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# ISOLATED CAVERNOUS HEMANGIOMA OF EYELID- A RARE CASE

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## ABSTRACT

**Purpose:** We describe clinical and pathologic picture of a young patient with isolated cavernous hemangioma of eyelid managed successfully. **Method:** A 24 years female presented to our OPD with firm globular, multi-lobulated & non-reducible mass over her left upper eyelid having indolent course With total duration of eight years of presentation, she was having defective vision for last four years due to presence of 5 by 5 cm dimension mass near medial canthus extending to lateral canthus and occupying space below eyebrow to lid margin of left eyelid. As an adaptation for better vision, she used to keep her head erect and chin elevated. MRI concluded with a T2 hyperintense lesion with patchy contrast enhancement. **Result:** Excision under local anesthesia followed by biopsy which confirmed our diagnosis as vascular hamartomatous lesion. Patient is on follow-up for last one year with last 1 year with acceptable cosmesis and preserved normal vision. **Conclusion:** Cavernous hemangioma is the most common benign non-infiltrative neoplasm of the orbit having an intraconal and lateral location. Similar pathology of eyelids are rare to be searched in English literature but should be considered as a differential in case of isolated eyelid lesions.

**KEYWORDS:** benign tumor, lid mass, adnexal lesions, vascular malformation, capillary hemangioma.

## INTRODUCTION

Cavernous hemangioma is the most common benign non-infiltrative neoplasm of the orbit. It represents a hamartoma that seldom appears prior to teen age, majority arising after second

decade. Mostly, orbital cavernous hemangioma lesions are intraconal and lateral in location.[1] To our best knowledge there is no reports on isolated cavernous hemangioma of eyelid. Cavernous hemangioma primarily involving the eyelid without orbital extension is very rare entity to be found in standard text books. We report a case of unilateral cavernous hemangioma of eyelid in a young female managed surgically & doing good on follow-up.

## CASE DETAILS

Young female aged 24 years presented to Eye OPD with a slow growing mass over her left upper eyelid. She was having this mass for last eight years. Initially, it was not associated with any other ocular complaints. For last four years, she developed defective vision as the mass was large enough to obscure her visual field. She had tried many alternative medicines for the same with no results. History of prior trauma or surgical attempt for removal or biopsy on eyelid was not present. Her general condition was satisfactory. On systemic examination, no significant abnormality was detected. On inspecting her general stature and position of head with respect to body axis, head was erect & chin elevated. Her eyebrows were elevated and frontal creases were prominent. On local inspection, Solitary mass of 5 × 5cm approx. dimensions with globular shape, lobulated surface was present over left upper eyelid. Extension of the lesion was from lateral canthus to 1 centimeter short of medial canthus. Mass was overhanging upper eyelid with preserved eyelashes and eyebrow. [fig. 1] Skin overlying the mass was pigmented having purplish hue. On palpation, it was firm, well-circumscribed, not fixed to overlying skin, comparatively mobile. Finger insinuation was possible below the supra-orbital rim. The mass was non-reducible & non-pulsatile. On auscultation, no bruit was heard. The mass didn't increase in dimension with Valsalva maneuver. Mass lesion was huge enough to cover the entire pupil. Her visual acuity was 6/6 in both eyes, all the extra ocular movements were full in all gazes. Intraocular pressure, anterior segment and posterior segment of both the eyes were normal. The differentials considered at this point were cavernous hemangioma, dermoid, neurofibroma or lymphangioma. On **Magnetic resonance imaging** (**MRI**) with contrast, hyperintense T2 weighted lesion of size 3 × 2.7 mm with peripheral patchy enhancement was noted in the left upper eyelid.

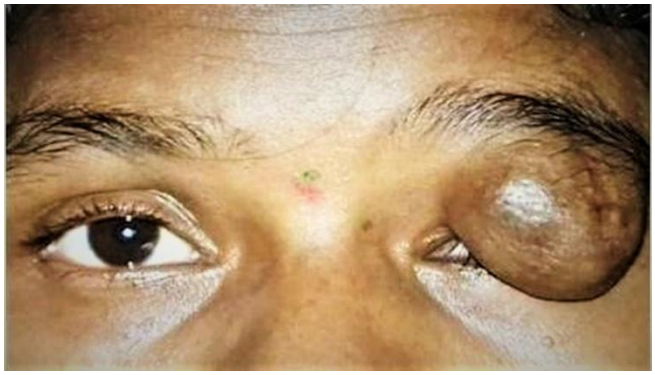

**Fig. 1 - Preoperative finding of left upper eyelid mass.**

Excisional biopsy was planned under local anaesthesia. The lesion was excised via transcutaneous approach with an incision over the mass. A multi-lobulated well encapsulated mass of purple-pink color was found. It was sparsely adhered to surrounding tissue and could be nicely delivered to the wound. [Fig.2] No significant bleeding was present. Minimal oozing from surgical field was managed. No feeding vessel was identified. Surgical incision was closed in two layers after excision of redundant skin.[Fig. 3]

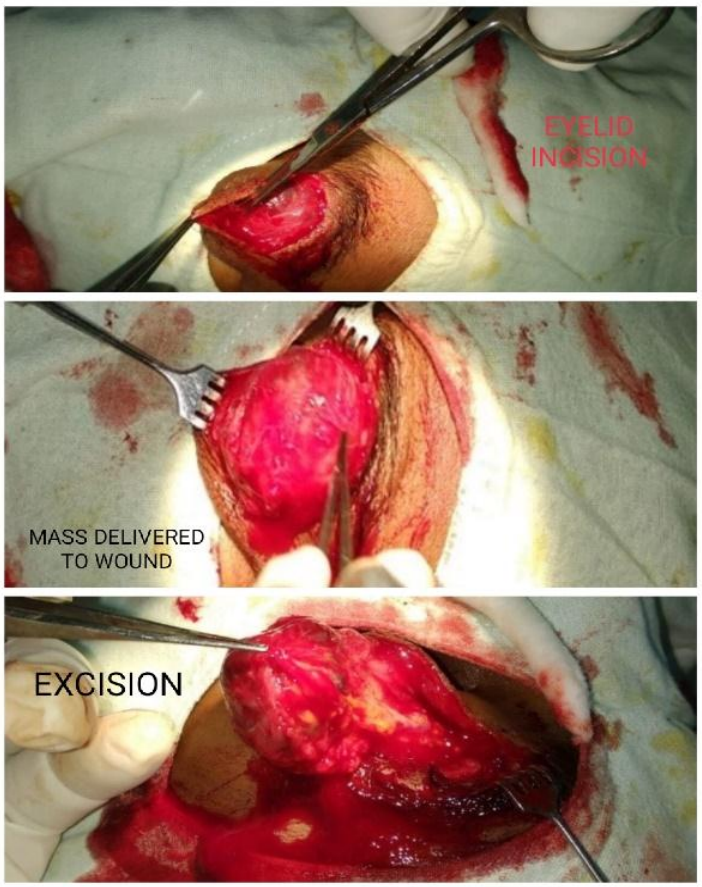


**Fig. 2: eyelid incision and release of flimsy strands from eyelide tissue with excision of cavernous hemangioma.**

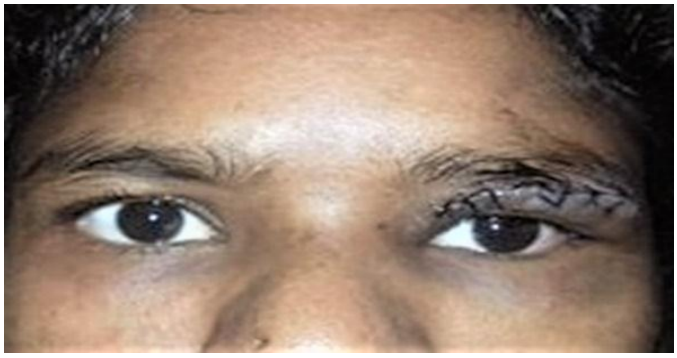

**Fig. 3: follow-up for suture removal after excisional biopsy.**

Histopathology revealed dilated vascular endothelium lined blood filled spaces suggestive of vascular hamartomatous lesion likely to be cavernous hemangioma. [Fig. 4] Postoperative course was uneventful and patient discharged on next day with dressing changed. Next visit was advised for suture removal at 1week of postoperative period. On removing sutures, wound seemed healthy with well apposed margin and resolving mild edema of left upper eyelid. Lid margin was normal with no ectropion or entropion. After one month, she visited for routine follow-up with satisfactory cosmetic result and normal vision. We found a curvilinear scar mark over the left upper eyelid only.

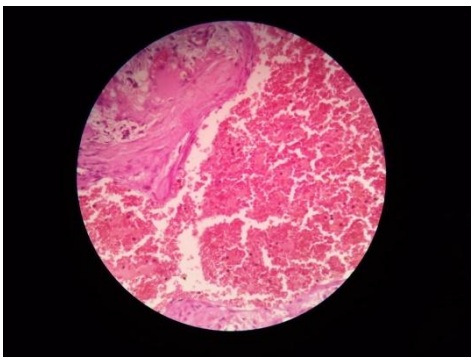

**Fig. 4: histopathology of excisional biopsy specimen.**

## DISCUSSION

Cavernous hemangioma is the most common benign orbital tumor of adults and only rarely occurs as a primary eyelid lesion. Even literatures showing eyelid involvement or eyelid presentation of orbital cavernous hemangioma are few. Out of 214 diagnosed orbital cavernous hemangiomas, only 11.7% of lesions presented with eyelid swelling.[2] Involved subcutaneous tissue of eyelid in a case of orbital cavernous hemangioma was published only recently in 2020 as a case report.[3] Our case involved the left upper eyelid without involving orbit & other adnexa. In contrast, a case report published in 2003 reported multifocal

cavernous hemangioma involving cheek, palate, temporal fossa, orbit, eyelid, and conjunctiva.[4]

As in orbital lesions having more incidence in females, this case is also a young female increasing slowly Cavernous hemangiomas usually present during adulthood only rare to undergo spontaneous regression. Whereas, capillary hemangiomas are the most common orbital tumor of vascular origin found in infants. They start regressing spontaneously after an initial phase of progression.[5]

Superficial skin lesions are dark blue, compressible and, unlike the orbital variety, not encapsulated.[3,6] In this case the mass was well circumscribed, lobulated with overlying hyper pigmented skin. The typical dark blue hue is more evident in white skinned people but in dark skinned people the color may vary. Treatment is surgical excision.

Clinical differentials for this diagnosis were other commonly found pathologies like dermoid, neurofibroma and lymphangioma. Considering regular surface and margin with absence in direction of bony fusion, dermoid could be ruled out.[7] Additionally, imaging might show bony adhesion or scalloping. Absence of café au lait spots, Lisch nodules or cutaneous multiple fibromas lead to exclusion of neurofibroma. Neurofibroma lesions had no skin color alteration as seen in this case.[8] Lymphangioma commonly presents in pediatric population and characteristically ill-circumscribed with infiltrative growth pattern. It also involve orbit and eyelids resulting in gross cosmetic facial deformity.[9]

Computerized tomography (CT) with contrast or MRI have been considered primary diagnostic methods for orbital lesion. But arteriography or venography for orbital cavernous hemangioma have limited role.[10] Cavernous hemangioma on CT scan are homogenously enhanced, well circumscribed mass lesions. We preferred MRI which showed a T2 hyperintense lesion with patchy peripheral enhancement in the left upper eyelid without orbital extension or bony invasion. Similar characteristic lesions were also imaged in cases of intraconal orbital cavernous hemangioma.[11] Confirming the diagnosis could also be done by technetium Tc 99m (99mTc) red blood cell scintigraphy which is done in few centers only.[12]

This case was further evaluated for liver or bowel involvement by sonogram of abdomen which turned out to be normal. Cavernous hemangioma may be associated with a rare syndrome termed as ‘Blue rubber bleb nevus’ syndrome. This syndrome is characterized by

varied vascular malformations like cavernous hemangioma, capillary hemangioma, vascular angioma and telangiectasia of multiple abdominal organs including eye.[13] Excisional biopsy specimen of this case was suggestive of vascular hamartomatous lesion consisting of dilated, endothelium-lined vascular spaces with thrombosis. A subtype of cavernous hemangioma termed as sinusoidal hemangioma has been described to involve the eye lid with a more aggressive growth pattern invading adjacent areas of the brow and cheek.[14]

Surgical treatment was warranted as the patient was a young student with difficulty of vision. We did a complete surgical excision of the mass under local anesthesia and the wound was closed with multiple interrupted skin suture with 5-0 silk. Cosmetic result was also essential in this case, so eyelid tissue were to be preserved. We had planned for reconstruction of eyelid if required. Many medical management options had been proven to be successful in capillary hemangioma but not for cavernous hemangioma.[15]

## CONCLUSION

Isolated cavernous hemangioma of eyelid is a rare tumor. Other differential diagnosis should be accurately ruled out by doing a proper examination of the mass, by doing relevant investigations like CT scan, MRI and Histopathology. A well circumscribed cavernous hemangioma of eyelid can easily be excised and all excision should be followed by excisional biopsy to rule out any malignant component.

## REFERENCES

1. Louisraj S, Ponnudurai T, Rodriguez D, Thomas PA, Nelson Jesudasan CA. Cavernous hemangioma of the orbit: an unusual acute presentation. Int Med Case Rep J, 2017 Jul 24; 10: 255-259. doi: 10.2147/IMCRJ.S133284. PMID: 28769595; PMCID: PMC5533564.
2. Yan J, Wu Z. Cavernous hemangioma of the orbit: analysis of 214 cases. Orbit, 2004 Mar; 23(1): 33-40. doi: 10.1076/orbi.23.1.33.28992. PMID: 15513018.
3. Bagheri A, Feizi M, Tavakoli M. Unusual Subcutaneous Presentation of Cavernous Hemangioma in the Lower Eyelid. J Ophthalmic Vis Res, 2020 Apr 6; 15(2): 236-239. doi: 10.18502/jovr.v15i2.6741. PMID: 32308958; PMCID: PMC7151512.
4. Bajaj MS, Nainiwal SK, Pushker N, Balasubramanya R. Multifocal cavernous hemangioma: a rare presentation. Orbit, 2003 Sep; 22(3): 155-9. doi: 10.1076/orbi.22.3.155.15613. PMID: 12868022.

5. Smoker WR, Gentry LR, Yee NK, Reede DL, Nerad JA. Vascular lesions of the orbit: more than meets the eye. Radiographics, 2008 Jan-Feb; 28(1): 185-204; quiz 325. doi: 10.1148/rg.281075040. PMID: 18203938.
6. Rootman DB, Rootman J, White VA. Comparative histology of orbital, hepatic and subcutaneous cavernous venous malformations. Br J Ophthalmol, 2015 Jan; 99(1): 138-40. doi: 10.1136/bjophthalmol-2014-305761. Epub 2014 Oct 15. PMID: 25320153.
7. Sezenoz AS, Arat YO, Tepeoglu M. An unusual eyelid mass: Tarsal dermoid cyst. Saudi Journal of Ophthalmology: Official Journal of the Saudi Ophthalmological Society, 2015 Oct-Dec; 29(4): 312-314. DOI: 10.1016/j.sjopt.2015.05.004.
8. Pe'er J. Pathology of eyelid tumors. Indian J Ophthalmol, 2016 Mar; 64(3): 177-90. doi: 10.4103/0301-4738.181752. PMID: 27146927; PMCID: PMC4869455.
9. Pang P, Jakobiec FA, Iwamoto T, Hornblass A. Small lymphangiomas of the eyelids. Ophthalmology, 1984 Oct; 91(10): 1278-84. doi: 10.1016/s0161-6420(84)34158-6. PMID: 6514291.
10. Ansari SA, Mafee MF. Orbital cavernous hemangioma: role of imaging. Neuroimaging Clin N Am, 2005 Feb; 15(1): 137-58. doi: 10.1016/j.nic.2005.02.009. PMID: 15927865.
11. Purohit BS, Vargas MI, Ailianou A, Merlini L, Poletti PA, Platon A, Delattre BM, Rager O, Burkhardt K, Becker M. Orbital tumours and tumour-like lesions: exploring the armamentarium of multiparametric imaging. Insights Imaging, 2016 Feb; 7(1): 43-68. doi: 10.1007/s13244-015-0443-8. Epub 2015 Oct 31. PMID: 26518678; PMCID: PMC4729705.
12. Polito E, Burroni L, Pichierri P, Loffredo A, Vattimo AG. Technetium tc 99m-labeled red blood cells in the preoperative diagnosis of cavernous hemangioma and other vascular orbital tumors. Arch Ophthalmol, 2005 Dec; 123(12): 1678-83. doi: 10.1001/archopht.123.12.1678. PMID: 16344439.
13. Raidas RC, Parihar D, Singhi MK, Kachhawa D. Blue rubber bleb nevus syndrome: A case report and review of literature. Indian J Paediatr Dermatol, 2015; 16: 210-3.
14. Enjolras O, Wassef M, Brocheriou-Spelle I, Josset P, Tran Ba Huy P, Merland JJ. Hémangiome sinusoïdal [Sinusoidal hemangioma]. Ann Dermatol Venereol, 1998 Sep; 125(9): 575-80. French. PMID: 9805543.
15. Bang GM, Setabutr P. Periocular capillary hemangiomas: indications and options for treatment. Middle East Afr J Ophthalmol, 2010 Apr; 17(2): 121-8. doi: 10.4103/0974-9233.63071. PMID: 20616917; PMCID: PMC2892126.