# EXTRA-ORAL HEMANGIOMA IN A 10 YEAR OLD GIRL – A CASE REPORT WITH LITERATURE REVIEW

R.Kannan[1] B. Sekar [2] S.Murali [2] Dominic Augustine[2]

*[1]Department of Oral and Maxillofacial Surgery, [2]Department of Oral and Maxillofacial Pathology, Vinayaka Missions Sankarachariyar Dental College & Hospital, Salem - 636308. Tamilnadu, India.*

**Corresponding Author:** Dominic Augustine, Post Graduate Student,Department of Oral and Maxillofacial Pathology, Vinayaka Missions Sankarachariyar Dental College & Hospital, Sankari main road, Ariyanoor, Salem - 636308. Tamilnadu, India. Ph: 09629495716, Email: dominic2germain@gmail.com

**Abstract**

The term hemangioma has traditionally been used to describe a variety of developmental vascular anomalies. In recent years, great progress has been made in the classification and understanding of these vascular lesions. Currently, hemangiomas are considered to be benign tumors of infancy that are characterized by a rapid growth phase with endothelial cell proliferation, followed by gradual involution. Most hemangiomas cannot be recognized at birth, but arise subsequently during the first 8 weeks of life. On the other hand, vascular malformations are structural anomalies of blood vessels without endothelial proliferation. A case of extraoral hemangioma affecting the left cheek of a 10 year old female is presented here with management and literature review.

Keywords: Hemangioma, tumor of infancy, vascular anomalies.

## Introduction

Benign vascular lesions are a consequence of blood vessel abnormalities or endothelial cell proliferation. The International Society for the Study of Vascular Anomalies (ISSVA), in 1996, approved a classification system modified from the one proposed by Mulliken, Glowacki. [1] The diseases were subdivided into (a) tumors: hemangioma (HEM), pyogenic granuloma, rapidly involuting congenital hemangioma, noninvoluting congenital hemangioma, hemangiopericytoma, tufted angioma and kaposiform hemangioendothelioma; and (b) vascularmalformation(VM).[2]

HEM is a benign proliferation of endothelial cells. It is the most common neoplasm of infancy. HEM frequently is not present at birth and develops in phases: a rapid growth phase with endothelial cell proliferation followed by gradual involution.[2] It presents as a red macula, papule or nodule, depending on the congestion degree and on how deep it is in the tissue. Although HEM is a benign lesion, in some cases, it may lead to compression of surrounding structures, formation of fissures, ulcers or hemorrhages, and functional and aesthetic problems. [3]

The most common location is the head and neck, which accounts for 60% of all cases. Eighty percent of hemangiomas occur as single lesions, but 20% of affected patients will have multiple tumors. Oral HEM can be found in the lips, tongue or buccal mucosa. Extra orally they can occur anywhere in the head and neck but more common in the parotid, lip, oral cavity, perinasal region and the larynx. [4] A case of extraoral hemangioma affecting the cheek of a 10 year old girl is presented here with literature review and management.

**Case Report**

A 10 year old female visited the dental clinic with a chief complaint of a red swelling on the left cheek since 6 months. Her medical history was non-contributory. On extraoral examination a well defined sessile cherry red swelling was seen on the left side of the cheek measuring 1x1 cm. [Fig. 1] It was present 3cms lateral to the corner of the mouth and 2cms above the inferior border of the mandible.

On palpation the growth was soft, mobile and non-tender. There was no local rise in temperature. It was partially compressible.

On intraoral examination no extension of the lesion onto the buccal mucosa was evident.No lymph nodes were palpable. A provisional diagnosis of hemangioma was made.

The biochemical parameters of the patient were within the normal ranges. Hemoglobin was 13.1 gm/dl, Hematocrit was 33.4%, and erythrocyte sedimentation rate was 12 mm/h. Her blood glucose levels and urine examination was normal.

A conservative surgical excision was planned based on the size and location of the lesion. Use of a sclerosing agent was not considered owing to the small size of the lesion. After taking aseptic precautions, using a no 15 B.P blade the lesion was excised under local anesthesia. [Fig. 2] The surgical site was closed using a 3-0 silk suture. [Fig. 3]

The specimen was sent for histopathological analysis which showed numerous proliferating blood vessels and budding capillaries engorged with red blood cells and extensive endothelial cell proliferation. [Fig. 4]. A final diagnosis of capillary hemangioma was established.

Patient was recalled after 15 days for follow up where the surgical site had completely healed. [Fig. 5] Patient was called for periodic follow up for a period of six months where no recurrence of the lesion was observed.

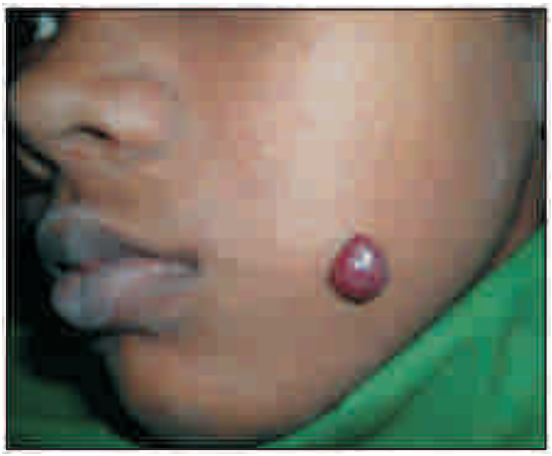

Fig 1: *A well defined cherry red swelling seen on the left side of the cheek.*

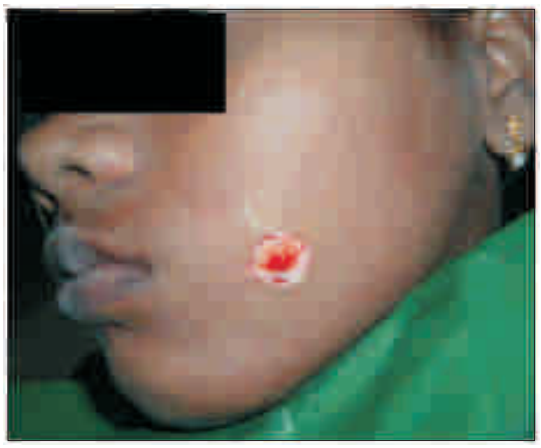

Fig 2: *Surgical procedure, the lesion was excised under local anesthesia*

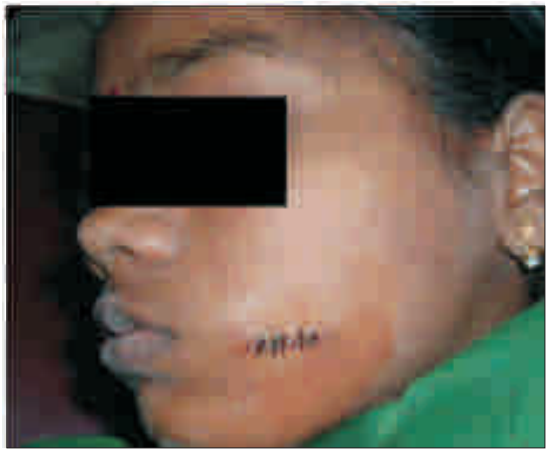

Fig 3: *The surgical site was closed using a 3-0 silk suture.*

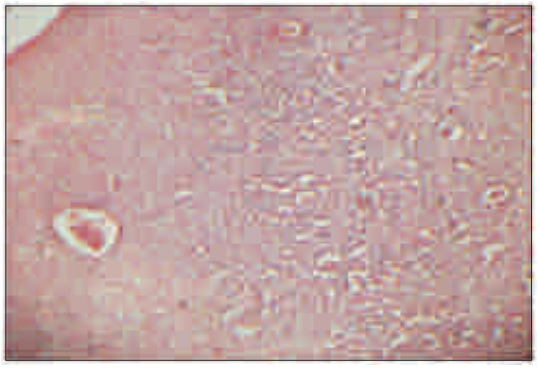

Fig 4: *10 X . H & E stain showing numerous proliferating blood vessels and budding capillaries engorged with red blood cells and extensive endothelial cell proliferation.*

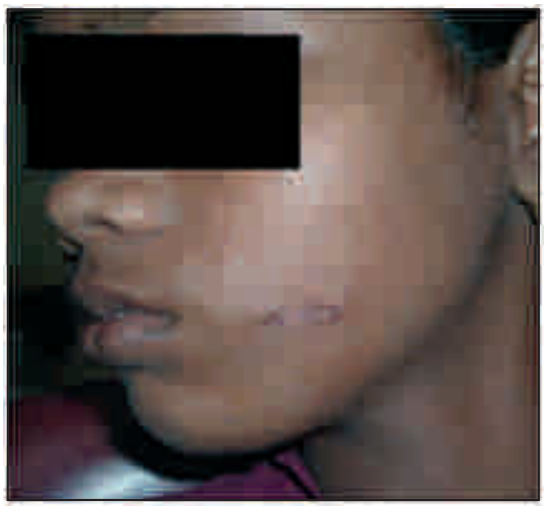

Fig 5: *During follow up where the surgical site had completely healed.*

## Discussion

The term hemangioma has traditionally been used to describe a variety of developmental vascular anomalies. The head and neck regions possess complete rich and intricate blood vessels which might be a predisposing factor for a variety of vascular lesions.

Hemangiomas are one of the most common neoplasm of infancy with an estimated prevalence of 1-3% of all the neonates and 10% by one year of age. 60% of hemangiomas arise in the head and neck. Capillary hemangiomas have a 3:1 female to-male ratio and occur more frequently among Caucasians than other racial groups. [5]

Clinically, hemangiomas of the oral soft tissue often appear as soft, flat or raised mucosal lesions. They are usually deep red or bluish red in color and may blanch on the application of pressure. They are seldom well circumscribed, and are most commonly seen in the lip, tongue and buccal mucosa. [6] Our case was located extraorally on the cheek; it appeared cherry red in color.

Hemangiomas are classified on the basis of their histological appearance as capillary, cavernous or mixed. Capillary hemangiomas are composed of many small capillaries lined with a single layer of endothelial cells supported in connective tissue stroma of varying density. Cavernous hemangiomas are formed by large, thin-walled vessels or sinusoids that are lined with a single layer of endothelium and are separated by thin septa of connective tissue. Mixed hemangiomas consist of both capillary and cavernous components.[7] Our case turned out to be capillary hemangioma by histopathology.

Hemangiomas can be associated with a number of abnormalities. One cluster of abnormalities has been referred to as the PHACE syndrome: posterior fossa abnormalities, facial hemangiomas, arterial abnormalities, cardiovascular defects, and eye abnormalities. [8]

Potential complications include Kasabach-Merritt syndrome (consumptive coagulopathy), compression of vital structures (e.g., airway, orbital structures), fissure formation, ulceration, and bleeding. These complications usually occur in the rapid proliferate phase and can be associated with a mortality rate as high as 20–30%. [9]

Klippel-Trénaunay syndrome is a combined capillary–lymphatic–venous malformation of the trunk or extremities in association with limb overgrowth. Sturge-Weber syndrome is a trigeminal nerve distribution capillary malformation with

intracranial abnormalities. Proteus syndrome includes cutaneous and visceral vascular malformations with pigmented nevi, hemihypertrophy, hand or foot overgrowth, exostoses, and lipomatosis. [9]

Our case was non syndromic hemangioma.

Angiography has proved to be useful as a diagnostic tool when clinical and radiographic characteristics suggest a diagnosis of hemangioma. This will demonstrate presence of a vascular lesion and delineate its boundaries.

Various treatment options available depend on size of lesion, location of lesion and age of patient. The range of treatment includes steroid therapy, sclerosing agents, irradiation, and surgical excision with or without ligation of vessels, embolization, laser therapy and replacement of resected area with iliac bone graft. Treatment is indicated only in some conditions like aesthetic disfigurement as uncontrolled bleeding is the most hazardous complication. Sclerotherapy has an indispensible role when surgery is not recommended. Sclerotherapy may cause complete regression of low vascular lesions.

Radiotherapy is useful to reduce the tumoral volume. Nevertheless, it has a lot of adverse effects such as damage to the normal adjacent tissues growth, residual scarring and malignization. So, radiotherapy is considered unacceptable therapeutic option. Simple curettage may lead to an uncontrollable bleeding as well as an incomplete excision of the lesion. [10] Surgical excision was the treatment of choice in our case.

The flash pumped pulsed dye laser (FPDL) is one of the treatments of choice for the superficial hemangiomas. FPDL emits short pulses of concentrated energy in the wavelength of oxyhemoglobin and induces thermolysis of vasculature. But their effects on hemangiomas are minimal especially if they are deep and its excessive or wrong use can cause thermal burns. The early detection and biopsy of capillary hemangiomas is necessary in order to identify the clinical nature of the tumor and avoid potential complications.

## Conclusion

At birth, the hemangiomas are often small and inconspicuous, with 60% absent at birth. Hemangiomas can have deep, superficial, or mixed components. The clinical appearance of hemangiomas varies with the degree of dermal involvement and the depth of the lesions. A characteristic strawberry appearance is present when the lesions involve the skin. Hemangiomas and vascular malformations are endothelial lesions that can present with a number of serious medical problems, hence rapid diagnosis and prompt management is essential.

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## References

1. *Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg. 1982; 69:412-22.*

2. *Enjolras O, Mulliken JB. Vascular tumors and vascular malformations. Adv Dermatol. 1997; 13:375-423.*

3. *Donnelly LF, Adams DM, Bisset GS 3rd. Vascular malformations and hemangiomas: a practical approach in a multidisciplinary clinic. AJR Am J Roentgenol. 2000; 174: 597-608.*

4. *Jackson IT, Carreno R, Potparic Z, Hussain K. Hemangiomas, vascular malformations, and lymphovenous malformations: classification and methods of treatment. Plast Reconstr Surg. 1993; 9:1216-30.*

5. *Braun IF, Levy S, Hoffman JC Jr. The use of transarterial microembolization in the management of hemangiomas of the perioral region. J Oral Maxillofac Surg 1985; 43: 239-48.*

6. *Shafer WG, Hine MK, Levy BM. A Textbook of Oral Pathology. 4th ed. WB Saunders Co. Philadelphia, p: 154-257, 1983.*

7. *Regezi JA, Sciubba JJ. Oral Pathology. 3rd ed. WB Saunders Co. Philadelphia, p:136 -8, 1993.*

8. *Frieden IJ, Reese V, Cohen D. PHACE syndrome: the association of posterior fossa Brain malformations, hemangiomas, arterial anomalies, coarctation of the aorta and Cardiac defects and eye abnormalities. Arch Dermatol* 1996*; 132:307–11.*

9. *Fishman SJ, Mulliken JB. Hemangiomas and vascular malformations of infancy and Childhood. Pediatr Clin North Am* 1993*; 40:1177–1200*

10. *Kenan S, Abdelwahab IF, Klein MJ, Lewis MM. Hemangiomas of the long tubular Bone. Clin Orthop Rel Res 1992; 256-60.*

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