Case Report

Unusual Case of Acquired Capillary Hemangioma of the Eyelid in an Adult

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Abstract

Acquired capillary hemangioma of the eyelid is very uncommon in adults. To our knowledge, only eight such cases have been reported in the literature till date. We report the case of a 25-year-old female with acquired capillary hemangioma of the left lower eyelid treated by cutting diathermy followed by reconstruction of the cutaneous and cartilaginous defect. This patient had a history of a similar lesion in the left malar region treated 5 years back. This case is being presented to highlight the occurrence of such lesions in an adult and to discuss the dilemmas in the diagnosis and management.

Keywords: Acquired capillary hemangioma, adult capillary hemangioma, vascular anomalies, vascular tumors

Introduction

Capillary hemangioma is the most common vascular tumor in infancy. It usually presents within the first few weeks or months of life.[1] It is characterized by rapid progression followed by spontaneous involution without the risk of recurrence. However, the occurrence of acquired capillary hemangioma of the eyelid is very uncommon in adults. Only eight such cases have been reported in the literature till date.[2-8] We report the case of a 25-year-old female with acquired capillary hemangioma of the left lower eyelid treated by cutting diathermy followed by reconstruction of the cutaneous and cartilaginous defect.

Case Report

A 25-year-old female presented with a growth in the left lower eyelid which started as a small lesion and gradually increased in size over the last 3 years. There was no preceding trauma. There was a history of a similar lesion in the left malar region just below the site of the present growth, which was operated elsewhere 5 years back. On examination, a single sessile, reddish growth measuring 4 cm × 3 cm in size was present in the left lower eyelid. It was firm and rubbery in consistency, noncompressible, with irregular margins, and nonpulsatile. There was no bruit. There were no similar lesions elsewhere. Fine-needle aspiration cytology was suggestive of vascular tissue. All routine blood investigations were normal. Slit lamp and fundus examination were normal. Ultrasound of the abdomen was normal. Left infraorbital Doppler showed well-defined superficial hypoechoic lesion in the left lower eyelid partially abutting the left globe. Computed tomography (CT) orbit (plain and contrast) and three-dimensional (3D) CT scan (volumetric rendering technique [VRT]) were suggestive of intensely enhancing mass lesion – hemangioma in the left infraorbital region with multiple feeding vessels, the prominent one being the left ophthalmic artery entering the lesion from the lateral aspect [Figure 1]. There was no evidence of bony involvement. The patient was subjected to surgical excision using cutting diathermy followed by reconstruction of the cartilaginous defect using auricular concha fabricated to suite the curvature of the lower eyelid contour. The orbicularis oculi muscle defect was closed by mobilizing the remaining muscles to cover the cartilage graft. The cutaneous defect was resurfaced with thick split-thickness skin graft taken from the medial aspect of the arm [Figure 2]. Excisional biopsy revealed a mass composed

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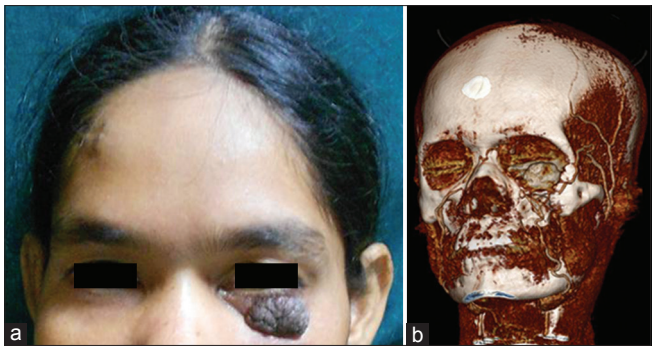

Figure 1: (a) Preoperative image of the patient with capillary hemangioma of the left lower eyelid (written consent of the patient taken). (b) Three-dimensional computed tomography scan (volumetric rendering technique) suggestive of intensely enhancing mass lesion – hemangioma in the left infraorbital region with multiple feeding vessels, the prominent one being the left ophthalmic artery

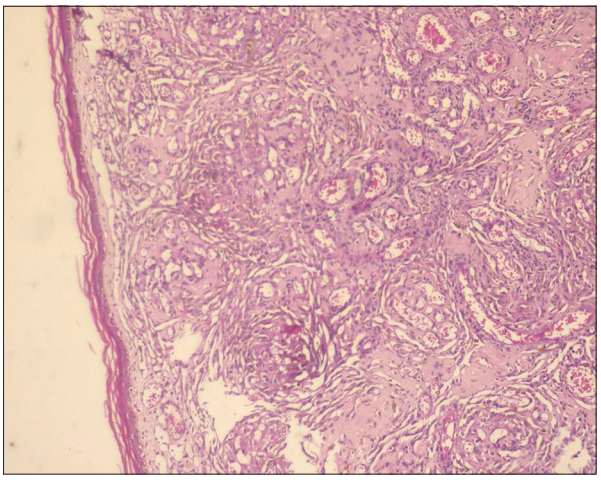

Figure 3: Low-power (×10) histological section of mass stained with hematoxylin and eosin showing lobules of thin- and thick-walled blood vessels lined by endothelial cells in the subepithelial tissue suggestive of capillary hemangioma

of lobules of thin- and thick-walled blood vessels lined by endothelial cells in the subepithelial tissue suggestive of capillary hemangioma [Figure 3]. No recurrence was noted postexcision. The patient was satisfied with the esthetic outcome [Figure 4].

Discussion

Vascular anomalies pose a diagnostic challenge due to inconsistent classification systems, poor understanding of natural history, and overlapping clinical and histological features.[9] The currently used classification, proposed by Mulliken and Glowacki, was adopted by the International Society for the Study of Vascular Anomalies in 1996.[9] Accordingly, vascular anomalies are classified into vascular tumors (lesions characterized by endothelial hyperplasia) and malformations (lesions characterized by dysmorphogenesis and normal endothelial turnover).[1,9,10]

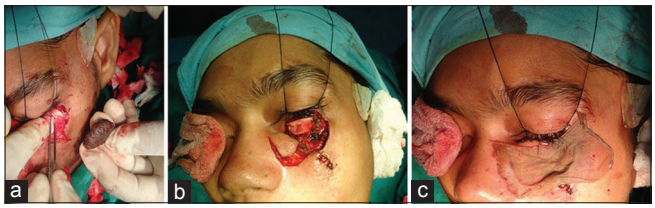

Figure 2: Intraoperative image showing (a) the excised lesion. (b) Reconstruction of the cartilaginous defect using auricular concha. (c) Resurfacing of the cutaneous defect with thick-split thickness skin graft

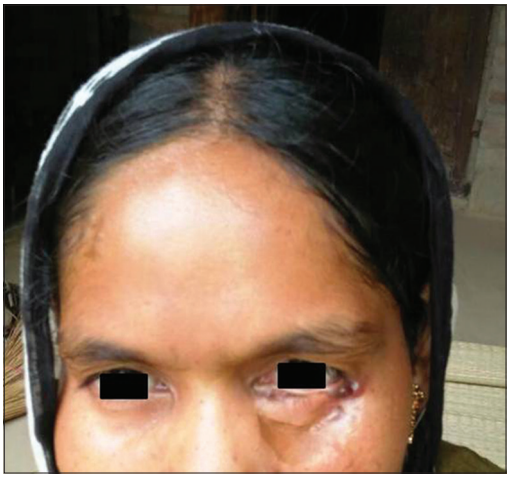

Figure 4: Two-year postoperative image of the patient

Hemangiomas are the most common vascular tumors.[1] They rarely present at birth, show a rapid growth during the first 6 months of life, and spontaneously involute with time.[1] Vascular malformations, on the other hand, are present at birth, show proportionate growth throughout the life of the individual, and are infiltrative in nature.[1,11] The hemangioma feels firm and rubbery and is difficult to compress as compared to a malformation which is readily compressible.[12] It is possible to differentiate a hemangioma from a vascular malformation clinically in 95% of the cases.[12]

Hemangiomas are further classified according to the time of presentation as "congenital" or "infantile."[1] Congenital hemangiomas are rare and present at birth.[1] They either rapidly involute in infancy (rapidly involuting congenital hemangioma) or never involute (noninvoluting congenital hemangioma).[1] Infantile hemangiomas are the most common tumor in infancy and occur in around 4%–10% of the population.[1,10] Hemangiomas can also be classified depending on their depth as superficial, deep, and compound.[9] The superficial hemangioma extends into the superficial dermis and appears red and nodular.[1,12] A deep hemangioma involves the lower dermis or subcutaneous tissue and presents as a protrusion with an overlying bluish hue.[1,12] Compound hemangiomas have both deep and superficial components.[1]

Female sex (preponderance of 3:1–5:1), prematurity, low birth weight, and antenatal chorionic villus sampling (10-fold

risk) are the predisposing factors.[1,10] Isolated lesions account for 80% of the cases, while 20% are multiple.[12] The head-and-neck region is most commonly involved, followed by the trunk and extremities.[12] Multiple lesions are usually associated with involvement of viscera, especially the liver, gastrointestinal tract, lungs, and brain, and should be appropriately investigated.[10,12] Most of the hemangiomas appear during the first 6 weeks of life.[12] It begins as a macular patch, blanched spot, or a localized area of telangiectasia surrounded by a halo followed by rapid proliferation.[12] This rapid growth leads to exhaustion of blood supply, resulting in ischemia and necrosis.[1] This phase is followed by the quiescence phase which lasts from 9 to 12 months where there occurs a slow or no growth of the hemangioma.[1] The final phase is that of involution associated with shrinking of the hemangioma.[1]

Vascular malformations are classified depending on the predominant type of vessels involved as capillary, venous, arteriovenous, lymphatic, or combined malformations.[9] According to the flow characteristics, the lesions can be subdivided as low-flow, high-flow, and combined lesions.[9]

Although this method of classifying vascular anomalies caters to a majority of clinical and histological entities and clearly differentiates between congenital and infantile hemangiomas, there still exists some controversy regarding the nomenclature of similar lesions arising in adults.[13] They are vaguely referred to as adult or acquired capillary hemangiomas by most authors and do not find a place in the current classification system.[13] The clinical and histopathological findings are usually consistent with that of capillary hemangiomas typically seen in infancy.[8] Acquired capillary hemangiomas appear to be true capillary neoplasms and need to be carefully differentiated from neoplastic conditions such as Kaposi's sarcoma, angiosarcoma, acquired tufted angioma, and intravascular papillary endothelial hyperplasia.[3,6] A close differential is pyogenic granuloma, a common cutaneous vascular tumor, which grows rapidly and is commonly confused with a hemangioma.[10] It occurs at any age with a slight female predilection, affecting 1% of pregnant women.[14] These lesions, however, are of smaller size (average diameter 6.5 mm) often associated with crusting of the surface epithelium followed by sloughing of the distal tissue.[10] Repeated and copious bleeding episodes are the rule.[10] Histologically, it is a perithelial, rather than an endothelial, tumor and consists of loose and vascular granulation tissue with an ulcerated or eroded surface epithelium and inflammatory cells.[11,14]

Acquired capillary hemangioma of the eyelid and periocular region is a very rare phenomenon. They have a predilection for the upper lid and brow, and cases involving the lacrimal sac have been also reported.[6] The exact etiology is unknown. It has been associated with hormonal changes and increased estrogen levels during puberty and pregnancy.[2,5] Overexpression of angiogenic growth factors, including vascular endothelial growth factor (VEGF), has been associated with capillary hemangiomas.[7] Adult or acquired "hemangiomas" do not involute like their infantile counterparts.[13] Nonregressive nature of the lesion, cosmesis, visual obstruction, and prevention of accidental trauma and bleeding are the main reasons for seeking treatment.[7]

More than 90% of infantile hemangiomas can be diagnosed on the basis of clinical history and physical examination.[10] Radiologic assessment with ultrasonography, CT, or magnetic resonance (MR) imaging is useful in the pretreatment evaluation of the patient.[10] Ultrasonography scan shows an irregular mass with variable echoes blending into surrounding tissues and is used to rule out orbital involvement and to monitor the size of the lesion.[15] On CT scan, capillary hemangiomas appear as homogeneous soft-tissue masses without destruction of the underlying bone.[15] The use of contrast CT and MR imaging helps in delineating the border and identifying the feeder vessels.[15] 3D CT VRT can be used to delineate the fine anatomical details which are difficult to evaluate with axial reconstructions alone. Angiography is rarely used to identify the feeder vessels for ligation or embolization in life-threatening hemangiomas unresponsive to other therapies.[15] Tissue biopsy may be rarely needed to differentiate the lesion from neoplastic and aggressive lesions.[15]

Although most of the infantile hemangiomas undergo spontaneous involution, some may require intervention.[15] Nonsurgical interventions include the use of corticosteroids which accelerate the regression of the lesion.[10] Intralesional corticosteroids (triamcinolone) are used for small, well-localized hemangiomas while systemic corticosteroids for large or multiple hemangiomas.[10] Recombinant interferon alfa (2a or 2b), vincristine, cyclophosphamide, imiquimod, and antiangiogenic agents such as bevacizumab are the other drugs found to be effective in life-threatening hemangiomas.[10,15] Their use is limited by their variable responses and toxicities.[10] Systemic propranolol has also been used successfully.[15] The exact mechanism of action is not known, but vasoconstriction, decreased expression of VEGF, and induction of apoptosis of capillary endothelial cells are supposed to cause regression of the lesion.[15] Surgical resection is recommended in cases where conservative therapy has failed, and where the hemangioma is blocking the airway or vision or is bleeding.[10,15] Preoperative imaging, including MR imaging, MR angiography, and possibly an angiogram, is necessary to identify the feeding vessels.[15] Low-level radiotherapy can speed the regression of the mass by creating microembolisms in the tumors.[15] Carbon dioxide, argon, neodymium–yttrium aluminum garnet, and flash-lamp pumped-dye laser have also been used in the treatment.[15]

There is, however, no standard treatment modality for the management of adult capillary hemangiomas, though many treatment options have been tried successfully. These lesions have been managed using intralesional corticosteroids, bevacizumab, and cutting diathermy without any evidence of recurrence. Our patient was treated with cutting diathermy

followed by reconstruction of the cutaneous and cartilaginous defect. There has been no recurrence till date.

Conclusion

This case is being presented to sensitize surgeons about the uncommon occurrence of acquired capillary hemangioma of the eyelid which responds to standard treatment modalities. It should therefore be included in the differential diagnosis of lesions in this region. Also, a complete understanding of the natural history of the tumor and its response to different therapies is essential, given the cosmetic significance of adequately treating these lesions.

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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