Malignant Vascular Tumors of Temporal Bone:
Review Article

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Abstract
Objective: The objective of this study was to present a review article about temporal bone vascular tumors.

Data Sources: Published English-language literatures in PubMed and Google scholar.

Review methods: PubMed and Google scholar were systematically searched using search terms: angiosarcoma, temporal and bone, hemangioendothelioma, temporal and bone. Kaposi sarcoma, temporal and bone, hemangiopericytoma, temporal and bone.

Study Selection: We included studies about vascular tumors of temporal bone.

Results: Forty-five studies were included in this study. The results showed the most patients were male, mass was the common symptom, external ear was the most common site and surgery was the essential treatment.

Conclusion: Malignant vascular tumors of temporal bone are rare malignant sarcomas that should be considered in the differential diagnosis of temporal bone mass.

Introduction
Malignant vascular tumors are extremely rare in children and adults. Vascular neoplasms are the most common subcutaneous deep soft tissue sarcoma and in pediatric age. Malignancy is only seen in 2% of blood vessel tumors. They include angiosarcoma, hemangioendothelioma, Kaposi sarcoma and hemangiopericytoma. A review by the University of Minnesota revealed only four case of malignant vascular tumor among 228 vascular tumor (3 angiosarcoma and one Kaposi cancer). A review by the Memorial Sloan-Kettering Cancer Center included six patients less than 21 years diagnosed between 1970 and 1995: four were angiosarcomas and two malignant hemangiendotheliomas. Farr et al. [1,2] reported 21 cases of head and neck malignant vascular tumor, ten of them were angiosarcomas and the other eleven cases were hemangiopericytomas. The absolute and net five year cure rate was 25% and was achieved by radical surgery alone.

Material and Methods
Literature review was conducted using PubMed (MEDLINE) and Google Scholar for English articles. The following keywords were used: angiosarcoma, temporal and bone; hemangioendothelioma, temporal and bone; Kaposi, sarcoma, temporal and bone; hemangiopericytoma, temporal and bone.

Inclusion criteria
All malignant vascular tumors of temporal bone published articles were included in the study.

Results: Forty-five studies about malignant vascular tumors of temporal bone were available in PubMed (MEDLINE) and Google scholar in English literature (Tables 1 & 2).

Table 1: Temporal bone malignant vascular tumor articles.

| Age | Sex | Type                  | Symptom                   | Location | Angiography                      | Treatment | Follow up |
|-----|-----|-----------------------|---------------------------|----------|----------------------------------|-----------|----------|
| 3   | M   | Hemangioendothelioma  | Retroauricular Swelling    | Mastoid  | A interferon                     |           |          |
|     |     |                       | Tenderness                |          |                                  |           |          |
|     |     |                       | Fever lymphadenopathy     |          |                                  |           |          |


| Name and Year | Age | Gender | Tumor Type | Symptoms | Presentation | Treatment | Follow-up |
|---------------|-----|--------|------------|----------|--------------|-----------|-----------|
| Panda et al. [4] | 38 | M | Hemangioendothelioma | Tinnitus, Hearing loss, Fullness of the ear, Dizziness | Middle ear | Surgery | 6 months |
| Moskowitz et al. [5] | 6 | F | Hemangioendothelioma | Facial palsy, Hearing loss, Dizziness, Tinnitus | Middle ear | Surgery |  |
| Lalaji et al. [6] | 1 | F | Hemangioendothelioma | Mass, mastoid | Surgery, Predisolon | 2 years |
| Jo-chaim et al. [7] | 19 | M | Hemangioendothelioma | Tinnitus, Earache, Discharge, Hearing loss | Mastoid | P Surgery, Radiotherapy | 3 years |
| Kim et al. [8] | 7 | M | Hemangioendothelioma | Mass, Mastoid | branches of the middle meningeal artery, Partial Surgery, Radiotherapy | Recurrent 2 year, Radiotherapy, Chemotherapy |
| Ibarra, et al. [9] | 5 | F | Hemangioendothelioma | Mass, Facial palsy, EAM | Middle meningeal artery | Surgery |  |
| Goldestien et al. [10] | 62 | N | Hemangioendothelioma | Tinnitus, Hearing loss, Vertigo, Mass | Middle ear, Normal angiogram | Surgery, Radiotherapy | 1 year |
| Chang et al. [11] | 1 | M | Hemangioendothelioma | Mass, Facial palsy, IAM | External carotid | Surgery, Cortison, Interferon | 1 year |
| JawadAkhterGilani et al. [12] | 45 | M | Kaposi | Multiple Auricle mass | Chemotherapy | 4 months |
| Acoglu et al. [13] | 45 | M | Kaposi | Auricle mass | Recurrent Surgery | 11 year |
| Busi et al. [14] | 72 | F | Kaposi | Auricle Multiple mass | Surgery | 18 months |
| Colletti et al. [15] | 6 | M | Kaposi | Auricle Multiple mass | Treat AIDS, AIDS HSV 8 positive |  |
| Rachadi et al. [16] | 64 | F | Kaposi | Auricle Multiple mass | Chemotherapy |  |
| Nauton et al. [17] | 68 | M | Kaposi | Auricle mass | Surgery, Recurrence 3 year |  |
| Authors                  | Sex | Diagnosis         | Location                     | Treatment Notes                                                                 | Follow-up |
|-------------------------|-----|-------------------|------------------------------|---------------------------------------------------------------------------------|-----------|
| Nervi et al. [18]       | M   | Kaposi            | Tragus Mass                  | Surgery                                                                         |           |
| STEARNs et al. [19]     | M   | Kaposi            | Tragus Mass                  | Surgery, 2 year                                                                 |           |
| Izquierdo Cuenca et al. [20] | M   | Kaposi            | Tragus Multiple Mass         | Surgery, HSV8                                                                     |           |
| Francés Rodríguez et al. [21] | M   | Kaposi            | Pinna Mass                   | Surgery                                                                         |           |
| Gibbas et al. [22]      | F   | Kaposi            | Multiple nodules on each ear mass | Surgery                                                                         |           |
| Babuccu et al. [23]     | M   | Kaposi            | auricle mass                 | Surgery, HSV 8                                                                   |           |
| Delbrouck et al. [24]   | M   | Kaposi            | EAM multiple Mass            | Systemic hormonal therapy. Local injection of a cytotoxic, radiotherapy resulted in a 50% regression of the tumour mass. T | AIDS     |
| Kusenbachet al. [25]    | M   | Kaposi            | Auricle Multiple Mass        | chemotherapy, HSV 8 positive                                                    |           |
| Kumarasamy et al. [26]  | M   | Kaposi            | EAM Multiple Mass            | Treat AIDS, AIDS                                                                |           |
| Kumarasamy et al. [26]  | M   | Kaposi            | EAM Multiple Mass            | Treat AIDS, AIDS                                                                |           |
| Linstrom et al. [27]    | M   | Kaposi            | Mastoid multiple Mass        | Treat AIDS, AIDS                                                                | Aids     |
| Pergolizzi et al. [28]  | M   | Kaposi            | Auricle Mass                 | Radiotherapy, Aids                                                              |           |
| Koscielny et al. [29]   | F   | Hemangiopericytoma| Tinnitus Red mass behind tm  | Surgery, 10 year, Angio Maxillary Occipital artery embolized                     |           |
| SUTBEYA ET AL. [30]     | F   | Hemangiopericytoma| Otorrhea, hearing loss, Otalgia Mass | Surgery, 1 year, No recurrence                                                   |           |
| Author(s)                | Gender | Age | Tumor Type         | Symptoms                                    | Treatment   | Duration |
|-------------------------|--------|-----|--------------------|---------------------------------------------|-------------|----------|
| Chotey et al. [31]      | F      | 60  | Hemangiopericytoma | Otorrhea, hearing loss, and a mass otorrhagia, hearing in the temporal bone | Middle ear surgery | 8 months |
| Castiglione et al. [32] | M      | 43  | Hemangiopericytoma | Auricular pain hearing sense of hearing in the temporal bone | Auricle surgery | 3 years |
| Tewfik et al. [33]      | M      | 21  | Hemangiopericytoma | Mass Hearing loss Tinnitus | Temporal bone EAM Surgery Radiotherapy | One year Pulmonary metastasis |
| Bignard et al. [34]     |        | 10  | Hemangiopericytoma | Mass Hearing loss Tinnitus | Tympanic bone mastoid process Soft tissue lesions surgery | Remission |
| Cross et al. [35]       | M      | 62  | Hemangiopericytoma | Otorrhea | Middle ear Surgery | 2 years |
| Bist et al. [36]        | M      | 35  | Hemangiopericytoma | Mass Hearing loss Tinnitus | Mastoid | Surgery Radiation | 2 years |
| Chin et al. [37]        | F      | 35  | Hemangiopericytoma | Mass | Mastoid | Middle ear surgery \Mannigreal retoauricle | Radiotherapy | 2 years |
| Birzagals et al. [38]   | M      |     | Hemangiopericytoma | Mass Retrarural mass | Middle Ear surgery | |
| Mahrous [39]            | M      | 50  | Hemangiopericytoma | Hearing loss Tinnitus Facial palsy | Juglar fossa surgery | |
| Megerian et al. [40]    | M      |     | Hemangiopericytoma | Facial Hearing loss Mass | Juglar fossa Ascending pharyngeal Radiotherapy surgery | |
| Schoksem et al. [41]    | M      | 26  | Angiosarcoma        | Mass | Mastoid | middle meningeal posterior auricular occipital artery tentorium cerebelli artery | Surgery Radiation Chemotherapy | 15 Months Pulmonary metastasis |
| Lu et al. [42]          | M      | 36  | Angiosarcoma        | Hearing loss otalgia | Middle ear mastoid | Surgery radiotherapy | 3 months |
A. Demography: There were 47 cases of malignant vascular tumors, 7 cases of angiosarcoma, 14 cases of hemangiopericytoma, 17 cases of kaposi sarcoma and 9 cases of hemangioendothelioma. There were 33 male and 14 female. There were 11 children and 36 adults. Angiosarcoma and hemangioendothelioma are seen more in child and young patients, while hemangiopericytoma and Kaposi sarcoma are seen more in adults (Figures 1-3) (Table 3).

### Table 2: Temporal bone vascular tumors patients’ age.

|                      | Angiosarcoma | hemangiopericytoma | kaposi | hemangioendothelioma |
|----------------------|--------------|--------------------|--------|----------------------|
| Age rang             | 9 - 57       | 10-64              | 3 - 81 | 1 - 62               |
| mean                 | 30           | 40                 | 49     | 15                   |

### Table 3: clinical symptoms of malignant vascular tumors.

|                      | Angiosarcoma | hemangiopericytoma | kaposi | hemangioendothelioma | all      |
|----------------------|--------------|--------------------|--------|----------------------|----------|
| Mass                 | 4/7          | 7/14               | 17/17  | 6/9                  | 34/47 (72%) |
| HL                   | 5/7          | 7/14               | 1/17   | 4/9                  | 17/47 (36%) |
| FP                   | 1/7          | 2/14               | 3/9    | 6/47                 | 12%      |
| Tinnitus             | 5/14         |                    | 3/9    | 8/47                 | 17%      |
| Vertigo              |              | 2/14               | 1/9    | 3/47                 | 6%       |
| otorrhea             | 4/7          | 3/14               | 1/9    | 8/47                 | 17%      |
| Otalgia              | 4/7          | 4/14               | 1/9    | 8/47                 | 17%      |

**Figure 1:** Different types of malignant temporal bone vascular tumors.

**Figure 2:** M/F Ratio.
B. Location: The most common location was middle ear for angiosarcoma and hemangiopericytoma, external ear for Kaposi sarcoma and mastoid for hemangioendothelioma (Figure 4).

C. Clinical symptom: Mass was the most common symptom in all types of malignant vascular tumors, followed by hearing loss and otorrhea (Table 2) (Figure 5).

D. Clinical course: Kaposi sarcoma of temporal bone usually appear as mass in external ear, hemangiopericytoma appear like a glamous tumor or mass, angiosarcoma appear as CSOM, while there is no specific form for angiosarcoma (Figure 6).

E. Treatment: Surgery is the essential treatment for malignant vascular tumors, radiotherapy used for high degree tumors or close margins, chemotherapy used for angiosarcoma and multiple lesions of Kaposi sarcoma. Interferon or cortisonmay be used for hemangioendotheloma. AIDS should be treated in positive patients with sarcoma Kaposi (Figures 7 & 8).

F. Recurrence: There were 6 cases of recurrence 12% (one case of angiosarcoma, one case of hemangiopericytoma, one case of hemangioendothelioma and 3 cases of Kaposi sarcoma).

Kaposi Sarcoma

There were 17 case of Kaposi sarcoma, 10 cases were multiple lesions (58%), 6 cases associated with AIDS (35%) and four cases associated with HSV 8 (23%).

Angiography

Middle meningeal artery was the most common artery giving blood supply to tumors followed by posterior auricle artery (Figure 7).

Discussion

Hemangioendotheliomas are neoplastic proliferations of blood vessel endothelial cells, forming thin-walled blood vessels and sheets of neoplastic cells. These lesions have variable malignant behavior, depending on their microscopic features. Hemangioendotheliomas usually show poor histologic differentiation, nuclear a typia and the existence of mitotic figures. These tumors may be divided into three groups: grade I, grade II, and grade III. The diagnostic grades I through III [3-7] are based on the presence of abundant vascular channels lined by only mildly atypical neoplastic endothelial cells and rare mitoses (in grade I). Grade III lesions show the most anaplastic features.

The radiographic findings of hemangioendothelium usually demonstrate an osteolytic sharply demarcated lesion. Surgical resection is the treatment of choice for hemangioendothelioma, radiation is advised for high grade lesions and non-operable cases. Chemotherapy has no significant role in the treatment.
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Some authors reported successful treatment with a interferon in two infants with hemangioendotheliomas [3].

KS is a vascular tumor that originates from vascular and lymphatic endothelium, it is characterized by a multifocal angiogenic process, frequently presenting as multiple vascular cutaneous and mucosal nodules. Nodal and visceral manifestations are usually observed in case of severe immuno suppression. Risk factors for KS development may include diabetes, advanced age and the use of corticosteroid medication. KS is also associated with viral infection such AIDS and HHV8. Typical lesions are reddish-purple papules, maculae, or nodules. KS mostly occurs in elderly men as multiple bilateral cutaneous lesions of the lower extremities. The involvements of the head and neck area have been observed in only 14% of cases of KS with only a few cases of ear involvement. A standard therapeutic guideline is not available; the treatment is depending on the subtype and stage of the neoplasm as well as on the immune status of the patient [15].

Haemangiopericytoma is extremely rare vascular tumor that usually seen in the trunk and lower extremities. It accounts 1% of all vascular neoplasms, about 15-25% of haemangiopericytoma occur in the head and neck region. The pericytes are the round or spindle shaped contractile cells, proliferations of these cells are responsible for the haemangiopericytoma evolution. McMaster et al. have divided haemangiopericytoma as histologically as: benign (low-grade); borderline (intermediate grade); and malignant (high-grade). They expected malignant behavior in cases having a slight degree of cellular anaplasia or one mitotic figure per 10 highpower fields or having a moderate degree of cellular anaplasia and one mitotic figure per 20 high-power fields. The majority of the reported head and neck cases are seen in the nasopharynx, nasal cavity, paranasal sinuses, mandible, maxilla and the orbit. Haemangiopericytoma of the temporal bone is extremely rare with only a few cases were reported in English literatures. Pre-operative embolization may help to obtain a better surgical result. Surgical excision is the treatment of choice. Radiotherapy may be helpful after incomplete resection of the tumor. The role of chemotherapy in the treatment of the haemangiopericytoma is still unclear. The recurrence rate of the tumour is relatively high [31].

Angiosarcoma is a rare malignant sarcoma with endothelial differentiation. They represent only 0.5-1% of malignant primary bone neoplasia. Bone angiosarcomas as usually occur in the limbs and less likely in the pelvis, ribs, and vertebra. The skull is an unusual site of angiosarcoma with only few cases reported in the literature. Parietal and frontal bones are more commonly involved than the sphenoid, occipital and temporal bones. Radiation therapy, arsenic exposure and previous trauma have not reported to be predisposing factors for skull angiosarcoma. Cranial angiosarcomas are often unicentric lesions. Most cases of skull angiosarcoma occurred in the third decade with a marked male predominance. Skull angiosarcoma often occur as growing mass or swelling, while the manifestations of temporal bone angiosarcoma are hearing loss, tinnitus, and otalgia with a mass in the temporal area. On CT scan, skull angiosarcoma usually appear as a well-demarcated lytic, hypervascularized and hemorrhagic mass. Angiosarcomas have variable malignant behavior related to their degree of vascular differentiation and microscopic appearance. Immunohistochemistry is necessary in poorly differentiated tumors for the identification of an endothelial lineage. Skull angiosarcomas have a rapid onset of symptoms and high percentage of local recurrence or metastasis. The prognosis of skull angiosarcoma is a worse due secondarily involvement of the meninges and the brain and the impossibility to perform a complete surgical resection. Extensive staging must be conducted in all cases at the time of diagnosis and the follow-up should include regular bone CT scan, cerebral imaging and chest radiography. Preoperative embolization may help to achieve a total excision of the tumor with minimal blood loss. The complete surgical removal of the tumor is the most effective treatment, but adequate margins are often difficult to achieve. Radiotherapy may be given as an adjunct to surgical therapy or as palliative treatment in non-operative cases of skull angiosarcoma. Adjuvant chemotherapy with paclitaxel may help the condition of patients with soft-tissue sarcoma of the face or scalp [41].

Conclusion

Malignant vascular tumors of temporal bone are very rare, mostly seen in adult male except hemangioendothelioma which is seen more in pediatrics. Kaposi sarcoma is the most common type of these tumors, it is usually seen as multiple lesions associated with HIV or HSV virus. Mass and hearing loss are the most common symptoms. Middle meningeal artery was the most common artery giving blood supply. Surgery is the essential treatment with adjuvant radiotherapy and chemotherapy for selected cases. There is a local recurrence in 12% of cases (Table 4).

Table 4: Temporal bone vascular tumors conclusion.

| Symptom      | Angiosarcoma | hemangiopericytoma | kaposi | hemangioendothelioma | All       |
|--------------|--------------|--------------------|--------|----------------------|-----------|
| M/F          | M=F          | M<F                | M=F    | F=M                  | M=F       |
| Child/Adult  | Adults<Children | Adults<Children  | Adults<Children | Children<Adults | Adults<Children |
| Location     | Middle ear   | Middle ear         | External ear | Mastoid    | External ear |
|              | Mass         | Mass               | Mass    | Mass                 | Mass      |
Clinical appearance | CSOM | Like glomus tumor or mass | Mass | Like glomus tumor or mass | Mass
--- | --- | --- | --- | --- | ---
Treatment | Surgery | Radiotherapy | chemotherapy | Surgery | Chemotherapy for isolated cases | mass | multiple case | Treat AIDS | Surgery | And radiotherapy for high degree tumor
Recurrence | 1/7 | 1/14 | 3/17 | 1/9 | 6/47

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