Surgical Outcome of Atypical Periocular Capillary Hemangiomas -
A Case Series

Abstract
Capillary hemangioma also called as “benign hemangio-endothelioma” and “strawberry nevus” is a
hamartomatous, localized proliferation of vascular tissue in a normal location. In children, periocular capillary
hemangiomas is one of the most common benign eyelid and orbital tumors. It affects nearly 2% of all infants,
has female predilection, manifests by 1-8 months of age and commonly regresses spontaneously by age of 7
years. Non resolving hemangiomas can be treated with steroids, laser or surgical excision. However there
may be recurrence after incomplete surgical excision. In this article the clinical profiles and the various
surgical managements of three atypical periocular capillary hemangiomas in respect to age and sex are
discussed.

Keywords: Capillary hemangioma, Eyelid tumor, Periocular tumor

Introduction
Capillary hemangioma (CH) is a most common benign periocular tumor comprised of hamartomatous localized
proliferation of vascular tissue in a normal location. It affects up to 2% of all infants with female predilection
3:2. It is a benign vascular tumor composed of proliferating endothelial cells within fibrous tissue. It may present in 1
-2% of newborns as a cutaneous, subcutaneous or deep lesion. It usually presents at birth or may become evident
in 10%-12% of children by the end of first year. Usually it is diagnosed clinically, but orbital ultrasound imaging is
helpful in subcutaneous and deep lesions to ascertain the extent of the involvement. Most hemangiomas regress
spontaneously by age of seven years.[1] The choice of treatment depends on lesion location, its size and potential
adverse effects of the treatment itself. Steroids, propranolol and surgical excision are some treatment options.[2] Here we present the various atypical presentations of periocular capillary hemangiomas in
respect to age and sex and also present their good surgical outcome in rural set up.

Case presentations
Case 1
A 7 year old male child, presented with painless, progressive swelling of left upper eyelid margin of 2
months duration. Ophthalmic examination revealed normal anterior and posterior segments and 6/6 visual acuity (VA)
in both eyes. Local examination of left upper eyelid showed 3x4 mm protruding, yellowish white colored lesion
with soft consistency. Total excision biopsy under general anesthesia was carried out. Histopathological examination
confirmed the diagnosis of capillary hemangioma. Patient was discharged with flat lid margin surface. One month
later painful left upper eyelid swelling reappeared with mild drooping. The swelling was 10mm in diameter,
circular, dome shaped, red colored with discrete yellow areas. It was soft and tender (Fig.1). Recurrence of
capillary hemangioma with infection was the clinical diagnosis. Total meticulous excision of the mass with good
hemostasis using cautery knife was carried out. Biopsy of the excised mass confirmed the recurrence of capillary
hemangioma with infection (Fig. 2). Postoperatively, local steroid-antibiotic ointment was administered. There was
no recurrence after 3 years (Fig. 3).
Histopathological examination showed small to medium sized capillaries and fibrous tissue in the intervening stroma and confirmed the diagnosis of capillary hemangioma. Antibiotic drops and ointment with oral anti-inflammatory and analgesics were prescribed postoperatively. Sutures were removed on 7th postoperative day. Followup examination after two years showed no recurrence in the asymptomatic patient (Fig. 8).

**Case 2**

A 11 year old boy presented with painless, gradually progressive red mass at medial corner of right eye since 2 months (Fig 4). Local examination showed normal 6/6 visual acuity in both eyes. Left Eye showed normal anterior and posterior segment. Right upper eyelid showed a reddish colour, lobulated, movable, nontender mass of 1 cm diameter with firm consistency, near the medial canthus (Fig 5). There was no crusting, ulceration or vascularisation over the mass. Anterior and posterior segment examination of right eye was normal. Clinical diagnosis of hemangioma was made and total meticulous excision biopsy of the mass (Fig. 6) and ligation of feeder vessel (Fig. 7) was carried out under general anaesthesia and sent for histopathological examination. The skin incision was sutured with 3 interrupted 7-0 nylon sutures.
Case 3

A 9 year old boy presented with painless, gradually progressive red mass at medial corner of right eye since 2 months (Fig 9). General and systemic examination of the patient was normal. Local examination showed 6/6 visual acuity in both eyes. Left eye findings were within normal limits. At the medial canthus the right eyelid showed a pinkish colour, nonlobulated, movable and tender mass of 1 cm diameter with firm consistency (Fig 10). Rest of the anterior and posterior segment examination of both eyes was normal. Clinical diagnosis of hemangioma was made and total excision biopsy (Fig. 11) with good hemostasis using cautery knife was carried out under general anaesthesia. Skin sutured with 3 interrupted 7-0 nylon sutures. Histopathological examination confirmed the diagnosis of capillary hemangioma. Postoperative antibiotic drops and ointment with oral anti-inflammatory and analgesics were prescribed and patient was discharged. Sutures were removed on 7th postoperative day. Follow-up examination after 1 year revealed no recurrence in asymptomatic patient (Fig. 12).

Discussion

Capillary hemangioma also called as “benign hemangioendothelioma” and “strawberry nevus” is a hamartomatous, localized proliferation of vascular tissue in a normal location.

Capillary hemangioma is the most common benign orbital tumor in children which affects 2 % of all infants, with a female predilection.[3:2] Capillary hemangioma is a tumour of primitive cells with capability to differentiate towards endothelial cells and pericytes and thus composed of proliferating endothelial cells within fibrous tissue. It shows two phases of growth, a proliferative phase and an involutional phase.[3] In proliferative phase, rapid growth occurs from 8-18 months and in involutional phase, there is slow regression of hemangioma. In proliferative phase there is increase in number of endothelial and mast cells which stimulates the vessel growth. Endothelial cell proliferation returns to normal following the proliferation phase. In involutional phase mast cell numbers decrease to normal and there is a decreased endothelial and mast cell activity. These vascular spaces become lined with endothelial cells without muscular support.

Approximately one-third of capillary hemangiomas are apparent at birth, while the remaining two-thirds or more
manifest by 6 months of age and may enlarge quickly over the first year, after which the tumor begins to involute. Half the lesions will involute by age 5 years, and 75% will involute by age 7 years. Involution typically begins centrally and then spreads to periphery. Most of the involution occurs by age of 4 to 7 years. Capillary hemangiomas are more frequent in premature or low-birth-weight infants. The history and presentation of a periorbital capillary hemangioma is typical and diagnostic. It may be cutaneous, subcutaneous or deep lesion involving orbit. The subcutaneous portion may extend into the orbit causing painless, non-pulsating proptosis of the involved eye.

Superficial hemangiomas produce an elevated strawberry-colored nodule and are likely to develop in the superonasal quadrant of the orbit and on the upper eyelid. Usually parents describe a growing reddish or purple spot which is absent at birth but later becomes larger and thicker over the first few months of life. The lesion blanches with pressure, which is helpful in distinguishing it from the non-blanching 'port-wine' stain of Sturge-Weber syndrome. It is non-pulsatile due to its low rate of blood flow. Many times during crying there is enlargement and deepened color of the hemangioma due to Valsalva maneuver however it is not pathognomonic for capillary hemangiomas. The peak size of the lesion is reached by 6-12 months. Spontaneous involution is the rule for untreated capillary hemangiomas and progressive involution continues until 7 years of age. Hemangiomas often follow the distribution of the first and second divisions of the 5th nerve. They are usually unilateral and located on the eyelid or brow and may result in significant ptosis of an involved eyelid. Reduced visual acuity may occur due to ptosis or astigmatism or glaucoma.[4,5] Amblyopia is seen in approximately 50% of patients with eyelid hemangioma. Orbital imaging with B-scan ultrasonography is helpful in determining the posterior extent of suspicious lesions, while chest or abdominal imaging may uncover other systemic hemangiomas.[6]

Advised Treatment Options [7]
1. Use of Corticosteroids [8]
2. Systemic propranolol [9]
3. Immunomodulator [10]
4. Surgical excision: Surgical removal is reserved for failed conservative therapy, unsightly residual lesions even after involutional phase and tumor causing vision loss. Thus surgical removal is useful for visual, cosmetic and diagnostic purpose and well-circumscribed superficial small lesions, in the early phases can be excised surgically with good results. However surgery carries the risks of general anesthesia, intraoperative bleeding and recurrence.[11,12]

In the present case series, all patients had relatively short 2 months history and late presentation at 7, 9 and 11 yrs of age. All patients were males. This is not classic presentation for capillary hemangioma in respect to age and sex. In our case no. 1 though there was recurrence after surgical excision which is a known phenomenon, we could prevent it by meticulous total excision of the lesion by using cautery knife and the vessel ligation during the surgical management of all cases. There was no intraoperative profuse bleeding or postoperative complication and recurrence in any case.

Conclusion
Eyelid capillary hemangiomas may present even in late childhood. Meticulous surgical excision with cautery knife and/or ligation of feeder vessel gives good cosmetic result without recurrence.

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References
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