pISSN 2454-5929 | eISSN 2454-5937

## **Case Report**

DOI: http://dx.doi.org/10.18203/issn.2454-5929.ijohns20171207

# An 80 year old lingual haemangioma: a case report

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Received: 24 January 2017 Accepted: 01 March 2017

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#### **ABSTRACT**

Haemangiomas are benign vascular tumours which are commonly encountered in head and neck regions. They are subdivided in cavernous haemaniogma and vascular malformations. Lingual haemangiomas pose a potential airway risk because of their position. This case report is about the management of a patient with a lingual haemangioma which was asymptomatic for 80 years but which was eventually removed in view of recurrent angio-oedema.

Keywords: Lingual haemangioma, Angio-oedema, Airway, Cavernous hameangioma, Vascular malforamation

#### INTRODUCTION

Haemangiomas are benign vascular tumours which affect the head and neck region in about 50% of cases. Management is dependant on patients' symptoms. Not all haemangiomas require removal. Among the different sites of head and neck haemangiomas, those that present on the tongue require special consideration in view of it being a mobile organ resulting in increased susceptibility to minor trauma and hence bleeding and ulceration. They are also associated with chewing and swallowing difficulties, breathing and cosmetic concerns. <sup>1</sup>

An 80 year old lady with a history of well-controlled hypertension, hypercholesterolaemia and smoking presented to the Otorhinolaryngology, Head and Neck Surgery Department at Mater Dei Hospital with a recurrent history of angio-oedema.

#### **CASE REPORT**

The Otorhinolaryngology, Head and Neck Surgery Department at Mater Dei Hospital with a history of recurrent angio-oedema.

She had 3 episodes of angio-oedema over the previous few months. It was noted that she was on oral angiotensin converting enzyme-inhibitors.

Intra-oral examination revealed a large, well-circumscribed lesion on the right side of the tongue, covering much of its anterior two-thirds, measuring 37 mm×32 mm×8 mm. The lesion had a bluish hue and was soft to palpation, with an irregular surface but no ulceration or active bleeding. A provisional diagnosis of a lingual haemangioma was made based on these clinical findings. The patient claimed that this had been present

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since birth and that it was never bothersome. She could speak and eat normally.

She was actively involved in a discussion about her management options. It was decided that, apart from a change in her anti-hypertensive treatment, this large lingual haemangioma also needed to be addressed as, in view of the history of recurrent angio-oedema, it posed a potential airway hazard.

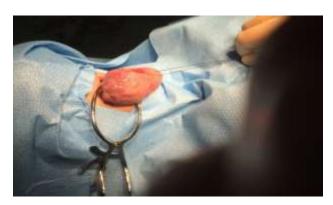


Figure 1: Photograph of patient intraoperatively prior to excision- lateral view.

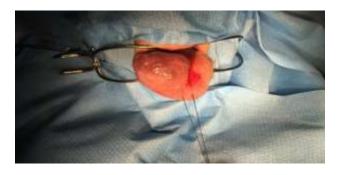


Figure 2: Photograph of patient intraoperatively prior to excision- dorsal view.

It was thereafter decided to surgically excise the lesion under general anaesthetic. A HARMONIC WAVE® open shears was utilised for the dissection, which was carried out without any immediate or delayed sequelae. The residual defect of the surface of the tongue was closed with absorbable sutures, which also helped with haemostasis.



Figure 3: Photograph of excised lingual haemangioma.

The patient was discharged on the first postoperative day. Pain was managed with oral paracetamol and codeine. NSAIDs were avoided in view of her co-morbid conditions. She was reviewed in clinic 4 days postoperatively and again at 6 weeks after surgery.



Figure 4: Postoperative appearance of tongue 4 days postoperatively.

Histologic examination revealed a cavernous haemangioma with multiple organising thrombi which did not exhibit any evidence of atypia or malignancy.

#### **DISCUSSION**

The head and neck region is the most commonly affected site by haemangiomas, especially the face, oral mucosa, lips and tongue. These lesions traditionally show a higher prevalence in females with a 3:1 ratio.<sup>2</sup>

Haemangiomas are one of two types of vasoformative tumours. The other type is vascular malformations, which is generally the main differential diagnosis for haemangiomas. Haemangiomas are histologically further classified into capillary and cavernous forms. Capillary haemangiomas are composed of many small capillaries lined by a single layer of endothelial cells supported in a connective tissue stroma of varying density, while the cavernous type is formed by large, thin walled vessels, or sinusoids lined by epithelial cells separated by thin layer of connective tissue septa.2 A provisonal diagnosis of haemangioma was done based on the clinical findings, with a differential diagnosis of arteriovenous (AV) malformation. Haemangiomas are often circumscribed lesions which rarely affect bone and are most commonly present on the tongue, lips and buccal mucosa. The lesion in this case was a well-circumscribed lesion on the ventral surface of the tongue. On the other hand, AV malformations, are poorly circumscirbed lesions which may affect bone. These are also associated with significant intra-operative haemorrhage that can make resection more difficult. There was no extensive haemorrhage during the resection of the lesion in this case, further supporting the diagnosis of haemangioma as opposed to an AV malformation. In our case, on histology, the lesion was indeed a haemangioma, of the cavernous type which was composed of irregular

anastomosed vascular channels containing multiple organising thrombi.

Management of haemangiomas depends on a variety of factors, and most true haemangiomas require no intervention. However, 10-20% require treatment because of the size, specific location, stages of growth or regeneration.<sup>2</sup> They might also need to be removed if they are causing disabling symptoms, such as increase in size, bleeding, difficulties in word articulation, dysphagia, difficulties in chewing, pain and airway compromise secondary to tongue enlargement. In our case, the lesion had to be removed in view of the recurrent episodes of angio-oedema, which together with the lesion's site and size on the tongue were posing a high risk for airway compromise.

A number of treatment options are available in the management of these lesions, comprising both medical and surgical interventions. Medical management includes systemic and intralesional administration of corticosteriods.<sup>3</sup> Pranitha et al describe a case of a 2 year old boy who had tongue haemangiomas treated successfully with prednisolone sodium syrup.1 Conservative or further aggressive forms of treatment modalities are available for haemangiomas of the tongue. These include embolisation, excision, cryotherapy, sclerotherapy, radiotherapy, laser photocoagulation and chemotherapy. Kucuk et al describe a case of a 32 year old female with a huge congenital haemangioma of the tongue who underwent sclerotherapy and radiofrequency ablation in the first instance, but since the tumour did not regress. definitive surgery with a partial hemiglossectomy was performed.<sup>3</sup> In another case, a 48 year old female with a tongue cavernous haemangiona was successfully managed with sclerotisation with ethanol via direct puncture, without proceeding to the previously planned surgery.4 In a different case, a sclerosing agent was administered topically and the mass observed for a period of one week. Since there was no change in appearance and size of the mass, surgical excision under local anaesthesia was performed.<sup>5</sup>

Risk benefit analysis needs to be performed when choosing a patient's treatment modality; namely focusing on the patient's comorbid state, risk of general anaesthesia, local expertise, recurrence rates, functional preservation, potential airway risk and cosmesis. Patients should be able to take an informed decision as to what treatment modality is chosen. In this case, we opted to proceed directly to definitive surgical excision due to the size of the lesion and the associated risk of airway compromise if the patient had to suffer another episode of angio-oedema.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Agius M, Chircop DM, Cutajar JC, Said ME. An 80 year old lingual haemangioma: a case report. Int J Otorhinolaryngol Head Neck Surg 2017;3:434-6.