Global Journal of
Otolaryngology
ISSN 2474-7556

Juniper Publishers
key to the Researchers

Review Article
Volume 2 Issue 3 – December 2016
DOI: 10.19080/GJO.2016.02.555589

Glob J Otolaryngol

Malignant Vascular Tumors of Temporal Bone: Review Article

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Submission: November 22, 2016; **Published:** December 06, 2016

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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: angisarcoma, 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: Fortyfive 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 hemangioendotheliomas. 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: angisarcoma, 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
Eliashar et al. [3]	3	M	Hemangioendothelioma	Retroauricular Swelling Tendeness Fever lymphadenophathy	Mastoid		A interferon	

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	
Lal.aji et al. [6]	1	F	Hemangioendothelioma	mass	mastoid		Surgery predisolon	2 year
Jochaim et al. [7]	19	M	Hemangioendothelioma	Tinnitus Earache Discharge Hearing loss	mastoid		P Surgery radiotherapy	3 year
Kim et al. [8]	7	M	Hemangioendothelioma	Mass	Mastoid	branches of the middle meningeal artery	Partial Surgery Radiotherapy	Recurrent 2 year Radiotherapy chemothe-rapy
Ibarra, et al. [9]	5	F	Hemangioendothelioma	Mass Facial plasy	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	Facial palsy	IAM	External carotid	Surgery Cortison interferon	1 year
JawadAkhterGilani et al. [12]	45	M	Kaposi	Multiple Auricle	Mass		chemotherapy	4 months
Acıoğlu et al. [13]	45	M	Kaposi	Auricle	Mass		Recurrent Surgery	11 year
Busi et al. [14]	72	F	Kaposi	Auricle Mutiple	Mass		Surgery	18 month
	6	M	Kaposi	Auricle	Mass		Treat AIDS	AIDS HSV 8 positive
Colletti et al. [15]	57	M	Kaposi	Auricle Multiple	Mass		Surgery	Recurrence 3year
Rachadi et al. [16]	64	F	Kaposi	Auricle Multiple	Mass		Chemotherapy	
Nauton et al. [17]	68	M	Kaposi	Auricle	Mass		Surgery	Recurrent 9 year

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Nervi et al. [18]	52	M	Kaposi	Tragus	Mass		Surgery	
STEARNS et al. [19]	66	M	Kaposi	Tragus	Mass		Surgery	2 year
Izquierdo Cuenca et al. [20]	81	M	Kaposi	Tragus Multiple	Mass		surgery	HSV8
Francés Rodríguez et al. [21]	70	M	Kaposi	Pinna	Mass		Surgery	
Gibbas et al. [22]	73	F	Kaposi	Multiple nodules on each ear	mass		surgery	
Babuccu et al. [23]	36	M	Kaposi	auricle	mass			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]	3	M	Kaposi	Auricle Multiple	Mass		chemotherapy	HSV 8 positive
Kumarasamy et al. [26]	40	M	Kaposi	EAM Multiple	Mass		Treat AIDS	AIDS
Kumarasamy et al. [26]	40	M	Kaposi	EAM Multiple	Mass		Treat AIDS	AIDS
Linstrom et al. [27]	35	M	Kaposi	Mastoid multiple	Mass		Treat AIDS	Aids
Pergolizzi et al. [28]	40	M	Kaposi	Auricle	Mass		Radiotherapy	Aids
Koscielny et al. [29]	64	F	Hemangiopercytoma	Tinnitus Red mass Behind tm	Middle ear	Angio Maxillay Occipital artery embolzed	Surgery	10 year
SUTBEYA ET AL. [30]	18	F	Hemangiopercytoma	Otorrhea, hearing loss, Otalgia Mass	Middle ear		surgery	1 year No recurrence

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Chotey et al. [31]	60	F	Hemangiopercytoma	otorrhea, hearing loss, and a mass otorrhagia,	Middle ear		surgery	8 months
Castiglione et al. [32]	43	M	Hemangiopercytoma	auricular pain hearing sense of hearing in the	Auricle		surgery	3 years
Tewfik et al. [33]	21	M	Hemangiopercytoma	Mass Hearing loss Tinnitus	Temporal bone EAM		Surgery Radiotherapy	One year Pulmonary meatstasis
Bignard et al. [34]	10		Hemangiopercytoma	Mass Hearing loss Tinnitus	Tympanic bone, mastoid process Soft tissue lesions		surgery	remmision
Cross et al. [35]	62	M	Hemangiopercytoma	otorrhea	Middle ear		Surgery	2year
Bist et al. [36]	35	M	Hemangiopercytoma	Mass Hearing loss tinnitus	Mastoid		Surgery Radiation	2 year
Chin et al. [37]	35	F	Hemangiopercytoma	Mass	Mastoid	Minnigeal retoauricle	Surgery Radiotherapy	2 year
Birzgaliset al. [38]			Hemangiopercytoma	Middle ear mass	Middle Ear		surgery	
			Hemangiopercytoma	Retrauricl mass	Mastoid	surgery	surgery	remmision
			Hemangiopercytoma	Deafness Dizziness Tinnitus paillodema	Petrous		surgery	
Mahrous [39]	50	M	Hemangiopercytoma	Hearing loss Tinnitus Facial palsy	Juglar fossa		surgery	
Megerian et al. [40]			Hemangiopercytoma	Facial Hearing loss Mass Hearing loss	Juglar fossa	Ascending pharyngeal	Radiotherapy surgery	
Scholsem et al. [41]	26	M	Angiosarcoma	Mass	Mastoid	middle meningeal posterior auricular occipitalartery tentorium cerebelli artery	Surgery Radiotherapy chemotherapy	15 Months Radiation Died 20 months Pulmonary metastasis
Lu et al. [42]	36	M	Angiosarcoma	Hearing loss otalgia	Middle ear mastoid		Surgery radiotherpy	3 months

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Küstermeyer et al. [43]	13	M	Angiosarcoma	Hearingloss Otalgia Facial palsy	Petrous angiosar-coma	from the middle meningeal artery	Radiotherapy Surgery chemotherapy	
Hindersin et al. [44]	57	F	Angiosarcoma	Hearing loss otorrhea	Middle ear	External carotis	Surgery Radiotherapy Chemotherapy	Recurrent
Buraïma et al. [45]	12	F	Angiosarcoma	otorrhea Mass Mastoid EAM	Middle ear		Surgery Chemotherpy	No recurrence 12 month
Masiah et al. [46]	9	M	Angiosarcoma	Otorhea Mass Hearing loss	Petrous		Surgery Radiotherapy	3 year
Durko et al. [47]	55	F	Angiosarcoma	Otorrhea Mass Hearing loss	Mastoid		Surgery	Recurrence Died after 10 months

Table 2: Temporal bone vascular tumors patients' age.

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

A. Demography: There were 47 cases of malignant vascular tumors, 7 cases of angiosarcoma, 14 cases of hemangiopercytoma, 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 hemangiendothelioma are seen more in child and young patients, while hemangiopercytoma and Kaposi sarcoma are seen more in adults (Figures 1-3) (Table 3).

Table 3: clinical symptoms of malignant vascular tumors.

	Angiosarcoma	hemangiopercytoma	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%)

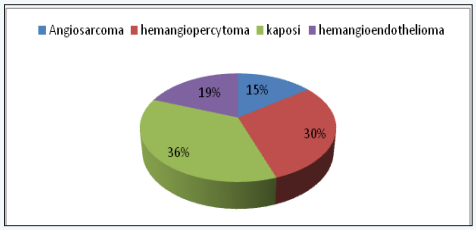

Figure 1: Different types of malignant temporal bone vascular tumors.

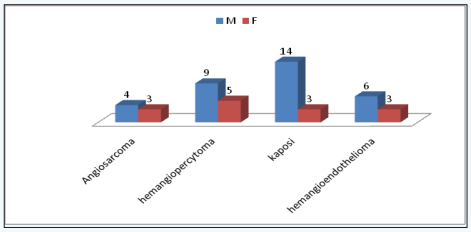

Figure 2: M/F Ratio.

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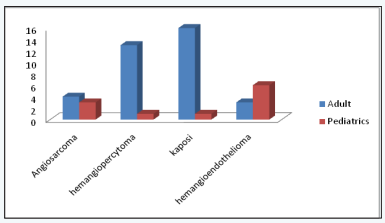

Figure 3: Adult / Pediatric Ratio.

B. Location: The most common location was middle ear for angiosarcoma and hemangiopercytoma, external ear for Kaposi sarcoma and mastoid for hemangioendothelioma (Figure 4).

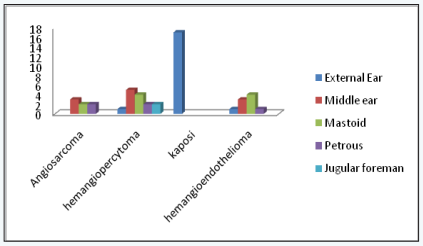

Figure 4: Location of temporal bone malignant vascular tumors.

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).

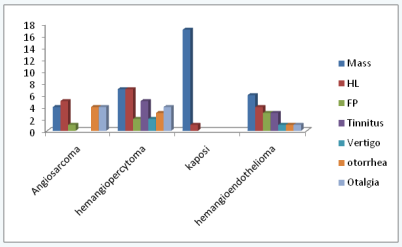

Figure 5: Symptoms of malignant vascular tumors.

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

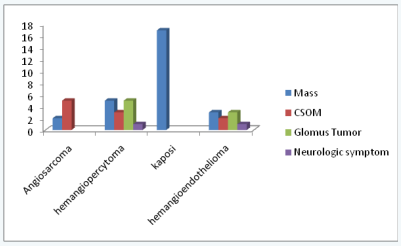

Figure 6: Clinical scenarios of different types of temporal bone vascular tumors.

E. Treatment: Surgery is the essential treatment for malignant vascular tumors, radiotherapy used for high degree tumors or close margins, chemotherapy used for angisarcoma 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).

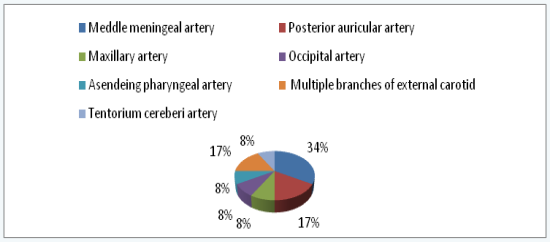

Figure 7: Angiography of Malignant vascular tumors of temporal bone.

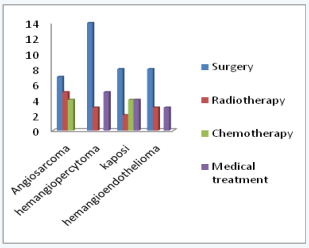

Figure 8: Treatment of temporal bone vascular tumors.

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

Kapos 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 e diabetes, advanced age and the use of corticosteroid medication KS is also associated with viral infection such AIDS and HHV8. Typical lesions are reddish-purplish 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 acounts1% of all vascular neoplasms, about 15-25% of haemangiopericytomas 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 haemangiopericytomas 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 angiosarcom as usually occur in the limbs and less likely in the pelvis, ribs, and vertebra. The skull is an unusual site of angiosarcom with only few cases reported in the literature. Parietal and frontal bones are more commonly involved than the sphenoid, occipitaland 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 poor 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-operable 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 commonly type of theses 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.

	Angiosarcoma	hemangiopercytoma	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
Symptom	Mass	Mass	Mass	Mass	Mass

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Clinical appearance	CSOM	Like glomus tumor or mass	Mass	Like glomus tumor or mass	Mass
Treatment	Surgery Radiotherapy chemotherapy	Surgery And radiotherapy for high degree tumor	Surgery for isolated cases Chemotherapy for 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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How to cite this article: Omar R. Malignant Vascular Tumors of Temporal Bone: Review Article. Glob J Otolaryngol. 2016; 2(3): 555589.DOI: 10.19080/GJO.2016.02.555589

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