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ORIGINAL ARTICLE

Lymphangiectasia Haemorrhagica Conjunctivae – An Uncommon Entity Revisited

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Abstract

We present a case of LHC, which presented to us with spontaneous appearance of reddish discoloration of the conjunctiva, and was evaluated for corkscrew conjunctival vessels by neuroimaging, which was normal. The haemorrhages subsided spontaneously in 3 days, revealing the conjunctival lymphangiectasia and unmasking the diagnosis.

Keywords

Conjunctival lymphangiectasia, Lymphangiectasia Haemorrhagica Conjunctivae

Introduction

Lymphangiectasia Haemorrhagica Conjunctivae (LHC) is a rare condition that was first described as early as 1880 but fails to be recognized still due to its unfamiliarity. The presentation is often insidious and dramatic and can raise a false alarm leading onto neuroimaging modalities which may be unwarranted if the condition can be recognized on presentation.

Case Description

A 54-year-old female attended the out-patient clinic with complaints of having noticed reddish discoloration of the left eye noticed the same day. This was unassociated with any pain or diminution of vision. There was no history of trauma or eye rubbing. She had not noticed any similar occurrences in the past. She was a known hypertensive for the past 6 years, with no other comorbidities and was on antihypertensive medications alone, but no antiplatelets or anticoagulants.

On examination, her Best corrected vision was 6/6, N6 in both eyes. Right eye examination was within normal limits. The left eye showed dilated and tortuous conjunctival vessels all around in the perilimbal region with Haemorrhage along the vessels in a tortuous vascular pattern unlike an ordinary subconjunctival haemorrhage (Figure 1).

A gonioscopy showed normal angles in the right eye and faint appearance of blood in Schlemm's canal in the temporal quadrant alone in the left eye, but the intraocular pressures recorded were within normal limits in both eyes. Dilated fundus evaluation and OCT imaging were within normal limits and did not reveal any evidence of choroidal haemangioma or any other vascular lesion.

Her Blood investigations were within normal range. She was advised Magnetic Resonace (MR) Imaging of brain and orbit and MR venography to rules out any venous outflow obstruction. MR imaging did not show any dilatation of superior ophthalmic vein. There was no lateral convexity of the cavernous sinuses. MR venography showed normal morphology of the cortical venous system, and the orbital contents showed normal morphology and signal.

However, when the patient presented 3 days later with the reports, the hemorrhages were resolving well with the irregular dilated sausage shaped conjunctival lymphatic channels being delineated in the areas where the hemorrhages had completely resolved (Figure 2). The patient was reassured and advised observation alone and to watch for recurrences.

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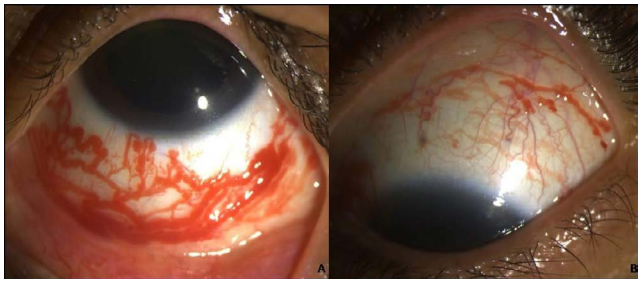

Figure 1: Dilated conjunctival vessels in the perilimbal region with haemorrhage along the vessels in a tortuous vascular pattern.

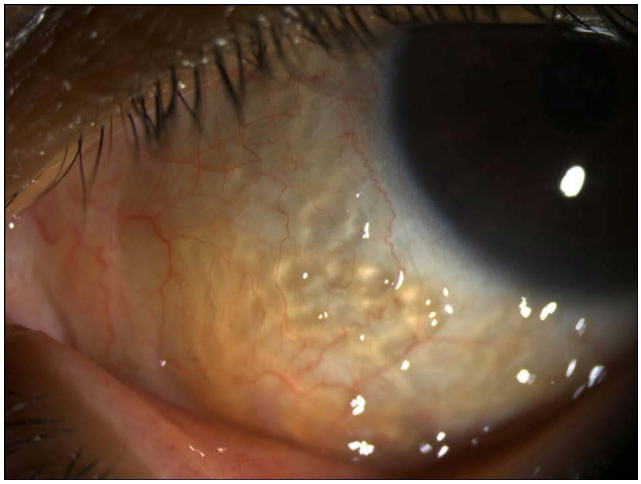

Figure 2: Resolving haemorrhages with the irregular dilated sausage shaped conjunctival lymphatic channels delineated in the areas where the haemorrhages had completely resolved

Discussion

LHC was 1st described and named by Leber as early as 1880 [1]. Since then, through the years several such cases have been described, but the first report of 5 cases of LHC in literature was in 1969 by Philip Awdry from Moorfields Eye Hospital [2]. Almost 4 decades later in 1998, the condition was redescribed in a series of 9 eyes by Lochhead et al. [3]. Still, it appears that the entity is under-recognized.

Conjunctival lymphangecatsia is rare condition, involving dilatation of the normal lymphatic vessels of the bulbar conjunctiva, which accounts for 1% or less of all conjunctival lesions. It appears as a diffuse form which appears clinically as chemosis or a focal form which results in cysts or a string of pearls' appearance [4].

The pathophysiology of LHC is not clearly understood. The pericorneal lymphatic ring (Lymphatic circle of Teichmann) forms a plexus of tiny vessels measuring about 1 mm in size along the limbus [4]. These coalesce to form radial vessels which drain from around the limbus into larger collector channels running circumferentially between 4-8 mm of the limbus [4]. There are connections between the deep conjunctival venous plexus and these collector channels. Unidirectional lumenal valves present in the lymphatic system prevent backflow, but these valves may not be completely effective, and it is believed that occasionally, retrograde flow results in lymphatic channels filling up with blood resulting in LHC [4].

When the whole circumferential channels get filled with blood, LHC can result in a dramatic picture as in our case and radiological imaging modalities may be immediately unwarranted as the condition typically resolves spontaneously in 3-4 days.

Conclusion

LHC may be more common than expected and an awareness of its characteristics will be useful in examining cases presenting as recurrent subconjunctival haemorrhages of bulbar conjunctiva. The difference in the pattern of haemorrhage conforming to a vascular tortuous pattern as well as the speedy resolution should give the clue to differentiate from a more diffuse and longstanding subconjunctival haemorrhage. Also, the delineation of the lymphatic channels when they are drained of blood revealing the characteristic appearance would confirm the diagnosis.

More than that, Conjunctival lymphangiectasia has also been described as a potential noninvasive, surrogate biomarker of Fabrys disease after it was described to have 85% incidence in a cohort of 13 patients with Fabrys disease [5]. Hence awareness among ophthalmologists about its existence and modes of presentation will help with early recognition and focused investigations.

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