Congenital capillary hemangioma arising from palpebral conjunctiva of a neonate

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Capillary hemangioma is the most common benign vascular eyelid tumor in childhood. The periocular lesion appears within the first few weeks after birth and usually has superficial or deep components. Primary conjunctival capillary hemangiomas are rarely reported. We present the case of a 2-day-old child with a pedunculated capillary hemangioma arising from superior palpebral conjunctiva. A complete surgical excision was performed under general anesthesia, and the child was asymptomatic at follow-up of two months.

Key words: Congenital capillary hemangioma, conjunctival capillary hemangioma, management, pedunculated capillary hemangioma

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Capillary hemangioma is the most common benign vascular eyelid tumor in childhood.[1] It is present in 1%–4% of all births[2] and is more common in premature infants and often following chorionic villus sampling.[3] It is usually a cutaneous, subcutaneous, or deep orbital lesion and commonly presents a few weeks after birth. The usual clinical course of infantile cutaneous or subcutaneous hemangiomas includes an initial engorgement (age 6–12 months) followed by regression

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(age 1–7 years). It usually regresses spontaneously and hence is kept under close observation. Active intervention is performed only if the lesion is very extensive and causes amblyopia, mechanical ptosis, exposure keratopathy, or optic neuropathy. Treatment modalities for cutaneous or subcutaneous hemangiomas include topical[4] and oral beta blockers,[5-8] local and oral steroids,[9,10] surgical excision, immunotherapy,[11] laser photocoagulation with pulsed dye laser,[3] and embolization.[12] Although conjunctiva may be involved as a part of capillary hemangioma of the lid, isolated conjunctival lesion is rare and its congenital presence is rarer. In a series of 1463 cases of conjunctival tumors studied, only 10 were capillary hemangiomas, forming 16% of conjunctival vascular tumors, only 1% of all conjunctival tumors.[13] Usually, hemangiomas of conjunctiva are small and require only observation. Chang and Estes, in a recent paper, report usage of topical timolol in the management of small bulbar conjunctival hemangioma.[14] Oral propranolol was also recently reported to be in use in the treatment of a case of bilateral sessile infantile hemangioma of posterior lamella of upper lid by Syed Ali Raza Rizvi *et al*., with clearing of visual axis within 12 weeks of treatment.

We report a case of capillary hemangioma in a newborn, which could be treated completely by surgical excision due to its unique pedunculated morphology. Such a case of congenital pedunculated conjunctival capillary hemangioma has not been reported so far to the best of our knowledge.

Case Report

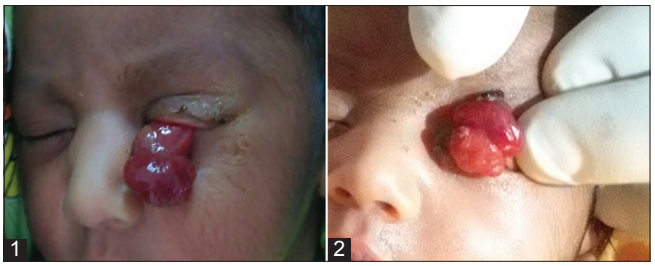
A 2-day-old female baby presented to our pediatric surgery department with a red mass arising from left upper eyelid since birth [Fig. 1]. In the antenatal history elicited, mother was found to be a primipara who had regular antenatal checkups, with no history of any invasive interventions, and the baby was born out of full-term normal vaginal delivery with no perinatal complications.

On examination, the right eye appeared to be normal. Examination of the left eye showed a red, pedunculated, highly vascular, nontender mass of 2.5 cm × 1.5 cm arising from the palpebral conjunctiva of upper lid. The mass had a flattened stalk with terminal lobulations, at the tip of which, there was dried up mixture of blood and discharge. On upper eyelid eversion, the base of peduncle was found to be at 4 mm from lid margin. The lesion appeared to cause mechanical ptosis and covered the visual axis. The eyelid opening appeared inadequate [Fig. 2]. There was minor bleeding from the surface of the swelling on manipulation. The rest of the conjunctiva was normal except for mild congestion. The cornea of the left eye was clear. There was no evidence of similar cutaneous lesions on the rest of the body.

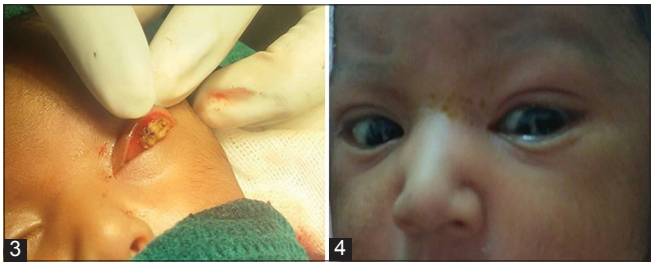
A provisional diagnosis of capillary hemangioma was made. There was also the possibility of pyogenic granuloma and rare possibility of rhabdomyosarcoma. Complete hemogram of the baby showed normal hemoglobin (17.3 g%), white blood cell (11,000/μL), and platelet (2.5 lakhs/μL) levels. Ultrasonogram (USG) showed high-internal reflectivity with irregular acoustic structures suggestive of capillary hemangioma. USG of cranium and abdomen were done to rule out internal hemangiomas but were found to be normal.

Taking into consideration the pedunculated nature, unsightly appearance, possibility of developing amblyopia due to the central location and also to obtain a histopathological diagnosis (to rule out any other tumor), surgical removal was finalized as the treatment. On postnatal day 6, the baby was prepared for general anesthesia. Under anesthesia, detailed examination of the conjunctiva of both eyes was done to rule out deeper lesions and bilaterality. Excision of the mass was done with bipolar electrocautery device by a team consisting of ophthalmologist and pediatric surgeon. It was cut close to tarsal plate, and adequate hemostasis was obtained [Fig. 3].

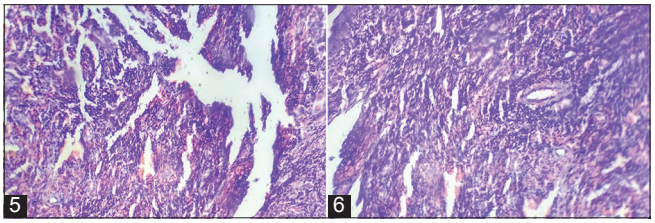
On the 1st postoperative day, the eye was quiet with adequate eyelid opening, clear cornea, and normal fundus [Fig. 4]. On follow-up visits, there was no conjunctival scarring and cornea appeared clear. Multiple sections studied showed a cellular neoplasm composed of plump endothelial cells with well-canalized and poorly canalized vessels, confirming the diagnosis of cellular infantile hemangioma [Figs. 5 and 6]. The



Figures 1 and 2: (1) Baby with left eye upper lid pedunculated swelling arising from palpebral conjunctiva (2) Upon lifting and resting the swelling on upper lid, thin broad pedicle appreciated



Figures 3 and 4: (3) On table image of baby after excision of lesion (4) Baby on first postoperative day



Figures 5 and 6: (5) HPE-Multiple sections studied showed a cellular neoplasm composed of plump endothelial cells with well-canalized and poorly canalized vessels, confirming the diagnosis of cellular infantile hemangioma (6) Multiple sections studied showed a cellular neoplasm composed of plump endothelial cells with well-canalized and poorly canalized vessels, confirming the diagnosis of cellular infantile hemangioma

baby was followed up for 2 months postsurgery and showed no deformities and had adequate lid movements.

Discussion

Capillary hemangioma is common in eyelids but uncommon in the conjunctiva. These are benign vascular tumors which consist of endothelial cell proliferation. It usually presents a few weeks after birth, sometimes at birth, grows for several months, and later regresses spontaneously over years. These occur singly or as part of syndromes (PHACES). Capillary hemangiomas of conjunctiva are rare, and mostly, the lesions are small, demanding only close watch. Usually, cutaneous and orbital hemangiomas are treated with oral steroids or propranolol. Recently, cases of effective management of small conjunctival hemangiomas with topical timolol have been reported. The current literature also shows management of sessile palpebral conjunctival capillary hemangiomas with oral beta blockers. In our case, the baby was brought to us with a red, lobulated mass with stalk present since birth and arising exclusively from the upper eyelid conjunctiva. In order to prevent amblyopia,the lesion was surgically excised,which was possible due to its pedunculated nature.

Conclusion

Conjunctival capillary hemangioma of pedunculated nature has not been reported in literature so far to the best of our knowledge.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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