

## OLGU BİLDİRİSİ / CASE REPORT

# Hemangioma of the hyoid bone: a rare case report

G. Saqulain, M.M. Khan

### Hiyoid kemik hemanjiyomu: Nadir bir olgu sunumu

Bu yazıda 30 yaşındaki erkek bir hastada ilk kez tespit edilen hiyoid kemik hemangioma olgusu sunulmaktadır. Kemik hemanjiyomları vasküler kaynaklı benign tümörlerdir. Primer kemik hemanjiyomları nadir tümörlerdir ve tüm kemik neoplazilerinin %1'den daha az bir kısmını oluştururlar. Kemik hemanjiyomları en sık vertebralarda ve daha sonra da kranial ve maksillofasiyal kemiklerde tespit edilir. Hemanjiyomların uzun ve kısa tübüler kemiklerde ve kaburgalarda görülmesi oldukça nadirdir. Literatür taramasında hiyoid kemik kaynaklı hemanjiyom olgusuna rastlanmamıştır. Hiyoid kemik hemanjiyomunun tedavisi cerrahidir. Prognoz iyidir ve nüks nadirdir.

**Anahtar Sözcükler:** Hiyoid kemik, hemanjiyom, primer kemik neoplazisi.

### Abstract

In this article, the first documented case of hemangioma of the hyoid bone in a 30-year-old male is reported. Bone hemangiomas are benign tumors of vascular origin. Primary bone hemangiomas are uncommon tumors. They constitute less than 1.0% of all bone neoplasms. Bone hemangiomas are most commonly found in the vertebral column followed by the cranial and maxillofacial bones. The occurrence of hemangiomas of long and short tubular bones and ribs is extremely rare. Literature search did not reveal any case of hemangioma arising from the hyoid bone. Treatment of the hyoid bone hemangioma is Surgery. The prognosis is good and recurrence is rare.

**Key Words:** Hyoid bone, hemangioma, primary bone neoplasm.

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## Introduction

Haemangiomas are benign vascular lesions with 4 histological variants: cavernous, capillary, arteriovenous and venous. Bone hemangiomas constitute less than 1% of all primary bone neoplasms.<sup>1-3</sup> They are predominantly of cavernous and capillary varieties. They occur most frequently in the verte-

bral column followed by the craniofacial skeleton and the long bones.<sup>1,2</sup> Involvement of other sites is extremely rare. In this study, we present the first documented case of hemangioma of the hyoid bone. Also we present the clinical and histological features of the case and our management strategy.

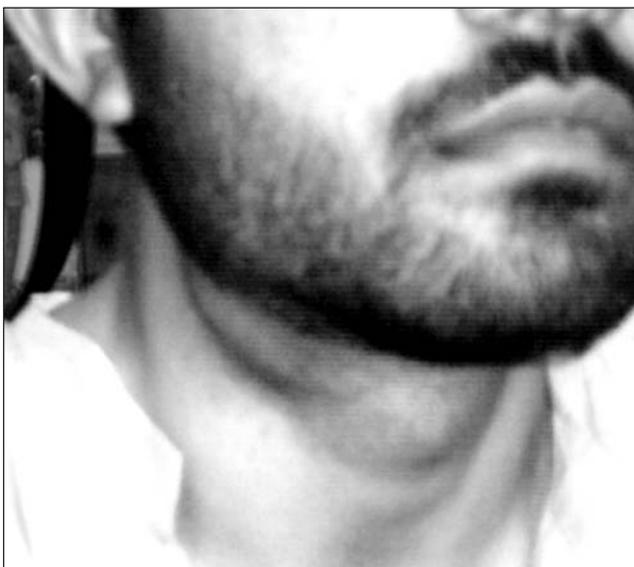
## **Case Report**

### **Clinical data**

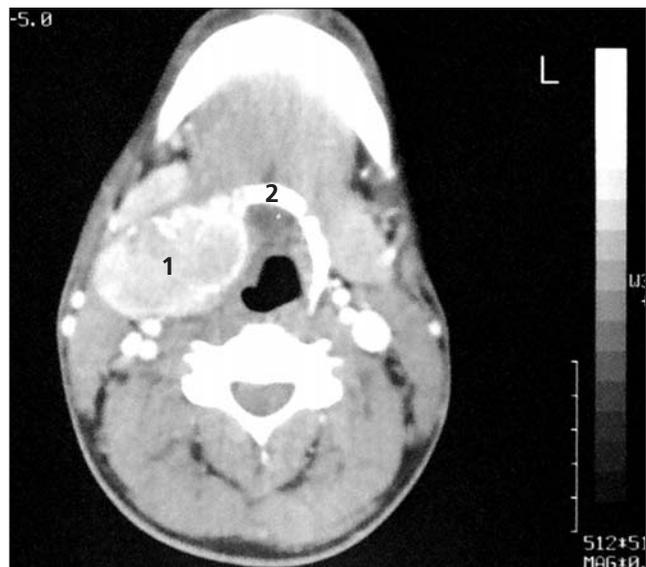
A 30-year-old male presented to the ENT Department of Capital Hospital, Islamabad, with five months history of a slowly enlarging swelling in the right submandibular region with no other symptoms. On examination an 8 x 5 cm, ovoid, mass was noted in the right submandibular region extending to the retromandibular area (Figure 1). There was no redness over the swelling and on palpation no tenderness or pulsation was noted. The swelling was firm in consistency with well defined, rounded

margins and appeared to move with the movement of the hyoid bone. Cranial nerve examination revealed no neurological deficit.

Routine investigations were all normal. Fine needle aspiration cytology (FNAC) was attempted without success. Computed tomography (CT) scan, of the head and neck region, showed an ill defined, heterogenous, enhancing mass in the right submandibular area with erosion and expansion of the hyoid bone and extension to the right parapharyngeal space (Figure 2). A Magnetic resonance imaging (MRI) scan showed a well defined, iso-intense, moderately enhancing mass lesion measuring 3.2 cms x 2.2 cms, occupying the right retromandibular region, attached to the hyoid bone and pressing on adjacent structures (Figure 3). The mass showed heterogeneous signal on T2W1 sequence. No significant lymphadenopathy was noted and the rest of the neck appeared unremarkable.



**Figure 1.** Mass in submandibular region extending to the retromandibular area.



**Figure 2.** Pre-operative axial computed tomography imaging demonstrating ill defined, heterogeneous enhancing (1) and remaining hyoid bone (2).



**Figure 3.** Pre-operative MRI scan showing a well defined, iso-intense, moderately enhancing mass lesion (1) presenting on adjacent structures



**Figure 4.** Operative photograph showing an ovoid mass (arrow) with well defined rounded margins arising from the hyoid bone with no extension into the surrounding soft tissue.

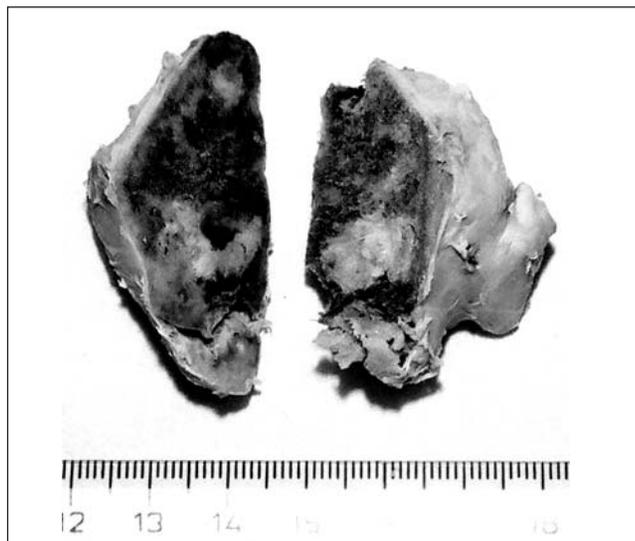
The case was discussed with our pathologist and an excision biopsy was planned. Under general anesthesia and endotracheal intubation, the mass was approached through an incision 2 cm below the mandibular margin. On exposure, a hard ovoid mass with well defined rounded margins was encountered arising from the hyoid bone (Figure 4). The mass appeared to be extending to the surrounding soft tissue and adhered to the submandibular gland and surrounding muscles with no definite invasion. The mass was removed in toto including part of the hyoid bone. The post operative period was uneventful with no palsies. Fifteen months after surgery, the patient remained free of tumor recurrence; he was subsequently lost to follow-up.

#### Pathological findings

Gross examination of the specimen revealed a bony mass measuring 4.5x3x1.5 cm (Figure 5). This was bisected after decalcification. Its cut surface was bony in appearance and showed dark brown and grayish white areas (Figure 6). Representative sections were taken and routinely processed. The slides were stained using hematoxylin and eosin. Microscopic examination revealed proliferation of thin walled blood vessels lined with a single layer of endothelial cells interspersed with trabeculae of mature bone (Figure 7). The lumina of the blood vessels were filled with red blood cells. The marrow showed fibrosis. There was no evidence of malignancy. These features are compatible with hemangioma of the hyoid bone.



**Figure 5.** Specimen showing bony mass measuring 4.5 x 3 x 1.5 cm.



**Figure 6.** Cut surface of the specimen is bony in appearance and showed dark brown and grayish white.

## Discussion

Primary vascular tumors of bone are rare.<sup>3</sup> Hemangiomas are benign vascular lesions with four histologic variants: cavernous, capillary, arteriovenous and venous. Clinically significant symptomatic

bone hemangiomas constitute less than 1% of all primary bone neoplasms.<sup>1,3</sup> They are predominantly of the cavernous and capillary varieties and occur most frequently in the vertebral column followed by the cranium.<sup>1,2</sup> Autopsy studies have identified them in the vertebrae of approximately 10% of the adult population.<sup>3</sup> They may also arise from the maxillofacial skeleton including the mandible, maxilla and frontal and nasal bones.<sup>2,4</sup> Rare lesions involving the zygomatic bone, orbital roof, pterygoid process, and temporal bone have been reported.<sup>5,9</sup> On a review of the literature this appears to be the first report of hemangioma of the hyoid bone.

Hyoid bone is an uncommon focus for the solid primary lesions. The reported cases include in addition to a handful cases of chondroma,<sup>10-13</sup> chondrosarcoma,<sup>14-17</sup> osteoma and osteosarcoma<sup>18,19</sup> rare cases like giant cell tumor,<sup>20</sup> aneurismal bone cysts,<sup>21</sup> and plasmacytoma;<sup>22</sup> all of which need to be considered in the differential diagnosis.

Bone hemangiomas are slow growing lesions occurring typically in women in the fourth and fifth decades of life.<sup>2</sup> The lesions in the craniofacial region are usually asymptomatic, presenting with painless facial swelling.<sup>2</sup> The male to female ratio is about 2:3.<sup>3</sup> The reported case occurred in a male. However, like the craniofacial lesions it also slow presented as an asymptomatic slow growing tumor.

About the diagnosis, plain radiology is useful for evaluation as the first-line imaging modality in most cases. The lesion can be appreciated as a round opaque mass which may show a prominent trabecular pattern or may present with a non specific pattern. However CT and MRI are the imaging techniques of choice because they can clarify the tumors origin and extension. The computed tomographic appearance of hemangioma of the hyoid bone is classically of expansile, well-circumscribed areas of

rarefaction with a sunburst pattern of trabeculation radiating from a common center. When viewed en face or on axial views, a honeycomb or soap-bubble configuration is characteristic.

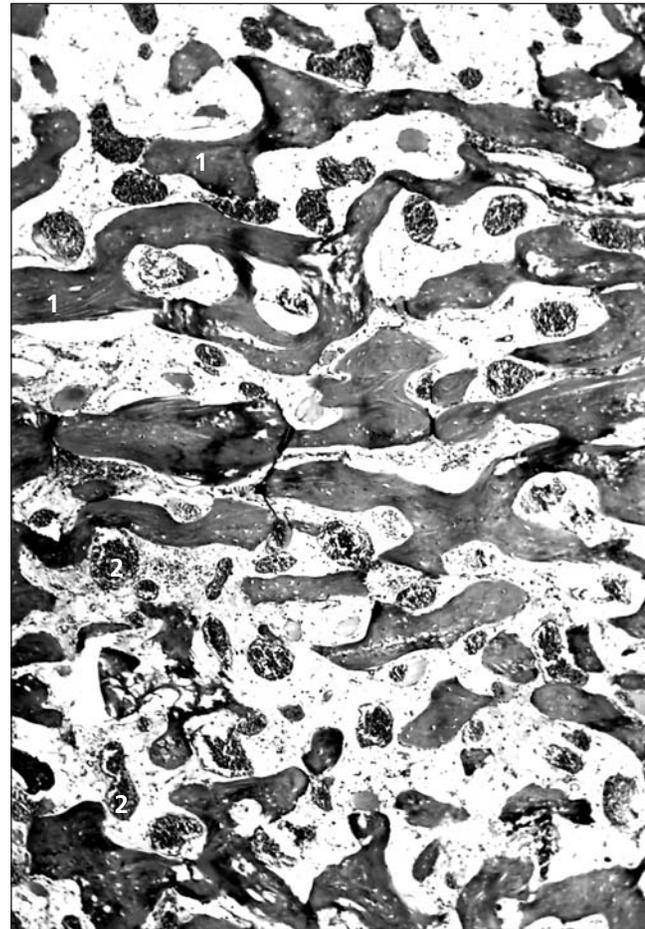
On MRI scan the signal characteristics are variable. The lesion usually appears mottled and heterogeneous. Symptomatic tumors usually show loss of fat and exhibit a low signal on T1-weighted images and a high signal on T2-weighting.<sup>3</sup>

Histopathologic confirmation of the tumor is the definitive method for diagnosis.<sup>1</sup> An FNAC&incision biopsy can be difficult or even unsuccessful, depending on tumor consistency. Though hemangiomas have variable histologic features, capillary and cavernous hemangiomas are composed of thin walled blood filled vessels lined by a single layer of flat, cytologically bland endothelial cells. The vessels permeate the marrow and surround preexisting trabeculae.<sup>3</sup>

The management consists of accurate diagnosis and surgical excision. The treatment of choice is the complete removal of the lesion along with its capsule and a wide enough border of healthy hyoid bone or total hyoidectomy. The hyoid bone provides attachment to muscles of the tongue and is also attached to the thyroid cartilage via the extrinsic laryngeal muscles and thyrohyoid membrane. Thus hyoidectomy along with tumor removal may result in changes in phonation and swallowing. However our patient did not develop any such symptoms as only partial hyoidectomy was performed. Radiotherapy and chemotherapy have no therapeutic role. The prognosis is good with a low recurrence rate.<sup>3</sup>

## Conclusion

The first documented case of hemangioma of hyoid bone origin that manifested as submandibu-



**Figure 7.** Histopathologic findings (HE x400). Bony trabeculae (1). Thin walled blood vessels (2) lined by single layer of endothelial cells and filled with RBCs.

lar mass in a male patient of 30 years with no other complaint is presented. Primary hemangiomas of bone origin are uncommon benign neoplasms and have not been reported in world literature. For the diagnosis CT scan and/or MRI are required. Also histopathological evaluation is necessary for the definitive diagnosis. The differential diagnosis is from chondroma, chondrosarcoma, osteoma, osteosarcoma, giant cell tumor, aneurismal bone cysts and plasmacytoma. Surgical excision of the tumor including hyoidectomy (partial or total) should be performed.

### References

1. **Khanam H, Lipper MH, Wolff CL, Lopes MB.** Calvarial hemangiomas: report of two cases and review of the literature. *Surg Neurol* 2001;55:63-7.
2. **Liu JK, Burger PC, Harnsberger HR, Couldwell WT.** Primary intraosseous skull base cavernous hemangioma: case report. *Skull Base* 2003;13:219-28.
3. **Fletcher CDM, Unni KK, Mertens F** (Eds.). World Health Organization Classification of Tumors. Pathology and Genetics of Tumors of Soft Tissue and Bone. Lyon: IARC Press; 2002. p. 320-1.
4. **Webb CJ, Porter G, Spencer MG, Sissons GR.** Cavernous haemangioma of the nasal bones: an alternative management option. *J Laryngol Otol* 2000;114:287-9.
5. **Ramchandani PL, Sabesan T, Mellor TK.** Intraosseous vascular anomaly (haemangioma) of the zygoma. *Br J Oral Maxillofac Surg* 2004;42:583-6.
6. **Kolasa P, Januszalwasiak, Kordek R, Boroński K.** Intraosseous orbital haemangioma. *Folia Neuropathol* 2001;39:37-9.
7. **Alba JR, Armengot M, Rausell N, et al.** Arteriovenous haemangioma of the pterygoid process. Report of a case. *Rev Laryngol Otol Rhinol (Bord)* 2002;123:175-7.
8. **Buchanan DS, Fagan PA, Turner J.** Cavernous haemangioma of the temporal bone. *J Laryngol Otol* 1992;106:1086-8.
9. **Valentini V, Nicolai G, Lorè B, Aboh IV.** Intraosseous hemangiomas. *J Craniofac Surg* 2008;19:1459-64.
10. **Özcan İ, Özcan KM, Selçuk A, Ergül G, Dere H.** Hyoid chondroma presenting as an external neck mass. *J Craniofac Surg* 2007;18:1184-7.
11. **Grayson A, Bain M.** Juxtacortical chondroma of the hyoid bone. Report of a case. *Arch Otolaryngol* 1967;86:679-80.
12. **Fedorova MA.** Chondroma of the hyoid bone. *Vestn Otorinolaringol* 1953;15:90-1.
13. **Nakagawa Y, Ishibashi K, Asada K, Sugawara N.** Chondroma of the hyoid bone: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;88:333-6.
14. **Dalla Palma P, Piazza M.** Peripheral chondrosarcoma of the hyoid bone. Report of a case. *Appl Pathol* 1983;1:333-8.
15. **Greer JA Jr, Devine KD, Dahlin DC.** Gardner's syndrome and chondrosarcoma of the hyoid bone. *Arch Otolaryngol* 1977;103:425-7.
16. **Itoh K, Nobori T, Fukuda K, Furuta S, Ohyama M.** Chondrosarcoma of the hyoid bone. *J Laryngol Otol* 1993;107:642-6.
17. **Verma S, Chokkalingam P, McRae D.** Chondrosarcoma of the greater cornu of the hyoid: a case report and literature review. *Ear Nose Throat J* 2008;87:E1-5.
18. **Weber AL, Loewenheim HM.** Osteoma arising from the stylohyoid chain and manifesting as a neck and oropharyngeal mass. *Ann Otol Rhinol Laryngol* 1993;102:399-401.
19. **Anderson TD, Kearney JJ.** Osteosarcoma of the hyoid bone. *Otolaryngol Head Neck Surg* 2002;126:81-2.
20. **Iype EM, Abraham EK, Kumar K, et al.** Giant cell tumour of hyoid bone: case report. *Br J Oral Maxillofac Surg* 2000;38:610-1.
21. **Shadaba A, Zaidi S.** Aneurysmal bone cyst of the hyoid. *J Laryngol Otol* 1992;106:71-2.
22. **Goel S, Moorjani V, Kulkarni P, Patange V, Sharma OP.** Plasmacytoma of the hyoid. *J Laryngol Otol* 1994;108:604-6.

### Conflict of interest statement:

No conflicts declared.

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