening of skull base foramen (► Fig. 2). An audiogram was obtained with profound right-sided hearing loss (► Fig. 3). She underwent a diagnostic cerebral angiogram, which demonstrated major arterial supply from the right ascending pharyngeal artery (APA) (► Fig. 4). Her exam improved to mild right facial weakness at rest with symmetry upon activation. This improvement was attributable to steroid intake. She was transferred to the ward while awaiting a surgical date. The day prior to surgery, she underwent an ophthalmological examination with normal findings. Standard of care surgical consent was obtained. Institutional Review Board approval was not required for single cases. An institutional authorization to use and disclose protected health information was obtained.

**Operative Technique**

On the day prior to surgery, she underwent an angiogram for particle and coil embolization of the right APA (► Fig. 5). A 4 French Pinnacle Introducer Sheath (Terumo, Somerset, NJ, United States) was placed in the right femoral artery. A 4 French Tegtmeyer catheter was navigated over a 0.035" Glidewire (Terumo, Somerset, NJ, United States) into the common carotid artery. Using roadmap imaging, the right APA was selectively catheterized. Next, a 1018 Excelsior microwire catheter (Stryker, Fremont, CA, United States) was navigated over a Fathom microwire (Boston Scientific, Marlborough, MA, United States) to superselectively catheterize the distal posterior branch of the APA. Embolization was performed using 355 to 500-micron polyvinyl alcohol

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**Fig. 1** Preoperative MRI showing a right jugular foramen heterogenous mass with solid and cystic components that appear hypervascular. (A) Axial T2. (B) Axial balanced fast field echo. (C) Coronal T1 with contrast. (D) Sagittal T1 with contrast.

**Fig. 2** Preoperative CT temporal bone protocol. No evidence of bone remodeling or osseous involvement.
particles (Cook Medical, Bloomington, IN, United States) followed by a 2 mm × 5 mm figure-eight coil (Boston Scientific, Marlborough, MA, United States). The microcatheter was removed under gentle aspiration. A final angiographic run revealed embolization of the feeding artery and a significant reduction of flow to the lesion.

The next day, she went for surgical resection (►Video 1). She underwent a right translabyrinthine approach for the resection of mass with an abdominal fat graft for closure. The patient was monitored with somatosensory evoked potentials, motor-evoked potentials, facial nerve electromyography, and electroencephalography. A neurotologist performed the surgical approach and closure, while a neurosurgeon performed the durotomy and resection. Intraoperatively, the mass was readily identifiable. There was a clear plane between the tumor and the cerebellum. Due to neoplasm size, internal bulking was required. The tissue had portions that were soft, while other portions were firm, requiring ultrasonic aspiration with an aggressive bit (CUSA Clarify, Integra, Princeton, NJ, United States) and dissection with microscissors (Kamiyama series, Takayashi Instrument, Inc, Natick, MA, United States). Frozen specimen revealed a low-grade neoplasm with a differential diagnosis including hemangioblastoma, meningioma, and schwannoma with low suspicion for papillary endolymphatic sac tumor. The mass was dissected off the trigeminal nerve and cranial nerves VII/VIII complex and found to likely originate from the jugular foramen as it was intimately involved with the lower cranial nerves (IX/X) with primary attachments to the petrous temporal bone around the jugular foramen. A small amount of residual tumor was left due to dense adherence to the lower cranial nerves. No changes were noticed in neuro-monitoring throughout the case including intact stimulation of the facial nerve. The patient went directly to MRI to obtain both a safety postprocedural scan and a postresection baseline brain scan. This MRI revealed a gross total resection with no obvious complication or radiographically residual disease (►Fig. 6).

| Frequency (Hz) | 250 | 500 | 1,000 | 2,000 | 3,000 | 4,000 | 6,000 | 8,000 |
|---------------|-----|-----|--------|--------|--------|--------|--------|--------|
| Right ear dB HL (masked) | NR up to 90 | NR up to 100 | NR up to 100 | NR up to 100 | NR up to 100 | NR up to 100 | NR up to 90 |
| Left ear dB HL | 15 | 15 | 10 | 5 | 5 | 5 | 0 | 0 |

Fig. 3 Preoperative audiometry showing no serviceable hearing on the affected side.

Fig. 4 Preoperative digital subtraction angiography. (A) Injection of the right common carotid artery during the arterial phase showing tumor blush from the posterior branch of the ascending pharyngeal artery. (B) Microcatheter injection of the ascending pharyngeal artery in the arterial phase.
Postoperative Management

The patient was extubated in the postanesthesia care unit and transferred to the PICU. She was noted to have some dysarthria and coughing with difficulty in swallowing postoperatively. She was evaluated by speech therapist who offered suggestions on safe swallowing techniques with a plan for regular diet and thin liquids. She was transferred to the stepdown unit on postoperative day (POD) 1. She was discharged home with her parents on POD 2. Her final histological diagnosis returned as AFH, positive for EWSR1 rearrangement by fluorescence in situ hybridization analysis (►Figs. 7 and 8). At a 2-week postoperative follow-up, her facial weakness had resolved. She underwent a full body positron emission tomography computed tomography (PET-CT) with no evidence of disease. At a 10-week follow-up, her diplopia had resolved. She was noted by her mother to cough throughout the day but had no difficulty swallowing and was on a regular diet. Brain MRI showed no progression of known residual disease. At a 5-month follow-up, she remained well with no progression on surveillance brain MRI with no further coughing. At a 9-month follow up, surveillance brain MRI noted disease progression. She showed no clinical signs of worsening disease. She was referred to radiation oncology for consideration of adjuvant radiotherapy with initial plan for 55.8 Gy over 31 fractions.

Literature Review

A systemic PubMed/Medline literature search was performed using the algorithm [“intracranial” AND “angiomatoid fibrous histiocytoma”] through December 2020. We reviewed the search results for intracranial cases of histologically proven AFH. Twenty-three manuscripts were found with 46 unique cases (►Table 1). We recorded patient age and gender, symptomatology at presentation, intracranial...
The lesion demonstrates a nodular proliferation with blood-filled pseudoangiomatous spaces and abundant hemosiderin deposition. The characteristic lymphoplasmacytic cuff often seen in this entity is absent in this case (x40 magnification). (B) Syncytial growth of bland spindled to epithelioid tumor cells; cytologic atypia is minimal, and mitotic figures are inconspicuous (x200 magnification). (C) The lesional cells show focal positivity for CD99 (x200 magnification) and (D) strong, diffuse staining for desmin (x200 magnification).

Discussion

AFH is a rare tumor that can infrequently appear intracranially. The tumor has rarely been reported in the skull base. There have been four reported cases of this tumor in the cerebellopontine angle (CPA) and an additional case at the petrous apex (Table 1). We report only the sixth case of a skull base AFH and the first at the jugular foramen. Alshareef et al described a case at the petrous apex in which the patient presented with right facial weakness, pain, and numbness. The first consideration on their differential diagnosis was a trigeminal schwannoma. The tumor was approached through a petrosal craniotomy with one-piece orbitozygomatic craniotomy. They were able to perform a gross total resection. The patient did develop a delayed cerebrospinal fluid leak from petrous apex air cells that was repaired. There was no evidence of recurrence on 6-month postoperative MRI.

Sloan et al reported four CPA masses in their series of 20 cases. Three of the four patients with CPA masses underwent a subtotal resection, and the final was a gross total resection. The surgical approach to these tumors was not described. One of the patients with subtotal resections underwent adjuvant radiation and chemotherapy. Unfortunately, they had metastases of their disease and passed away 27 months postoperatively. The second patient with subtotal resection did not undergo adjuvant therapy with ultimate progression of their disease. They passed away 1 month postoperatively. The final patient with subtotal resection has not undergone adjuvant therapy and remains alive at 13 months postoperatively with stable disease. The gross total resection has had local recurrence without adjuvant therapy. They remain alive at 30 months postoperatively.

Our case is unique in that it is the first described AFH case at the jugular foramen. The patient presented with multiple cranial nerve abnormalities and symptoms concerning for increased intracranial pressure. Imaging was concerning for an extra-axial mass along the petrous bone concerning for meningioma, schwannoma, solitary fibrous tumor, endolymphatic sac tumor, or other rare pediatric tumor. None of these diagnoses seemed to fit based on patient demographics, patient presentation, and radiographic appearance. Intraoperatively, the mass was found to originate from the jugular foramen. Postoperatively, the patient largely recovered with expected hearing loss. Her diplopia resolved and there was no evidence of facial weakness. Her mother noted that she coughed more throughout the day, which may be a sign of lower cranial nerve dysfunction, but she has had no difficulty swallowing and has remained on a regular diet.

Based on brain MRI, the mass appeared to be dural based and hypervascular and the patient underwent a preoperative diagnostic cerebral angiogram, which identified the primary arterial supply from the APA. A prior study has shown high rates of success for embolization of the APA. It was decided that the patient should undergo a preoperative embolization to aid in surgical resection. Our case is unique in that our treatment management included the preoperative use of endovascular arterial embolization followed by surgical resection.

The preoperative use of endovascular embolization assisted in the ease of surgical resection. This has been demonstrated in preoperative embolization of meningiomas. A systematic review of preoperative embolization of meningiomas found less intraoperative blood loss and less overall operative time. This has also been found in preoperative embolization of other hypervascular pathologies like arteriovenous malformations, paragangliomas, and carotid body tumors. Another systematic review found that preoperative embolization has a low morbidity/mortality profile. It has also been found to be safe in pediatric...
| Series           | Year | Age (years) | Sex | Presentation                                      | Location                      | Size (cm) | Gene fusion       | Adjuvant therapy                                                                 | Follow-up (months) | Status  |
|-----------------|------|-------------|-----|---------------------------------------------------|-------------------------------|-----------|-------------------|---------------------------------------------------------------------------------|-------------------|---------|
| Dunham et al    | 2008 | 25          | M   | HA, N/V, right HH                                 | Left occipital                | 5.3       | EWS/ATF-1         |                                                                                  |                   |         |
| Ochiński et al  | 2010 | 35          | M   | HA, right facial weakness                         | Left mesial temporal          | 0.5 × 0.5 | EWSR1 gene rearranged | Seven repeat surgeries for clot evacuation and/or debulking; two radiosurgeries | 49                | Deceased|
| Hansen et al    | 2015 | 17          | F   | HA, blurry vision, anemia, left arm hyper-reflexia | Bioccipital (extra-axial)     |           |                   | Repeat resection                                                                | 16*               | Alive   |
| Ablouff et al   | 2016 | 58          | F   | Weight loss, right facial weakness, right hearing loss | Medet's cave                  | 6.1 × 4.8 × 2.9 | EWSR1 rearranged |                                                                                  | 6                 | Alive   |
| Kao et al       | 2017 | 15          | F   | Meninges                                          | EWSR1-CREM                    | 2.5 × 2.3 × 1.0 | EWSR1-CREB1 |                                                                                  | 17                | Alive   |
|                 |      | 23          | F   | Meninges, occipital                               | EWSR1-CREB1                   |           |                   |                                                                                  |                   |         |
|                 |      | 20          | M   | Frontal                                           | EWSR1-CREB1                   |           |                   |                                                                                  | 156*              | Alive   |
| Spatz et al     | 2018 | 22          | F   | HA, seizure, left HH                               | Right occipital (extra-axial) | 3.1 × 3.1 × 2.6 | EWSR1-ATF1 |                                                                                  | 3                 | Alive   |
| Bale et al      | 2018 | 14          | F   | HA, N/V, diplopia                                 | Left lateral ventricle        | 3.8 × 3.6 × 3 | EWSR1-CREB1 |                                                                                  | 47*               | Alive   |
|                 |      | 18          | M   | Seizure                                           | Right frontal                 | 3.0 × 2.0 × 1.5 | EWSR1-CREM  |                                                                                  | 12                | Alive   |
| Gareton et al   | 2018 | 19          | M   | Seizure                                           | Right temporo-occipital (extra-axial) |           | EWSR1-CREM        | API/AI type chemotherapy; radiation 61.2 Gy in 34 fractions; repeat resection for recurrence | 120               | Alive   |
| Sciort et al    | 2018 | 17          | F   | Seizure, right hemiparesis                        | Left frontal                  | 5.9       | EWSR1-ATF1        | Repeat resection for recurrence; radiation 59.4 Gy; additional resection for recurrence | 90                | Alive   |
| Gunness et al   | 2019 | 32          | F   | HA, neck pain, papilledema                        | Right lateral ventricle       |           |                   | Repeat resection for recurrence; shunt for recurrent cyst                        | 24                | Alive   |
| Konstantinidis et al | 2019 | 13          | F   | HA, nystagmus                                     | Right frontal (extra-axial)   |           | EWSR1-ATF1        | Repeat resection for recurrence                                                  | 132               | Alive   |
|                 |      | 12          | F   | HA, N/V, blurry vision, right pronator drift      | Left frontal                  |           | EWSR1-CREB1       |                                                                                  | 28                | Alive   |
| Ghanbargi et al | 2019 | 58          | F   | Seizure, left hemiparesis                         | Right parietal (extra-axial)  | 1.6       | EWSR1-CREB1       |                                                                                  | 3                 | Alive   |
| Aizpurua et al  | 2019 | 9           | M   | HA, N/V, transient vision loss, papilledema, left facial weakness, right uvula deviation | Left precentral gyrus         | 2.5 × 2.0 × 1.8 | EWSR1-ATF1 |                                                                                  | 12                | Alive   |
| White et al     | 2019 | 9           | M   | Fatigue, weight loss, abulia                      | Right frontal (extra-axial)   | 2.1       | EWSR1-CREM        | Repeat resection for recurrence; radiation 50 Gy in 26 fractions with a boost of 10 Gy to cavity | 6                 | Alive   |
| Bin Abdulqader et al | 2020 | 10          | M   | HA, N/V, seizure, left hemiparesis, left facial weakness, left pronator drift | Bifrontal                    | 4          | EWSR1 rearranged   |                                                                                  | 3                 | Alive   |

(Continued)
| Series | Year (years) | Age | Sex | Presentation | Location | Size (cm) | Gene fusion | Adjuvant therapy | Follow-up (months) | Status |
|--------|--------------|-----|-----|--------------|----------|-----------|-------------|-----------------|-------------------|--------|
| Komatsu et al | 2020 | 53 | F | HA, dizziness | THrd ventricle | 2.8 x 1.9 | EWSR1-CREB1 | Radiation | 3 | Alive |
| Domingo et al | 2020 | 36 | F | HA, N/V, diplopia, lower extremity weakness | Interhemispheric (extra-axial) | 1.9 | EWSR1-ATF1 | Radiation | 3 | Alive |
| Ballester et al | 2020 | 67 | M | Confusion, expressive aphasia | Left temporal (extra-axial) | EWSR1-ATF1 | Radiation | 3.5 | Alive |
| Valente Aguiar et al | 2020 | 58 | F | HA, N/V, confusion, gait imbalance, right hemiparesis | Left lateral ventricle | EWSR1-CREB1 | Radiation | 6 | Alive |
| Ward et al | 2020 | 48 | F | HA | Left lateral ventricle | EWSR1-ATF1 | Repeat resection for recurrence; radiation for recurrence 35 Gy in 5 fractions | 16 | Alive |
| Gilbert et al | 2020 | 52 | M | HA, N/V, imbalance, weight loss | Vermian (extra-axial) | EWSR1-CREB1 | Radiation | 12 | Alive |
| Sloan et al | 2020 | 12 | M | | Parietal | EWSR1-ATF1 | Radiation 59.4 Gy | 24 | Alive |
| | 2020 | 9 | F | | Frontal | EWSR1-ATF1 | | 63 | Deceased |
| | 2020 | 24 | F | | Occipital | EWSR1-ATF1 | | | |
| | 2020 | 13 | F | | Frontal | EWSR1-ATF1 | | 24 | Alive |
| | 2020 | 34 | F | | Tentorium | EWSR1-ATF1 | | 81 | Alive |
| | 2020 | 17 | F | | CPA | EWSR1-ATF1 | Radiation; chemotherapy | 27 | Deceased |
| | 2020 | 70 | M | | CPA with spinal dissemination | EWSR1-ATF1 | | 1 | Deceased |
| | 2020 | 17 | F | | CPA | EWSR1-ATF1 | | 13 | Alive |
| | 2020 | 14 | F | | Lateral ventricle | EWSR1-CREB1 | | 59 | Alive |
| | 2020 | 39 | F | | Lateral ventricle | EWSR1-CREB1 | | 6 | Alive |
| | 2020 | 10 | M | | Falx (parietal) | EWSR1-CREB1 | | 57 | Alive |
| | 2020 | 25 | F | | CPA | EWSR1-CREB1 | | 30 | Alive |
| | 2020 | 14 | F | | Parietal | EWSR1-CREB1 | | 57 | Alive |
| | 2020 | 15 | F | | Spinal cord (thoracic) | EWSR1-CREB1 | Radiation | 30 | Alive |
| | 2020 | 14 | F | | Lateral ventricle | EWSR1-CREB1 | | 38 | Alive |
| | 2020 | 5 | F | | Frontal | EWSR1-CREB1 | Chemotherapy | 11 | Alive |
| | 2020 | 30 | M | | Falx (frontal) | EWSR1-CREB1 | Radiation 54 Gy | 6 | Alive |
| | 2020 | 4 | F | | Occipital | EWSR1-CREB1 | | 36 | Alive |

Abbreviations: API/AI, doxorubicin-cisplatin-ifosfamide; CPA, cerebellopontine angle; F, female; Gy, Gray; HA, headache; HH, homonymous hemianopsia; M, male; N, nausea; V, vomiting.

*updated follow up from Ballester et al.
*updated follow up from Valente Aguiar et al.
*updated follow up from Sloan et al.
*updated follow up from Bin Abdulqader et al.
patients and in skull base pathologies. Like meningiomas, AFH is most commonly extra-axial or intraventricular with radiographic dural attachments. As the feeding vessel for this tumor was from the APA, the major arterial supply was from the external carotid artery and deep to the tissue mass. Embolization involved the external carotid instead of the internal carotid circulation, which decreases catheter time in the intracranial circulation and decreases risk of stroke. However, attention must be paid to the APA itself as the internal carotid mass. Embolization involved the external carotid instead of this tumor was from the APA, the major arterial supply with radiographic dural attachments. As the feeding vessel resection.

The arterial feeder for this tumor was also deep and obtaining surgical devascularization would have come late in the procedure.

Similar to meningiomas, it appears that AFH outcomes are linked to extent of resection. Sloan et al reported in their case series that patients who received gross total resection had better survival and local recurrence rates, but those values did not reach statistical significance. Due to their propensity for local recurrence, follow-up radiation was suggested by their group. Since these can be hemorrhagic masses with deep arterial feeders, we suggest preoperative embolization may allow for the best chance at safely achieving gross total resection.

**Conclusion**

AFH is a rare tumor that mimics more commonly found intracranial neoplasms. We present a rarely reported skull base AFH and the first located at the jugular foramen. Due to the importance of gross total resection, we recommend preoperative evaluation with diagnostic cerebral angiogram to evaluate for preoperative embolization, which may improve rates of gross total resection. In the setting of gross total resection, adjuvant radiation therapy may be avoided. This is beneficial as the tumor is more common in children and young adults for which radiation carries long-term risks.

Conflict of Interest
None declared.

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