

CASE REPORT

**HEMANGIOMA OF THE TONGUE:
A CONSERVATIVE TREATMENT WITH CIRCUMFERENTIAL LIGATION**

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تعتبر الأورام الدموية التي تصيب نسيج الفم اللينة الأكثر شيوعاً عند الأطفال
والبالغين .
تظهر هذه الأورام عادة عند الولادة إلا أنها تصبح واضحة سريراً في نهاية
مرحلة الطفولة .
يشكل علاج هذه الآفات تحدياً للجراح .
تناقش هذه الورقة ثلاث حالات لأورام دموية كبيرة الحجم في اللسان تم علاجها
بنجاح عن طريق الربط المحيطي .

Hemangiomas affecting the soft tissues of the oral cavity represent the most common growths in infancy and childhood. They are usually present at birth but become clinically obvious in late infancy or early childhood. Treating those lesions pose a challenge to the surgeon. This paper presents three cases of extensive hemangiomas of the tongue which were treated successfully by circumferential ligation.

Introduction

The oral cavity, head and neck regions possess complex, rich and intricate blood vessels which might be a predisposing factor for a variety of vascular lesions. These lesions represent the most common growths in infancy and childhood, and may vary from small innocent birthmarks to large disfiguring tumours. Based on their clinical behaviour and the endothelial cell characteristics,^{1,2} these lesions have been recently categorized into two groups as vascular malformation or hemangiomas. It

is agreed that vascular lesions originate from a persistent angioblastic tissue that normally reorganises and regresses.³

Vascular malformations are present at birth but become clinically obvious in late infancy or childhood. They tend to grow correspondingly with the child. Infection, trauma and endocrine changes tend to affect their size and they do not have tendency to involute. They are characterized by a normal endothelial cycle and a normal number of mast cells.

Hemangiomas usually appear 2-4 weeks after birth, grow rapidly till the age of 6-8 months and then slowly. By age 5-8 years, they start to involute and spontaneously regress in 70% of the

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cases.⁴ They are characterized on the cellular level by increased endothelial cells turnover, a number of mast cells during rapid growth phase, diminished cellularity with fibrous tissues and low mast cell counts during involution. The most common incidence sites in the orofacial region are the tongue, cheek and lips. Females appear to have a slightly higher incidence than males.⁵

This paper discusses three cases of extensive tongue hemangiomas treated with circumferential ligation with satisfactory results supporting this particular approach in the management of these lesion.

Materials & Methods

Table 1 shows the distribution of three cases with patients' ages as 6, 13 and 25 years age. One was a male child and the other two were females with a history of swelling involving the tongue. The chief complaint in two cases were repeated bleeding from the lesion while the main complaint in the third case was speech difficulty. The right side of the tongue near the tip was involved in one case. The left side was involved in

the other two cases and the lesion was occupying almost the whole anterior 2/3 of the tongue. The lesions have been present at birth and have gradually increased in size. In one patient, the mass became stationary and started to regress to reach a size of 1x2 cm. The mucosal covering was dark purple in colour with no break or ulceration. All patients had bilateral external carotid angiograms to establish the presence of a main feeder.

All were negative and no main feeders were found. Although the main complaint in two of the cases was recurrent bleeding from the lesion, all had normal blood pressure with normal Haemoglobin and red cell count. All other investigations were within normal [Figs. 1, 2 and 3].

Using general anaesthesia, the lesions were isolated and multiple ligatures were placed around the periphery of the lesions in an attempt to tie the small feeder vessels circumferentially with 3.0 black silk sutures. Multiple intratumoural ties were placed randomly, draining most of the accumulated blood **and** consequently, the tongue was reduced and consequently, the tongue was reduced to normal dimensions [Figs. 3 and 4].

Table 1. Patient Distribution.

Patient	Age	Sex	Site	Chief Complaint	Investigation	Treatment
1	26	F	Lateral border and dorsal of left tongue	Repeated bleeding	Selective external carotid angiogram	Circumferential ligation twice
2	6	M	Lateral border anterior 2/3 left tongue	Severe bleeding episodes	Left external carotid angiogram	Circumferential ligation 75%
3	16	F	Tip and lateral border right side of left tongue	Speech difficulty	Selective external carotid angiogram	Circumferential ligation followed by excision



Figure 1. Left lateral border of the tongue with large, dark, red lesion raising the dorsum.



Figure 3. Circumferential ligation in an attempt to block feeder vessels.

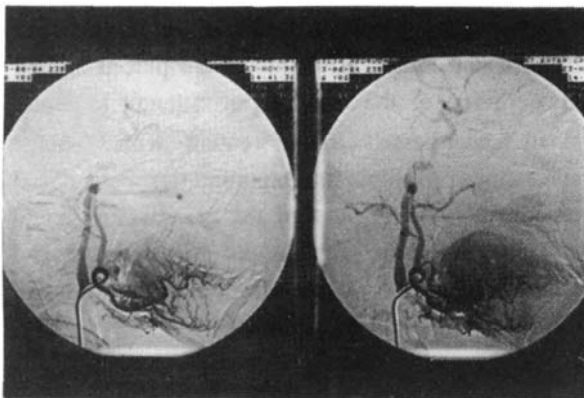


Figure 2. An angiogram of the right and left lingual arteries with increased vascularity on the left side with no feeder identified.

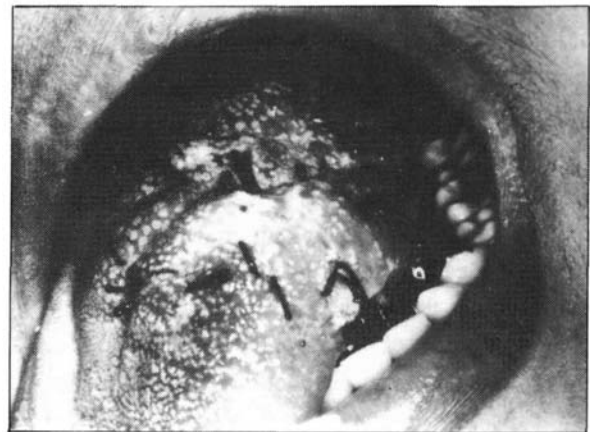


Figure 4. An intra-tumoral ligation with the tongue mass reduced to normal size and the presence of papillae evident.

Results

Over a period of 4 weeks, the lesions considerably diminished in size and the patients were able to return to normal function. By eight weeks, in two cases, the lesions almost disappeared completely and the tongue attained a normal appearance and the mucous membrane became pink in colour and normal in consistency [Fig. 5]. For the third case, although the mass was reduced by more than 50%, it seems that another intervention is needed.

Discussion

Hemangiomas, because of their tendency to involute and regress spontaneously, are generally managed conservatively, usually by close follow up and observation.⁴ Intervention is indicated only if the lesion starts to bleed, ulcerates, or interferes with function.⁶ Several treatment modalities are suggested for these lesions including surgical excision with blade or laser, cryosurgery, injection of corticosteroids or sclerosant (Sodium tetradecyl), radiotherapy, and embolization with steel coil, gel foam, silicone beads or cyanoacrylate.^{7,10}



Figure 5. The tongue, six weeks later, with small residual lesion and normal texture.

These above treatment modalities have notable disadvantages including excessive bleeding, fibrosis, scarring, cosmetic and functional deficiency due to the long term effect of steroid and radiation in children. Surgical excision and cryosurgery are effective treatment for small lesions and superficial ones, however. The technique of circumferential (intratumoral) ligation takes advantage of the easy accessibility of the feeders vessels. Several stitches are inserted through the tissues beyond the margins of the lesion isolating it even if the feeders are not readily identified. This approach is a simple, slightly invasive and nondeforming method of eliminating or reducing the lesion size.¹¹

Circumferential ligation tends to overcome the problems associated with other treatment modalities with minimal or no reconstructive procedures required to improve function and aesthetics. The efficacy of the procedure lies in the fact that it intercepts the arterial and venous connections with the regional circulation. Obstructing the vascular channels to and from the tumour, eliminates the blood flow leading to stasis and blood clots between the ligations and progressive atrophy of the vascular endothelium and the substitution of the angiomatous tissues by a fibro-connective tissue mass.¹² The clinical significance of

oral hemangiomas is based on their degree of invasion, involvement of the adjacent anatomic structures and their lack of encapsulation which inhibits and makes eradication hazardous.

Conclusion

While surgical removal of oral hemangiomas is indicated for small lesions, its use with large lesions lead to extensive tissue defect and rapid bleeding which might be difficult to control. Cryosurgery, on the other hand, is very effective in small superficial lesions but is completely ineffective in large deep ones. Embolization, utilizing various materials, is used to obstruct the vessels, with excellent results but requires considerable expertise from the technical point of view and serious complications such as tearing the vessels and overdilatation might occur. Circumferential ligation is therefore a simple technique requiring minimal experience while aiming at achieving strangulation when placing the ligature without cutting through the tissues.

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