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Isolated Conjunctival Lymphangioma and Epithelial Inclusion Cysts

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Abstract

We report a case of an isolated conjunctival lymphangioma combined with epithelial inclusion cysts on a 34-year-old male. Examination disclosed a yellow-greyish multiloculated cystic lesion of the superior-nasal bulbar conjunctiva in the right eye with dilation of lymphatic vessels. The patient was asymptomatic, the best-corrected vision was not impaired, and the rest of the ocular examination was unremarkable. Excision of the lesion with amniotic membrane graft replacement was performed. Histopathological examination of the lesion demonstrated numerous anastomotic, thin-walled, dilated lymphatic vessels with flattened endothelium and intervascular lymphocytes accumulation and also, there were several small inclusion cysts in the conjunctiva, which is consistent with conjunctival lymphangioma diagnosis combined with epithelial inclusion cysts. The patient remains asymptomatic, and there has been no relapse of the lesion during one year of follow up. We consider the surgical excision of isolated conjunctival lymphangioma as an optimal treatment modality, with a very low risk of recurrence.

Keywords: Conjunctival lymphangioma; Lymphatic vessels; Superior-nasal ulbar conjunctiva; Nonne-milroy-meige disease

Introduction

In January 2021, a 34-year-old male presented to the Cornea-Uveitis department at Malayan Eye Center, Yerevan, with red eyes after exposure to dust in windy weather. Bacterial conjunctivitis was diagnosed and treated in 10 days.

The slit-lamp examination revealed also an asymptomatic yellow-greyish multiloculated cystic lesion of the superior-nasal bulbar conjunctiva in the right eye with dilation of lymphatic vessels. On ocular examination, his BCVA was 20/20 in both eyes, IOP and pupils' reflexes were normal. Anterior and posterior segment examinations were within normal limits. The patient had no proptosis and no limitations on eye movements.

Past medical history was unremarkable for any systemic disorders.

Case Presentation

The patient claimed to have noticed the described lesion about five years ago, and according to him, the increase in size was negligible since then. Past ocular history was remarkable also for the presence of prior blunt trauma to the right eye, although its relation to the lesion occurrence was unclear.

A decision was made to perform an excisional biopsy to reach a diagnosis. The lesion was removed under topical anesthetic, and an amniotic membrane graft was placed in the region of scleral exposure.

Histopathological examination of the lesion demonstrated a thickened conjunctival segment, formed numerous anastomotic, thin-walled, dilated lymphatic vessels with flattened endothelium. Lymphocytes were accumulated in the intervascular spaces and peripheral areas. In addition to the described neoplasm, there were several small inclusion cysts in the conjunctiva (Figures 1 and 2).

A rare diagnosis of isolated conjunctival lymphangioma combined with epithelial inclusion cysts was made.

Removal of the lesion led to the resolution of the case, and there has been no relapse to date (February 16, 2022).

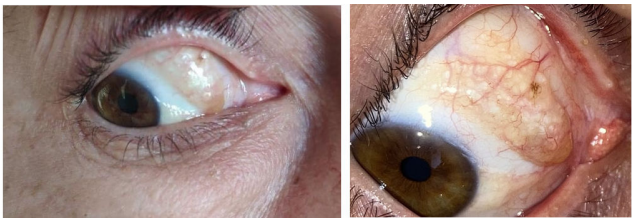

Figure 1: Preoperative appearance of the described lesion.

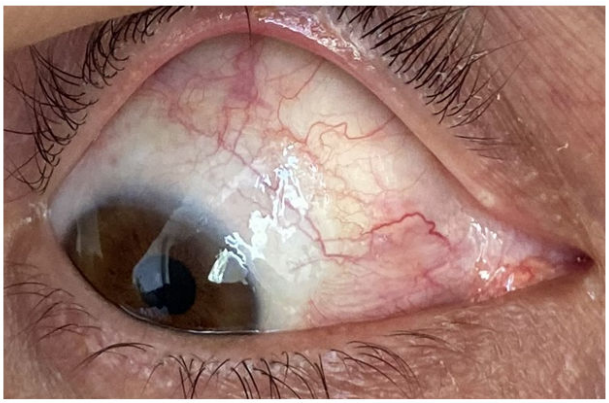

Figure 2: Postsurgical appearance, mild conjunctival scar.

Results and Discussion

Lymphangioma is a benign, hamartomatous tumor of the lymphatic vessels. Solitary or multifocal involvements can be observed. It can be presented as an isolated conjunctival lesion, but often represents a superficial component of an orbital lymphangioma that can be associated with pain, proptosis, motility problems, and vision loss. Lymphangioma is usually present at birth and may enlarge slowly [1,2].

Biopsy and a histopathological report are essential for the diagnosis [3].

Histopathologically it appears as a nonencapsulated, irregular mass composed of numerous cyst-like ectatic channels that are lined by flattened endothelial cells. These channels are separated by loose connective tissue that contains lymphocytes that sometimes form lymph follicles.

The etiology of conjunctival lymphangioma is not clear. It is usually a sporadic unilateral occurrence, which can be exacerbated by intralesional hemorrhage with large "chocolate cyst" formation or by upper airway infections that lead to lymphoid hyperplasia. It also has been recognized as part of Turner's syndrome and Nonne-Milroy-Meige disease.

The role of trauma in lesion growth progression has not been described in the literature.

Nevertheless, as relevant to our case, trauma is a stimulus for epithelial inclusion cysts formation, which usually is derived from the inclusion of conjunctival epithelium in the substantia propria. In these cases, complete excision is necessary to prevent a recurrence.

Isolated conjunctival lymphangioma is a rare condition. In the clinical series of 1643 conjunctival tumors, there were 15 lymphangiomas, accounting for 24% of conjunctival vascular lesions and for less than 1% of the 1643 lesions.

The average age at presentation is 25 years (range from birth to 65 years), with an average 3-years duration of symptoms at presentation [4-6].

In a review of 13 cases of orbital-adnexal lymphangiomas, classified these lesions as superficial, deep and combined. Superficial conjunctival lymphangiomas generally appear as a visible mass, consisting of isolated multicystic vascular abnormalities of cosmetic significance only, without affecting vision or the globe, as in our case.

There are multiple treatment modalities for lymphangiomas of the ocular adnexa.

Surgical debulking in cases of superficial lymphangioma is often curative, as occurred with our patient. In similar cases and the small classical lesions of skin and underlying subcutaneous tissue, this treatment option is 91-100% curative [7].

Alternative treatments also have been described, as CO_2 laser ablation, intralesional bleomycin, β-irradiation using strontium-90 applicator, and brachytherapy also can be used with partial success. Spontaneous resolution due to conservative, nonsurgical management has also been reported [8-12].

Conclusion

In summary, we present the first case of isolated conjunctival lymphangioma with epithelial inclusion cysts at Malayan Eye Center in the last 20 years. Our patient claimed to have had blunt trauma to the eye in the past. We assume it could have been a trigger or a precipitating factor, but further investigations are needed to prove the possible relationship to its occurrence.

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