



A cavernous hemangioma of the tongue base presenting as an ectopic thyroid: A case report

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ABSTRACT

Introduction: Hemangioma is a common benign tumor of blood vessels, that rarely develops in the oropharynx. Cavernous hemangioma is characterized by thin-walled dilated blood vessels lined with endothelial cells.

Case report: We report a case of a 46 years-old woman who was admitted to our department with a complaint of chronic dysphonia and dysphagia. Clinical examination, radiological investigations and thyroid scintigraphy found a mass of the tongue base, compatible with a lingual thyroid. Treatment consisted of surgical excision through a cervical approach. The histopathological examination found a cavernous hemangioma.

Conclusion: Though hemangioma is very rare in the base of tongue, it should be a part of the differential diagnosis while investigating a lesion in the oral cavity and the oropharynx, alongside the other possibilities.

1. Introduction

Hemangiomas represent a vast and heterogeneous group of benign vascular tumors that are characterized by similar histologic features, namely a marked proliferation of blood vessels [1]. Histologically, hemangiomas can be classified into capillary and cavernous lesions, depending on the size of the vascular spaces [2–5]. Lesions containing both types of lesions are called mixed hemangiomas, and may be more common than purely cavernous hemangioma [1–5].

They can be either congenital or acquired. While most of them appear in childhood, there have been occasional reports of occurrence in adults [6–8].

Although hemangioma is one of the most common soft tissue tumors of the head and neck region, it remains relatively rare in the oral cavity, and is particularly rare in the tongue base [9,10].

We report here a Case of a 46 years-old patient who was diagnosed with a voluminous hemangioma of the tongue base, mimicking an ectopic thyroid.

This work is reported by following the surgical Case report (SCARE) guideline [11].

1.1. Case report

A 46 years-old woman, with no relevant medical history, was

referred to our department of otorhinolaryngology and head and neck surgery for chronic and persistent dysphonia associated with progressive dysphagia, which first appeared 5 years prior to her admission.

The patient reported no associated symptoms, such as dyspnea and bleeding exteriorized through her mouth.

The clinical examination of the oral cavity found a hard and painless lump located in the tongue base, measuring approximately 4 cm in diameter. The examination of the neck found no palpable lymphadenopathy.

An inspection of the upper aero-digestive tract through nasofibroscope allowed the discovery of a large pedunculated red – blue mass in the oropharynx, of which the exact anatomical origin was uncertain. The vocal cords' shape and movements were normal.

Computed tomography (CT) imaging showed the presence of an oropharyngeal tumor occupying 70% of the oropharynx, with inherent micro-calcifications (Fig. 1).

Facial magnetic resonance imaging (MRI) revealed a bulky oropharyngeal tissue mass, located in the tongue base, isointense in T1 weighted images and hyper-intense in T2 weighted images, heterogeneously enhancing after injection of gadolinium measuring 44 × 42 mm, pushing down on the epiglottis and filling the valleculas (Fig. 2).

Additionally, a thyroid scintigraphy showed a thyroid gland with normal position and function and an abnormal uptake in the tongue base. This aspect was deemed compatible with an ectopic thyroid

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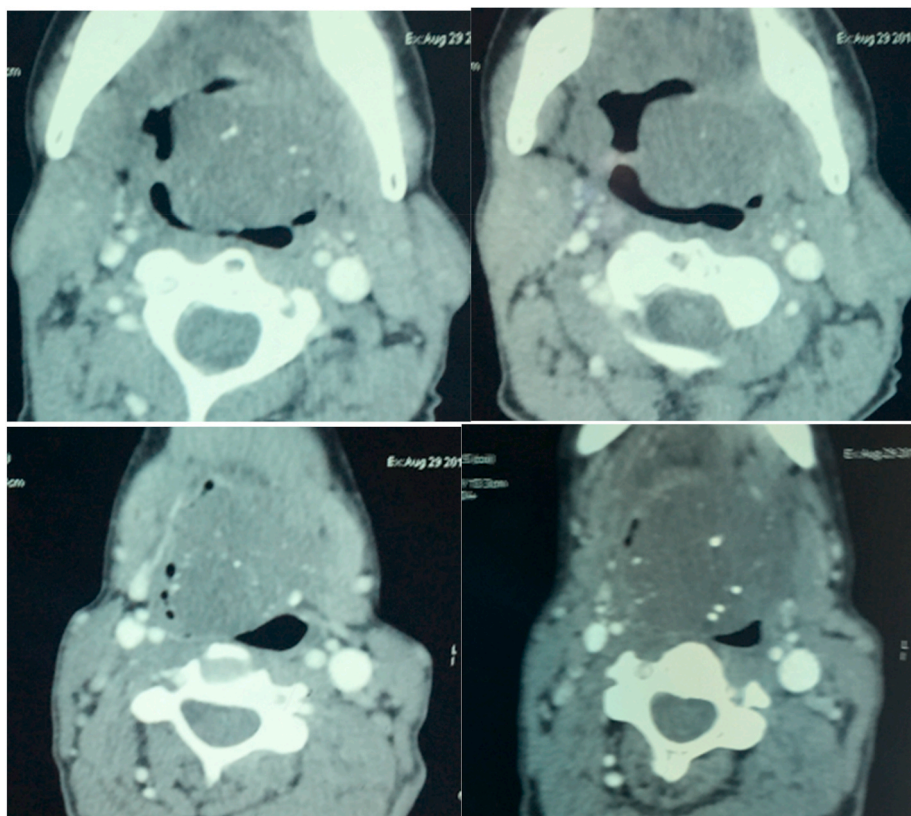


Fig. 1. Axial CT images showing the voluminous oropharyngeal mass, located in the tongue base, with inherent micro calcifications.

(Fig. 3). The normal Thyroid-stimulating hormone (TSH) confirms a euthyroid state in this patient.

Based on these investigations, the oropharyngeal mass was diagnosed as an ectopic thyroid located in the tongue base.

The patient underwent surgery under general anesthesia, which required a tracheotomy.

We informed the patient about the surgical procedure, its potential complications and the expected outcomes which were accepted. A complete excision of the mass was realized by a senior head and neck surgeon through a cervical incision (Fig. 4). The intraoperative exploration found a mass of the base of the tongue, situated above the valleculas. The tongue base was carefully preserved. No active bleeding or other intraoperative complications were reported during surgery.

Post-operative care included feeding through a nasogastric tube for the first week. Oral feeding was initiated on the 6th day after surgery. The tracheotomy cannula was removed on the 6th day as well.

The macroscopic examination of the excision piece showed a soft lobulated mass measuring 50 × 35 mm (Fig. 5).

Histopathologic analysis identified a proliferation of void capillary vessels bordered by flattened endothelial cells, in favor of a cavernous hemangioma of the tongue base (Fig. 6).

A complementary immunohistochemistry found a positive expression of CD31 and CD34, which confirmed the vascular nature of the tumor.

1.2. The post-operative TSH was normal

The patient received a 1-month check-up, which found a patient with normal speech and swallowing, and with a healed surgical wound and tracheotomy orifice (Fig. 7).

A six-month follow-up found the patient with no sign of a local recurrence of the hemangioma.

2. Discussion

Hemangiomas are presumed to be one of the most common congenital vascular anomaly [12], and are considered benign tumors characterized by three evolutionary stages: endothelial cell proliferation, rapid growth, and Involution [4,13].

Even though they are fairly common in the head and neck region, their occurrence in the pharynx, especially in the tongue base, is extremely rare [9,12,14,15].

The most frequent symptom is a difficulty to swallow or dysphagia, which can be associated to a dyspnea in Case of upper airway obstruction [14]. Other patient complains include hoarseness of the voice and oral bleeding [12]. Our patient only had chronic and persistent dysphonia.

Clinical examination will often require the use of endoscopy, since the base of tongue is not accessible to a regular examination of the oral cavity. Cavernous hemangioma usually appears as a deep red or blue elevated lesion, that can be smooth or lobulated, and sessile or pedunculated [1,3]. Its size may vary from a few millimeters to several centimeters in giant hemangiomas [5,14,16,17].

Radiologic explorations can be used to further characterization of the lesion and help to determine its nature and origin [4,18]. While CT scan is the most accessible and used imaging technique, MRI is more useful in studying the tongue base, since it is particularly efficient in the analysis of soft tissue lesions.

CT imaging provides information concerning the composition, the size and the extent of the lesion. Hemangioma is usually seen as a tumor of tissue density, with a weak enhancing pattern, which is due to intratumoral non-enhanced thrombosis [12].

On MRI, hemangioma is isointense on T1-weighted images, hyperintense on T2-weighted images, and usually shows bright enhancement after injection of gadolinium, with a varying level of homogeneity [19,20].

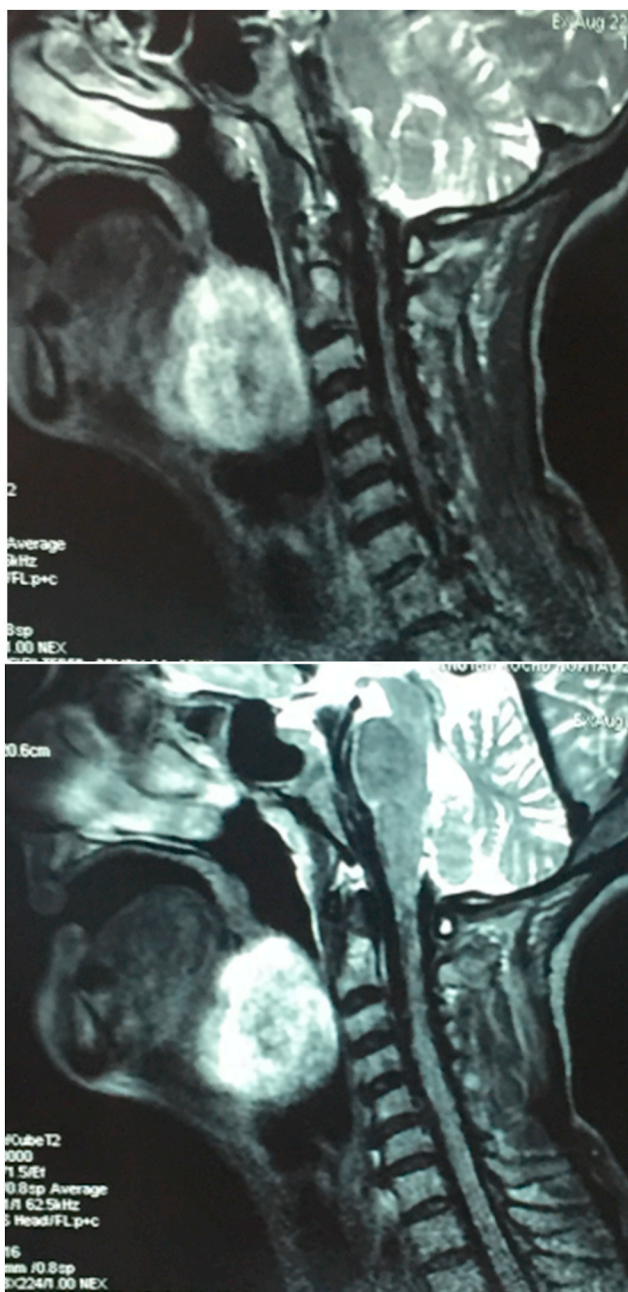


Fig. 2. Sagittal T2-weighted MRI images showing the tumor, which is hyperintense, in the tongue base.

In this particular Case, both CT imaging and MRI were used to analyze the volume and the characteristics of the tumor, since its location in the tongue base made clinical evaluation difficult.

Cavernous hemangioma can mimic clinical and radiologic aspects of other tumoral lesions. Differential diagnosis includes other type of vascular lesions, such as an arteriovenous malformation, as well as any possible etiology for a mass of the tongue base: a malignant or benign tumor, a remnant of the thyro-glossal duct, or an ectopic thyroid. In the Case presented here, the location as well as the radiologic characteristics of the mass indicated that it was probably a lingual thyroid.

Thyroid scintigraphy is usually highly specific and sensitive in identifying either normal or ectopic thyroid tissue [21]. In this Case, thyroid scintigraphy wrongly diagnosed the lingual tumor as an ectopic thyroid.

The confirmation of a hemangioma is based on histopathological

analysis, which shows well-developed, flattened capillary channels of various sizes, lined with endothelium. The cavernous hemangioma is further characterized by the presence of large and thin-walled dilated blood vessels or sinuses lined with endothelial cells, and separated by connective tissue septa or stroma [4,22]. The sinusoidal spaces are often filled with blood, though they may be void altogether [14,23].

While some hemangiomas may spontaneously disappear [24], especially in children, others will require specific treatment. Hemangiomas in adults are prone to persist over time, and do not respond well to medical treatment [12].

Several factors intervene in the therapeutic management of hemangiomas, including the age of the patient, the size and the location of the lesion, as well as its clinical characteristics [1,3,12,25].

There are many treatment modalities reported in the literature for head and neck hemangiomas, including a wait and watch policy, corticosteroid treatment, embolization, electrolysis, sclerotherapy, laser photocoagulation, immunomodulatory therapy, and surgery [4,18,26–29].

Aggressive treatments are indicated in Case of severe symptoms or functional disorders, such as airway obstruction, dysphagia, and bleeding [30].

Surgical excision for small, superficial hemangiomas is relatively easy. Bigger and deeper seated lesions, however, may call for a wider surgical approach. In this Case, the mass was large and was initially diagnosed as an ectopic thyroid in the base of the tongue, so it was surgically removed through a cervical incision.

Recurrence of hemangiomas after treatment has been reported in literature [7–31], regardless of the initial treatment used. Therefore, patients should benefit from a regular and meticulous follow-up after treatment. There is no general consensus on the recommended rhythm or duration of the follow-up in literature. Our patient has been thoroughly examined during the 1-month, 3-month and 6-month follow-ups, and no signs of a local recurrence have been found. Further check-ups are planned in the future.

3. Conclusion

Cavernous hemangioma is a benign tumor that is rarely found in adults, especially in the tongue base. Clinical symptoms include dysphagia, dysphonia, bleeding, and in extreme cases dyspnea.

While clinical findings and radiologic or even nuclear investigations may be in favor of a particular diagnosis, it is ultimately the histopathological analysis of the lesion that defines its true nature.

Consent

Written informed consent was obtained from the patient's family.

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Ethical approval

I certify that this kind of manuscript does not require ethical approval by the Ethical Committee of our institution.

Consent

Written informed consent was obtained from the patient for publication of this research study. A copy of the written consent of each patient is available for review by the Editor-in-Chief of this journal on request.

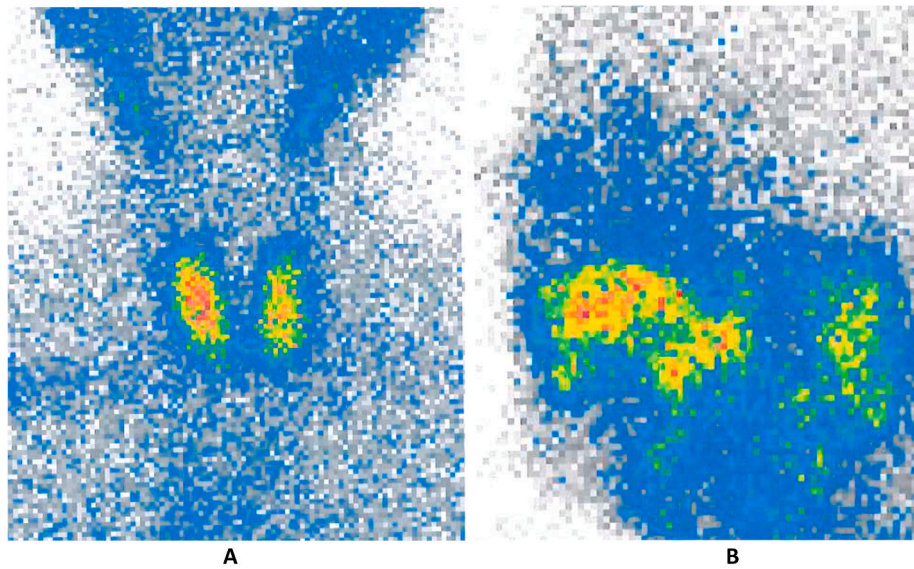


Fig. 3. (A) Normal volume of thyroid with homogeneous uptake an regular contours.
 (B) Basilingual mass with moderate uptake compatible with an ectopic thyroid

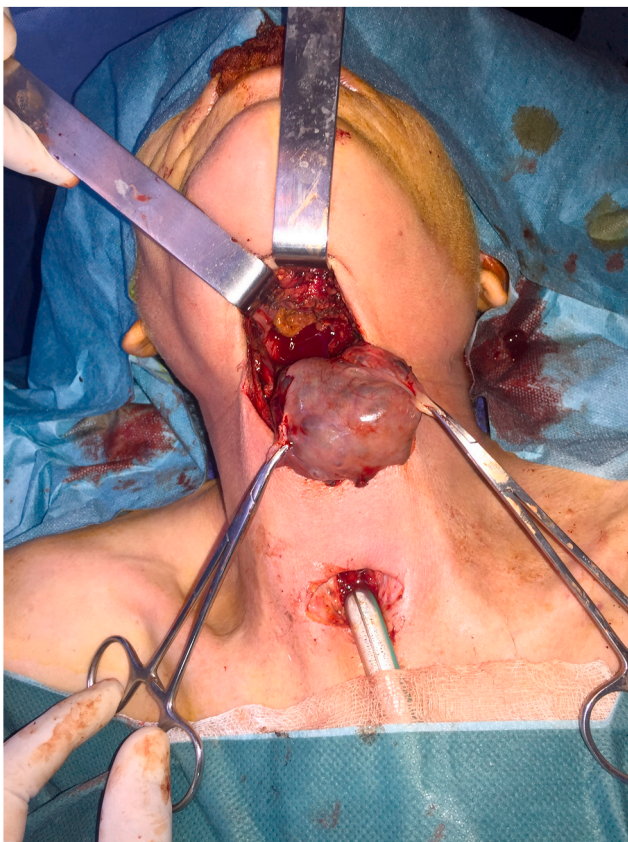


Fig. 4. Complete excision of the mass through a cervical incision.



Fig. 5. Macroscopic examination of the tumor showing a lobulated, red – blue mass, measuring 35 × 50 mm. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

S. Rouadi: drafting the article
 R. Abada: revising the article.
 M. Roubal: revising the article
 M. Mahtar: final approval of the version to be submitted

Registration of research studies

This is a Case report that does not require a research registry.

Guarantor

M. Beghdad.

Provenance and peer review

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Author contribution

M. Beghdad: conception and design of the study
 K. Choukry: conception and design of the study
 A. Mkhatri: acquisition of data
 Z. El Krimi: acquisition of data
 Y. Oukessou: drafting the article

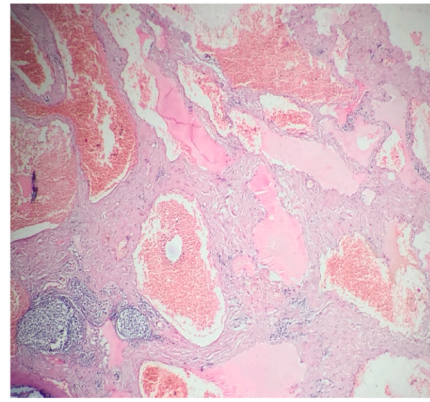
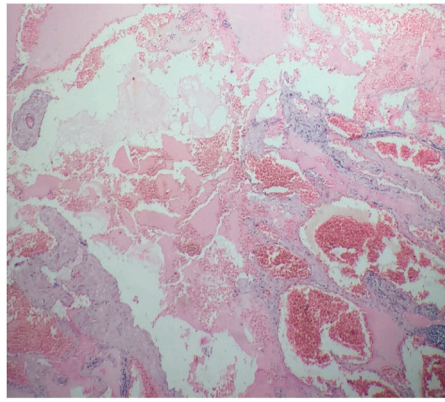


Fig. 6. Proliferation of capillary-type vascular cavities dilated and bordered by a flattened endothelium.



Fig. 7. Picture of the patient during the 1-month check-up. We can see the healed scars of the surgical incision, and of the orifice of the tracheostomy.

Declaration of competing interest

The authors declare that they have no competing interests.

Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.amsu.2020.10.053>.

The interstitial tissue is fibrous, sometimes calcified, and contains clusters of inflammatory cells.

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