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Research Article

An Unusual Case of Angiofibroma at the Base of the Tongue in a Male Child

Tangara Mariam[1, *], Dicko Ibrahim[2], Toure Tata[3], Konate Oumar[2], Ouane Aissata[2], Coulibaly Assitan Kole[2], Konate Moussa[4], Keita Moussa Bourama[2], Berthe Ismael[2], Simpara Gaoussou[2], Cisse Naouma[2], Traore Nouhoum[5], Konate N'faly[2], Diarra Kassim[2], Kone Fatogoma Issa[2, 3], Guindo Boubacary[2, 3], Soumaoro Siaka[2, 3] Singare Kadidiatou[2, 3], Keita Mohamed Amadou[2, 3]

[1]Department of ENT and Head and Neck Surgery, Mother-Child Hospital Naomi Harris, Kayes, Mali

[2]Department of ENT and Head and Neck Surgery, Gabriel Toure University Hospital, Bamako, Mali

[3]Faculty of Medicine and Odontostomatology, University of Sciences, Techniques and Technologies of Bamako, Bamako, Mali

[4]Department of ENT and Head and Neck Surgery, Main Medical Center of the Air Force, Bamako, Mali

[5]Department of ENT and Head and Neck Surgery, Fousseyni Daou Hospital, Kayes, Mali

Abstract

Angiofibroma is a benign but very aggressive tumor due to its tendency for local invasion. It is composed of myofibroblasts and vasogenic elements, making it a highly vascularized tumor. The oropharyngeal angiofibroma is an atypical angiofibroma in terms of location and symptoms. The general objective of our study was to examine the diagnostic and therapeutic characteristics of an angiofibroma with an exceptional location at the base of the tongue through a case observation accompanied by a literature review. This involves a 13-year-old patient who consulted for solid dysphagia. The onset would date back to 2 years after his admission, marked by the progressive installation of a ronchopathy associated with a feeling of a foreign body in the throat, and firm rhinophonia. This clinical picture prompted traditional treatments based on fumigations, which were unsuccessful. The evolution was marked after 3 months by the addition of solid dysphagia and dyspnea in the supine position. Faced with this clinical picture, they decided to consult us for management. The physical examination revealed a budding mass at the base of the tongue with vascular hyperemia that is mobile during tongue prostration and swallowing, with contact and palpation showing no bleeding or pulsation. A nasofibroscopy objectively showed a submucosal lesion budding at the base of the tongue, mobile during swallowing, extending towards the posterior pharyngeal wall and narrowing the oropharyngeal canal. A computed tomography scan with contrast injection suggested a hyperdense tissue mass, well-circumscribed, developing at the expense of the base of the tongue with significant obstruction of the oropharyngeal tract. The histopathological examination of the operative sample after complete transoral surgical excision under general anesthesia was in favor of an angiofibroma rearranged from the base of the language. The patient was lost to follow-up for 2 years due to financial difficulties as all the medical expenses were borne by the patient without any assistance or health insurance.

[*]Corresponding author: mariamtangara063@gmail.com (Tangara Mariam)

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Keywords

Angiofibroma, Tumor, Benign, Oropharynx, Mali

1. Introduction

Angiofibroma is a benign tumor but very aggressive due to its tendency for local invasion. It is composed of myofibroblasts and vasogenic elements, making it a highly vascularized tumor [1]. However, it has also been reported in very young ages, in elderly female patients, as well as in sites other than the nasopharynx. These rare variants have been called atypical angiofibromas [2]. Oropharyngeal angiofibroma is considered an atypical angiofibroma in terms of location and symptoms. Angiofibromas account for 0.5% of all cases of head and neck neoplasia, with an incidence ranging from 1 in 6,000 to 1 in 55,000 in the United States [1]. It seems that the incidence is higher in certain parts of the world, such as Mexico, Egypt, Southeast Asia, and the Pacific, but no significant cause has been identified yet [3]. The angiofibroma of the base of the tongue is the rarest and at the same time the most severe form, in which a benign formation develops in an area that is difficult to access. The diagnosis is challenging at an early stage due to its similarity to other hypertrophic and inflammatory pathologies of the pharyngeal region; it must always be considered as a differential diagnosis for any oropharyngeal mass, regardless of their vascularity, typical age, or sex of the patient. The presence of a single layer of cells, without the smooth muscle component, combined with the rigidity of the fibrous stroma, gives the tumor a hemorrhagic character [4]. Angiofibromas are isolated or associated with genetic disorders [5]. Surgery is considered the gold standard for the treatment of angiofibroma, and preoperative embolization has significantly reduced intraoperative complications and bleeding with minimal risk of residual disease. External management and the extent of excision may hinder lingual functions, but endoscopic or robotic surgery is not developed in our context. This justifies our clinical presentation.

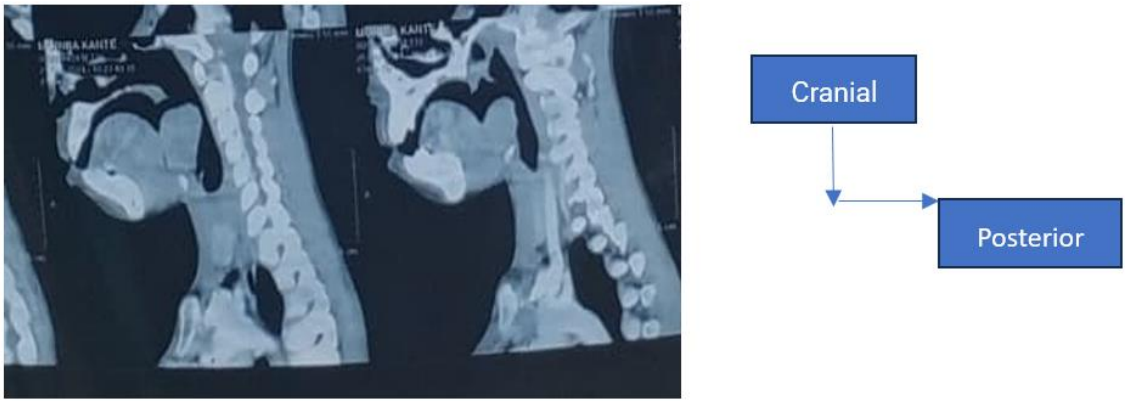


Figure 1. *Sagittal section with PC injection suggesting a large expansile tissue process, well-defined developing at the base of the tongue with significant obstruction of the oropharyngeal passage appearing benign.*

2. Observation

This is a 13-year-old male patient who consulted for solid dysphagia. The onset dates back to 2 years prior to his admission, marked by the gradual establishment and permanent evolution of a ronchopathy associated with a sensation of a foreign body in the throat, firm rhinophonia without dysphonia, dyspnea, dysgenesis, and oral bleeding. This clinical picture led to traditional treatments with fumigations without success. The evolution was marked after 3 months by the addition of solid dysphagia and dyspnea in the supine position. Faced with this clinical picture, they decided to consult the CSREF of the CIV, which referred him to us for management. He had no notable medical-surgical history. The patient is in good general condition with an OMS performance index of 01. No medical history has been reported. The physical examination revealed a budding mass at the base of the tongue with vascular hyperemia; the mass was mobile during tongue protrusion and swallowing, upon contact and palpation, without bleeding, without pulsation, with an inflammatory posterior pharyngeal wall and saliva stasis. Nasofibroscopy showed a submucosal budding lesion at the base of the tongue that

attached to the mobile tongue upon ingestion, extending to the posterior pharyngeal wall and narrowing the oropharyngeal tract. Laryngeal mobility was good. The rest of the physical examination was normal. The computed tomography with contrast injection suggested an expansive tissue process developing at the expense of the base of the tongue with significant obstruction of the oropharyngeal tract appearing benign.

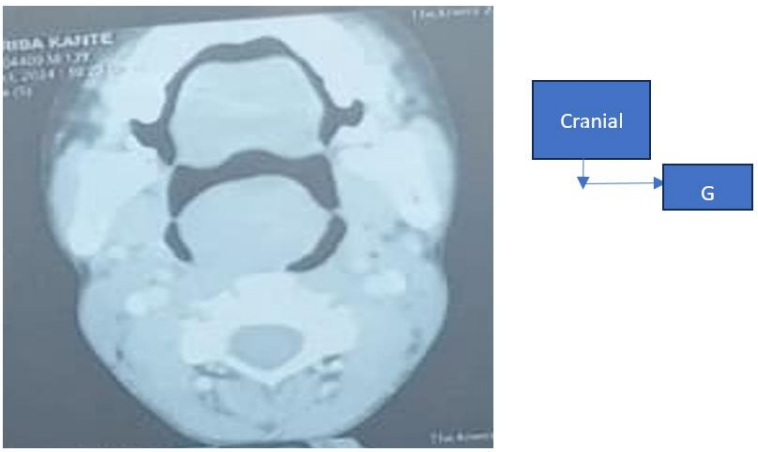


Figure 2. *Axial section with PC injection suggesting tissue hyperdensity at the expense of the base of the tongue, with regular contours and significant obstruction of the oropharyngeal pathway.*

The laboratory tests and a chest X-ray were unremarkable. Angiography and embolization were not available. After obtaining verbal consent from the guardian, we performed a complete excision of the mass through the transoral approach under general anesthesia. The patient was positioned in the supine position, with a bolster under the shoulders and a headrest under the head, under general anesthesia with orotracheal intubation, setting up the mouth gag and tongue depressor allowed for complete exposure of the mass. The tumor was hypervascular, non-bleeding, with regular contours, non-pulsatile, non-blowing, of firm consistency, measuring 4 cm in diameter. We proceeded to grasp the mass with a grasping forceps via the transoral route. We then made a fine circular incision on the mucosa followed by dissection of the submucosa until complete sectioning of the tumor was achieved with an electric scalpel. There was no loss of anatomical substance, two separate stitches were made with vicryl 4/0 thread to ensure hemostasis, there was minimal bleeding (around 30cc). The post-operative effects were simple. The patient was placed under observation in the ward for 5 days with analgesics (paracetamol 15 mg/kg every 6 hours) and a short-term corticosteroid treatment (Prednisolone 1 mg/day). The histopathological examination of the surgical sample revealed a proliferation composed of vascular clefts associated with a fascicular proliferation of fibroblasts. In some areas, the tumor is infiltrated with lympho-plasma cells. The conclusion was an angiofibroma of the base of the tongue.

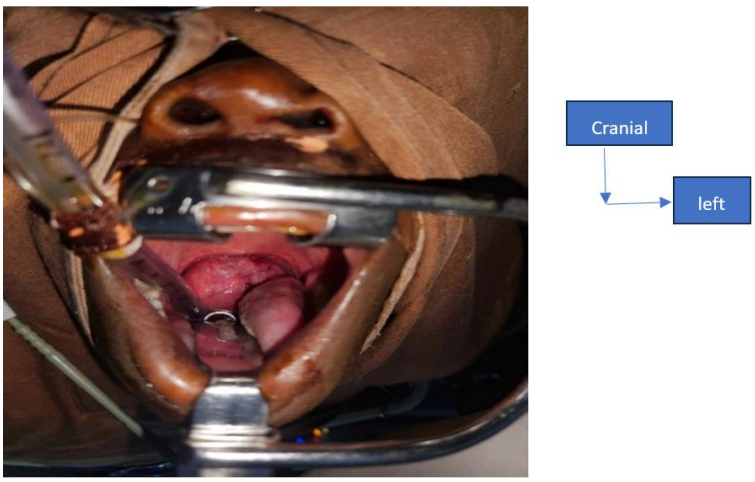


Figure 3. *Exposure of the tumor in the oropharynx during surgery.*

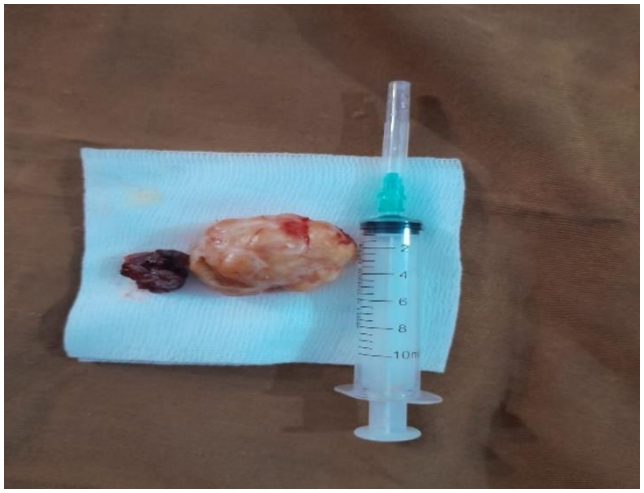
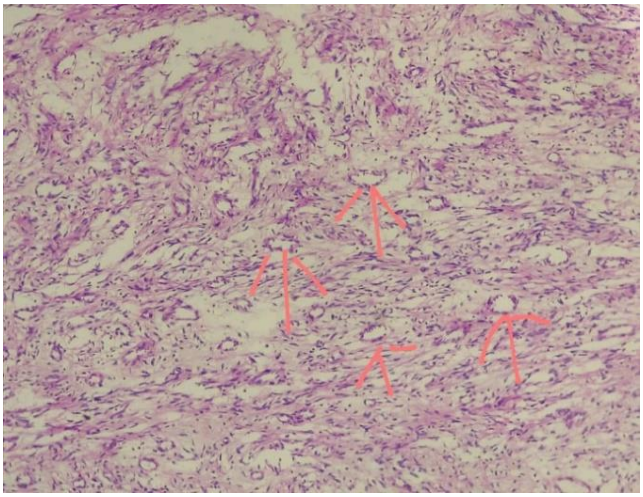
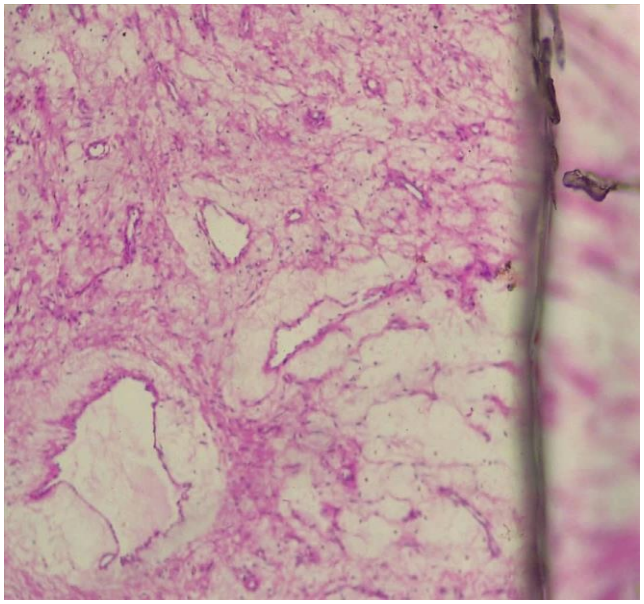


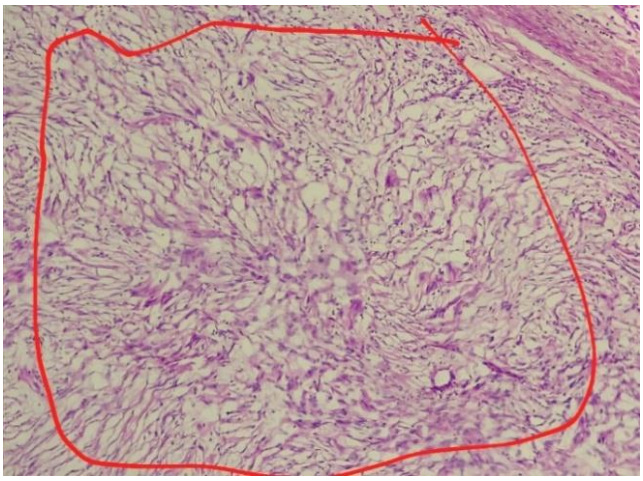
Figure 4. *Image of the surgical site after complete excision.*



C. Spindle cells or fibroblasts

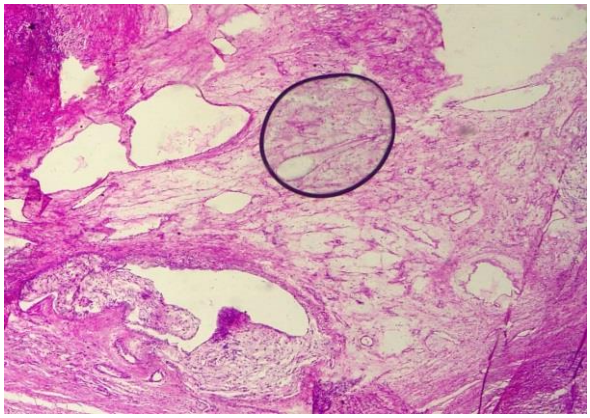


A. The well-defined fibrovascular tissue and a proliferation composed of fibroblasts with fleshy and visible endothelial cells.



D. Presence of lymphoplasmocytes around the

Figure 5. *Histological images.*



B. Infiltration of lymphoplasmic cells

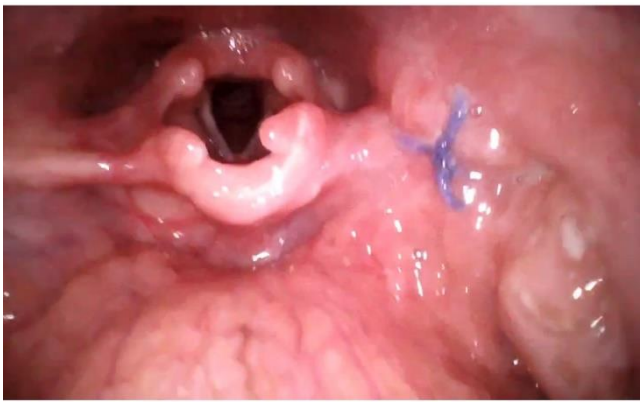


Figure 6. *Nasofibroscopic image of the base of the tongue J15 postoperatively.*

3. Discussion

Angiofibromas are rare benign vascular neoplasms that have a strong predilection for the nasopharynx. Although many theories have been proposed about their origin, current evidence indicates that angiofibromas do not originate from the nasopharynx, as is commonly assumed, but rather from a fibrovascular nidus in the retro-lateral wall of the nasal cavity near the sphenopalatine foramen [7]. It represents less than 0.05% of all head and neck tumors, with an incidence of 1 in 6,000 to 1 in 55,000 in the United States [8]. The extranasopharyngeal fibroma angioma (NEA) is a rare entity. Compared to AJN, NEA occurs at an older age, with a mean age of 22 years, and the male-to-female ratio was 2.75 [9]. The first case was reported by Beeden AD in 1971 [10]. The fibrous angioma at the base of the tongue is distinct, extremely rare, and has not been frequently reported by authors. The current literature on the subject mainly consists of individual case reports. In Mali, this is rarely reported. Despite this data from the literature, our study reports an unusual location in a 13-year-old boy. Among the 174 cases examined by Windfur and Vent until 2015, only 9 cases originated from the oropharynx. Celik et al. proposed in 2005 that angiofibromas presenting at least one of the following criteria, such as an origin or location other than nasopharyngeal, complaints other than nasal obstruction or epistaxis, an age younger than seven years or older than 25 years, female. Sex, atypical histopathology, and multifocality were considered atypical [2]. Our subject met the criteria via 2 parameters such as: the location and complaints, and is therefore labeled as an atypical angiofibroma. The angiofibroma of the base of the tongue becomes symptomatic due to a mass effect on the upper airways and adjacent structures. In the literature, angiofibroma clinically presents as a globular, well-defined, and non-encapsulated mass; covered by mucosa, the color can be pink-burgundy or pale and whitish [4]. In our observation, the mass was encapsulated according to histological results. The patient's symptoms were primarily marked by dysphagia, severe dyspnea, firm rhinolalia, ronchopathy, and miscarriages; which was consistent with the literature [11]. The angiofibroma is not associated with risk factors; it is often linked to a genetic predisposition [5]. In our clinical case, no medical-surgical history or alcohol-tobacco lifestyle was found. Computed tomography and MRI were recommended as optimal diagnostic techniques for the evaluation of AEN [12]. However, signs of suspicion of hypervascularization indicate the need for arteriography and carotid angiography before surgical procedures to organize necessary precautions, such as embolization, and to reduce the risk of rapid bleeding during tumor removal. A cervical CT scan was performed on our patient, which clarified the exact location, size, borders, and extent of the tumor. These results are consistent with the literature. Although preoperative imaging studies can provide some basis for the diagnosis of an angiofibroma, it can ultimately only be diagnosed through histopathological examination [13]. For a pathologist, while the precise diagnosis of an angiofibroma is not overly difficult, when its location is extremely rare, immunohistochemical analysis will aid in its diagnosis. Note that immunohistochemical analysis was not available in our technical platform. According to Shweta, Mittal et al. [14], the histopathological examination revealed a lesion with a complex mixture of blood vessels, irregular fibrous stroma with an edematous area and loose fibrous tissue, as well as multinucleated stromal cells that suggested an angiofibroma. In some areas, the tumor is infiltrated with lymphoplasmacytes; moreover, classic features, variations in microscopic results can rarely be observed in AF. These different characteristics do not alter the histological diagnosis. In our observation, the examined samples were the site of a proliferation consisting of vascular septa associated with a fascicular proliferation of fibroblasts. Complete surgical excision of the mass is the treatment of choice, preventing any recurrence. The aim of surgery is to completely remove the tumor while preserving the integrity of the tongue and restoring tongue function [15]. The surgical approach depends on the size, accessibility, and extent, so endoscopic laser surgery or instrumental microsurgery is indicated for small angiofibromas and external surgery for larger tumors [16]. Robot-assisted transoral surgery is another recently described therapeutic modality for the excision of angiofibroma with oropharyngeal localization according to a study by Nicola ïP, Villaret AB, Farina D, et al [17]. We performed a transoral endoscopic approach in our study, which allowed us to achieve complete excision of the tumor. A lignified transformation has been reported in the literature in cases of JNA, with radiation therapy being the main cause [18]. However, in cases of ENA, no malignant transformation has been reported to date. Long-term follow-up is necessary to ensure that there are no signs of recurrence. X-ray endovascular occlusion is used when complete surgery is not possible as a first step in radiation therapy. The application of the method reduces the volume of intraoperative blood loss. Radiation therapy may be effective in about 50% of cases of angiofibromas but may be accompanied by a large number of complications. For this reason, it is only used if it is not possible to perform a complete surgical procedure [6]. Recurrence is a constant concern in these patients and the literature reports recurrence rates ranging from 6% to 24% [19-20]. The absence of a tumor capsule, which prevents surgical cleavage, and the tendency to disseminate into the submucosa are the main causes of recurrence in surgically treated cases [21]. The failure of radiotherapy is generally attributed to the so-called 'geographic misses' in the radiation area and insufficient doses. In our study, after a follow-up of 2 weeks: the surgical wound was well healed, the tongue function was preserved, and the nasofibroscopy performed was normal. The 2-year follow-up after mass removal showed no tumor recurrence. The recurrence rate of angiofibromas associated with tuberous sclerosis can reach 80% [22].

This pathology can resemble other ENT conditions such as

cysts or lipomas and fibrous tumors (see Table 1).

Table 1. *Differential diagnosis table.*

PATHOLOGIES	Clinics	TDM	Histology	References
Angiofibroma	Dysphagia, severe dyspnea, firm rhinolalia, bronchopathy, and miscarriages, An emerging mass at the base of the tongue, hyperemic, vascular, mobile, without bleeding or pulsation.	A hyperdensity of tissue at the expense of the base of the tongue, with regular contours and significant obstruction of the oropharyngeal pathway.	The examined samples were the site of a proliferation consisting of vascular septa associated with a fascicular proliferation of fibroblasts. In some places, the tumor is infiltrated with lymphoplasmacytes.	Our article
Cysts	Snoring, without true dyspnea, a rounded, firm regular swelling forming a bulge covered with normal mucosa.	Median mass syndrome coming into contact with the hyoid bone, a liquid density and a capsule.	Cystic cavity bordered by a richly vascularized fibrous wall containing slightly eosinophilic hematic serosity punctuated by inflammatory elements and histiocytes.	Guerina; N. [23]
Lipomas	Lipomas generally appear deep in the soft tissues, rarely located under the mucosa; a yellowish color is visible through the stretched mucosa, which also contains some telangiectasias.	A negative fat density with sharp, generally homogeneous contours and without contrast uptake.	Presents adipose cells. With a few exceptions, they are surrounded by a fibrous capsule, or they are at least clearly delineated from the surrounding tissue, on which they generally rest with a wide non-pedunculated (sessile) base.	Bassetti, R., and al. [24]
Fibrous tumor	Dysarthria, dysphonia, dysphagia, bleeding and/or diagnosis may be present, with a well-defined, rounded, smooth, pale submucosal nodular mass.	Shows a hypodense area in the center and an increased fixation of the CT at the periphery.	Numerous thin-walled vessels often made of deer antler associated with a proliferation of spindle-shaped cells with hypercellular and hypocellular areas.	Betsalel, AD. [25]

4. Conclusion

Since the angiofibroma of the base of the tongue is an atypical angiofibroma in terms of location and symptoms presented, it poses a diagnostic and therapeutic challenge. Computed tomography (CT) and MRI have been recommended as optimal diagnostic techniques for the evaluation of this tumor. Complete surgical excision of the mass is the treatment of choice, preventing any recurrence. The definitive diagnosis of angiofibroma is made through histopathological analysis. The prognosis is excellent provided that complete excision is performed.

Abbreviation

AF	Angiofibroma
JNA	Juvenile Nasopharyngeal Angiofibroma
ENA	Extra-nasopharyngeal Angiofibroma
ATCD	Medical History
MRI	Magnetic Resonance Imaging
CT	Computed Tomography
WHO	World Health Organization

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Conflicts of Interest

The authors declare no conflict of interest.

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