

Case Report

Compound haemangioma of lower lip: an interesting and rare case report

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ABSTRACT

Haemangiomas are common benign vascular tumours of infancy and childhood. Haemangiomas occur in 10-12% of children of 1 year of age and most of them are self-resolving within 9 years of age. Most common sites are head and neck (around 90%) and lip is one of the preferred sites. Sometimes persistence of haemangiomas may require surgical intervention. Capillary haemangiomas generally located superficially and cavernous haemangiomas located deep. Mixed capillary-cavernous haemangiomas or compound haemangiomas are one of the rarer types and location at lip rarest. Here we report a 14-years-old boy presented to paediatric surgery outdoor with swelling in lower lip for last 3 years. The excisional biopsy done with a clinical diagnosis of granuloma and sent for histopathological study. On microscopy of tissue sections given from lesion showed stratified squamous epithelium with sub epithelium revealing two distinct areas of capillary haemangioma component and cavernous haemangioma component within the lesion. So, final diagnosis of capillary-cavernous haemangioma (compound haemangioma) was made without any granuloma or malignant component. Patient followed up for six months and he was completely asymptomatic without any residual disease and recurrence.

Keywords: Cavernous, Capillary, Lip, Child

INTRODUCTION

Haemangiomas are the most common benign vascular tumor of infancy and childhood. In around 90% of cases, it usually resolves by 9 years of age. Majority of the haemangiomas occur in head and neck region (60%) with lip, tongue, and palate being the most preferred sites.¹ They are also seen in trunk (25%) followed by extremities (15%). Its incidence is found more in girls, whites, premature infant and twins.²

Clinically, it presents as a smooth or lobulated mass, ranging in size from a few millimetres to a few centimetres. Vascular anomalies can be correctly labeled either as a tumor or malformation without the need of further tests 90% of the time, based on appearance and

typical growth characteristics.³ Treatment of persistence lesions may require surgery but depending upon size and location.^{4,5} Still there is no standardized treatment available.⁶

Lip as one of the preferred sites, the haemangiomas of lip can be superficial, deep or mixed type. Superficial is capillary haemangioma, deep is cavernous haemangioma or mixed as capillary-cavernous or compound haemangioma.^{7,8} Only few cases are available in literature as compound haemangioma of lower lip.

CASE REPORT

A 14-years-old boy who presented in paediatric surgery OPD with a swelling in left side of lower lip for last three

years. The swelling was gradually increased in size not associated with pain and fever. There was no previous significant medical history. No important family history was there. Systemic examinations were within normal limits on local examination the size of swelling was 1.5×1.0 cm, soft and pink without any signs of inflammation. Clinical diagnosis of granuloma was made and planned for excisional biopsy. His routine haematological and biochemical investigations were within normal limits. The lesion was excised and sent for histopathological examination. Grossly there was greyish white to greyish black irregular tissue bit measuring 1×0.8×0.5 cm. Tissue bisected and all embedded in one block. Hematoxylin and eosine (H and E) stained section on microscopic examination showed normal stratified squamous epithelium and sub epithelium showing a lesion (Figure 1).

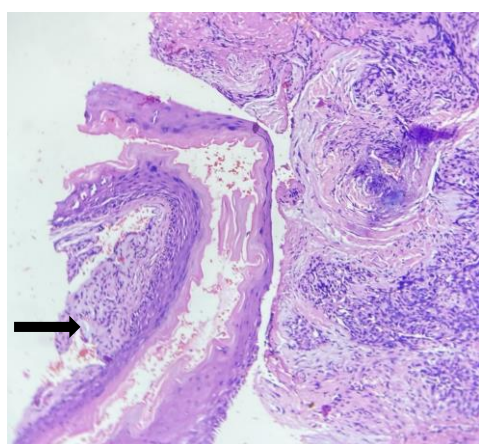


Figure 1: Photomicrograph showing stratified squamous epithelium (arrow) along with a vascular lesion below epithelium (H and E, at 100X).

The lesion revealed a vascular tumour with areas showing lobular arrangement and proliferation of small capillaries intermingled with large anastomosing thin walled dilated vessels with larger caliber having ill-defined circumscription (Figure 2-4).

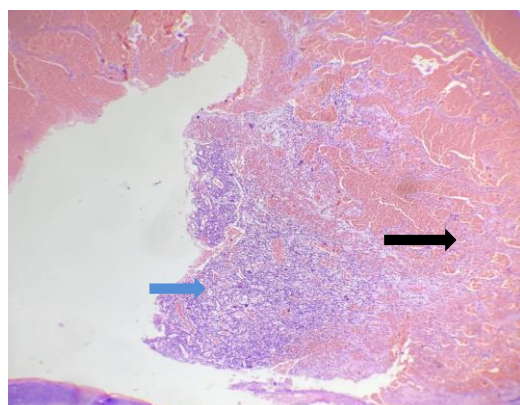


Figure 2: Photomicrograph showing areas of both capillary hemangioma (blue arrow) and cavernous hemangioma (black arrow) (H and E, at 40X).

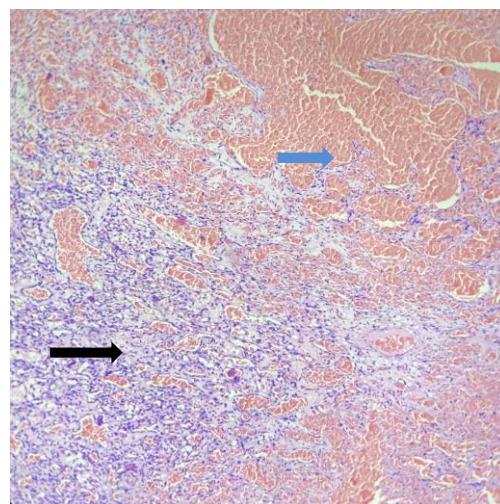


Figure 3: Photomicrograph showing areas of cavernous hemangioma with dilated blood vessels engorged with red blood cells (blue arrow) and capillary hemangioma with proliferating capillaries (black arrow) (H and E, at 100X).

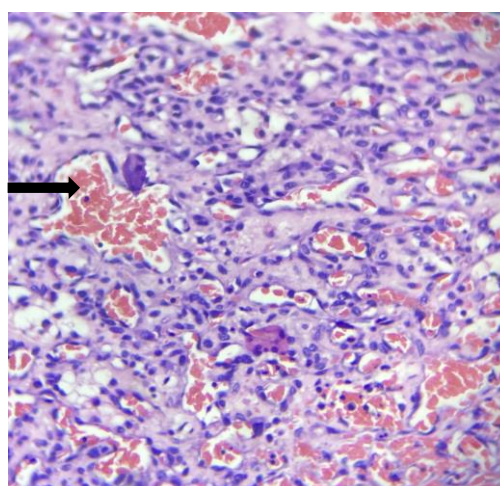


Figure 4: Photomicrograph showing thin walled capillaries lined by a single layer of flat endothelial cells without pleomorphism or increased mitosis (black arrow) (H and E, at 400X).

There was no pleomorphism or increased in mitosis noted in the endothelial cells. So, final histopathological diagnosis of capillary-cavernous haemangioma/compound haemangioma without evidence of malignancy or granuloma was made. Post excisional status were uneventful and patient was followed for six months without any residual disease or recurrence.

DISCUSSION

Haemangiomas and vascular malformations are two different and distinct groups of vascular lesions, which are often confused with each other. These terms have been used wrongly and interchangeably. Vascular lesions are classified on the basis of their anatomical, structural

features and biological behavior.¹ The classification developed by Mulliken and Glowacki in 1982 was based on the cellular kinetics of anomalous vessels. In this classification, two entities exist: haemangiomas and vascular malformations.⁹ The International Society for the Study of Vascular Anomalies (ISSVA), in 1996, approved a classification based on the fundamental separation of vascular anomalies into those lesions with a proliferative component (named 'vascular tumors') versus relatively static 'vascular malformations', following Mulliken and Glowacki's essential work. Wassef et al updated official ISSVA classification of vascular anomalies in 2015.¹⁰ The General biological scheme of the classification is retained. The section on tumors has been expanded and lists the main recognized vascular tumors, classified as Benign, locally aggressive or borderline, and malignant.⁹

Vascular neoplasms have increased endothelial cell turnover and they undergo mitosis. In contrast, malformations are not true neoplasms and therefore do not have increased endothelial cell turnover and do not exhibit mitosis. Instead, vascular malformations are defined as structural abnormalities of capillary, venous, arterial and lymphatic system.³ Vascular tumors arise from angioblasts that give rise to primitive blood vessels by a process known as vasculogenesis. Various proliferative cellular markers have been identified such as vascular endothelial growth factor (VEGF).¹¹ Imaging is useful in questionable lesions. Investigations such as MRI, CT scan, ultrasound with Doppler can be used. Recent studies show various immunohistochemical markers expressed by vascular tumors and malformations out of which GLUT 1, merosin, Lewis Y antigen, CD34, and Fcy-RIIb are the most important markers.³ Management and treatment of vascular lesions is mainly guided by various factors such as location of the lesion, flow characteristics, symptoms, functional disability and cosmetic deformity caused by the lesion.⁹

Haemangiomas are benign vascular tumours. Capillary haemangiomas are composed of many small capillaries lined with a single layer of endothelial cells supported in connective tissue stroma of varying density. Cavernous haemangiomas 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 haemangiomas consist of both capillary and cavernous components.¹ Although lip is a preferred site of haemangioma, mostly capillary haemangiomas seen in superficial part but in our case, there was clear distinct morphology of both capillary and cavernous type present within same lesion.

Although size of the lesion was not so big getting such type of compound morphology was surprising. Haemangiomas recur that's why patient should be followed up as in our case we followed the little boy for six months with a happy outcome.

CONCLUSION

Compound haemangiomas of lower lip is an extremely rare pathological entity. In each case careful histopathological examination needed and associated malignancy or other differentials like granuloma should be ruled out.

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