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Case Report
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Prominent lymphatic vessel in a functioning bleb after repeat XEN gel stent implantation surgery in pseudoexfoliative glaucoma: a case report

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Abstract
XEN gel stent implantation (XEN) surgery is becoming more popular due to its safety profile and efficacy, with conjunctival lymphatics thought to be critical in maintaining bleb drainage. We are reporting the case of a 75-year-old man with right eye pseudoexfoliative glaucoma who had two previous episodes of failed XEN surgery despite given needling and antimetabolite. He had a repeat XEN surgery in our center and his intraocular pressure was under control at the time of writing without any topical glaucoma medication, needling, nor antimetabolite. On examination, there was presence of prominent, dilated, and irregular lymphatic vessel originating from the distal end of the bleb. Anterior segment optical coherence tomography identified the lymphatic vessel located in the superficial Tenon's layer, which is most likely an initial lymphatic but larger in diameter. This dilated lymphatic vessel may be an important factor for his currently successful filtration surgery.

Keywords: *bleb drainage, conjunctival lymphatics, glaucoma, pseudoexfoliation glaucoma, XEN surgery*

Introduction

Microinvasive glaucoma surgery (MIGS) is becoming more popular due to its safety profile and efficacy. Morgan *et al*. reported that gelatin micro-fistulae implantation had 83% success rate with intraocular pressure (IOP) less than 21 mmHg and reduction of IOP-lowering medications from 3.0 to 0.9 at 6 years follow-up with no significant complications.[1] Similar microfistulae being marketed as XEN gel stent (XEN) with smaller dimension (Allergan, Irvine, CA, USA), is one of the newer modalities to lower IOP by forming a drainage bleb. Conjunctival lymphatics are thought to be critical in maintaining bleb drainage as evidenced by extensive experimental study.[2] It is arguably acceptable to say

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that more dilated and developed lymphatic vessels may have increased aqueous humor drainage, resulting in lower IOP. We are reporting a case of prominent lymphatic vessel in a functioning bleb after repeat XEN surgery.

Case presentation

A 75-year-old man with underlying right eye (RE) pseudoexfoliative glaucoma had XEN implant performed by an external ophthalmologist in 2018 which failed after 2 months due to unknown reasons. It was removed and a second XEN stent was implanted. Unfortunately, despite treatment with needling and 5-fluorouracil injection, the second implant failed and the patient was started back on topical antiglaucoma medication soon after the surgery. The patient was then referred to our institution.

Initial assessment in our institute showed RE visual acuity was 6/6, cornea was clear, conjunctiva was not inflamed with mobile conjunctiva superonasally, pseudophakic, and open angle in all four quadrants on gonioscopy. On gonioscopy, the XEN stent was seen superiorly in the 1 o'clock position entering the ciliary body with the proximal portion angled towards the iris and with the iris plugging the lumen of the XEN, which was considered the cause of failure. The IOP was 14 mmHg with guttae timolol-travoprost and brimonidine-brinzolamide fixed combinations and cup-to-disc ratio (CDR) of 0.7. His left eye (LE) visual acuity was 6/7.5, pseudophakic, IOP of 13 mmHg without any topical antiglaucoma medication, and CDR of 0.4. The patient complained of asymmetrical skin color changes due to topical eyedrops and was keen for surgery. Options and risks regarding XEN stent and glaucoma drainage device were discussed with the patient. The mobile conjunctiva within the superonasal quadrant suggested that little fibrosis had occurred from the prior two XEN stents, thus making this region potentially suitable for reimplantation of XEN. Our usual recommendation in the setting of failed XEN stent is for a glaucoma drainage device given that the usual cause of failure is fibrosis around the stent distal tip. After discussion with the patient regarding the likely outcomes, the patient wished to attempt a third XEN implant, which was performed at our institute. RE ab-interno XEN implant with subconjunctival mitomycin-C 0.01% (5 μg) was performed using eye fixation to free one hand allowing continuous gonioscopy during surgery and reasonably precise localization of the XEN stent through the trabecular meshwork, as previously described.[1] No intraoperative complications occurred.

Postoperative review at week 1 showed superonasal diffuse bleb extending for 3 clock hours with IOP of 9 mmHg. At 1-month, postoperative RE IOP maintained at 10 mmHg with slightly elevated diffuse bleb, as previously noted. In addition, there was a prominent lymphatic vessel noted originating from the distal end of the bleb at approximately 1 o'clock running circumferentially towards the

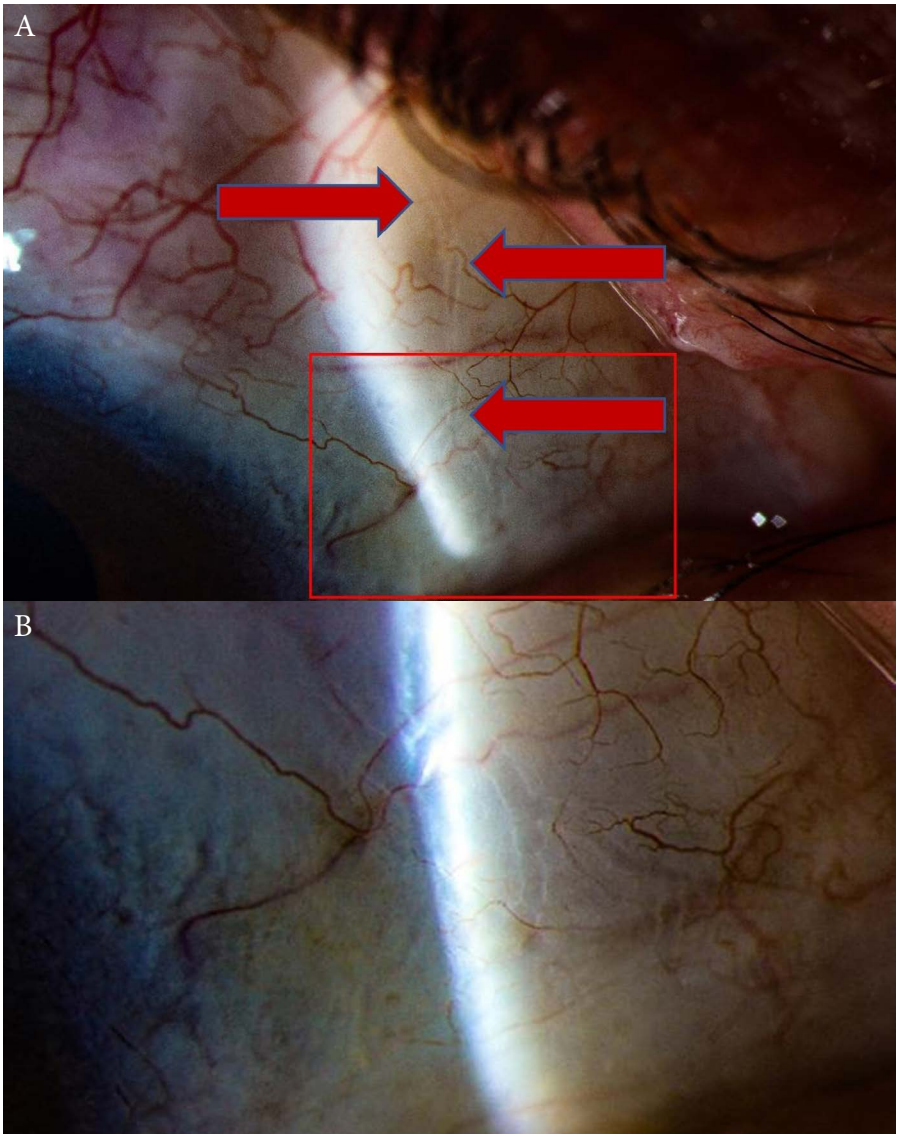

Fig. 1. (A) Prominent and dilated lymphatic vessel (red arrow) captured on slit lamp examination. The lymphatic vessel started superonasally, where the XEN bleb is located, and drained downwards nasally. (B) Magnified area of prominent lymphatic vessel (red square). The vessel with whitish wall and translucent inner segment is clearly demarcated from other blood vessels. The vessel's irregular caliber is clearly noticeable, which further supports that it is likely a lymphatic vessel.

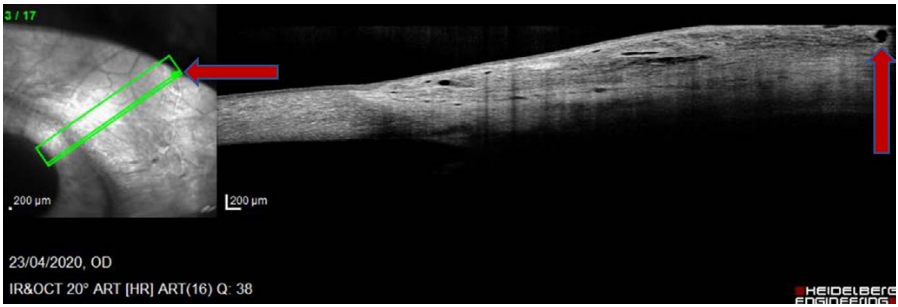

Fig. 2. Anterior segment optical coherence tomography scans of the bleb. The circular lymphatic vessel (red arrow) is located at the superficial Tenon's layer.

inferonasal quadrant (Fig. 1A) at a distance of 3–4 mm from the limbus. Enlarged image also showed the vessel having an irregular caliber (Fig. 1B). This vessel was flat with no cystic dilation and no surrounding chemosis, which tends to occur with lymphangiectasia.[5] Additionally, the vessel was not present prior to surgery. We concluded that this was likely to be a dilated normal lymphatic vessel.

Anterior segment optical coherence tomography identified the lymphatic vessel located in the superficial Tenon's layer (Fig. 2), which was most likely an initial lymphatic vessel. It was well circumscribed, round, and hyporeflective, measuring approximately 111 µm in diameter. Subsequent follow-up at 3 months revealed RE IOP of 13 mmHg without topical antiglaucoma medication. No needling or 5-fluorouracil injection were given after the latest XEN implant.

Discussion

It is postulated that aqueous humor is removed through transconjunctival filtration, reabsorption through walls of degenerated veins, or into aqueous vein or absorption into superficial conjunctival lymphatics.[3] The conjunctival lymphatic network has two layers, initial lymphatics and precollectors. Initial lymphatics are mostly located in the superficial region of Tenon's capsule and are much smaller in caliber. The caliber of the initial lymphatic vessel in our patient was large compared to our initial study on lymphatic and lymphatic capillaries, which are much smaller in caliber.[2] Hence, it was most likely formed after the last XEN implant, allowing aqueous outflow into the superficial Tenon's space and subsequently into this lymphatic vessel, causing its engorgement. It is important to notice that the appearance of the conjunctiva and its blood vessels are almost normal, indicating that there was no significant inflammation and scarring at the site of the bleb.

In our previous experimental study, fluorescein dye was injected into the anterior chamber to determine aqueous humor outflow pathway from the

anterior chamber into subconjunctival tissue, followed by its removal from subconjunctival tissue. A critical role of conjunctival lymphatic drainage in successful outcomes of glaucoma filtration surgery has been demonstrated.[2] Khoo *et al.* found that eyes with lymphatics that connected to drainage trabeculectomy bleb had greater IOP reduction and required fewer eye drops.[4] Similarly, our patient had a prominent dilated lymphatic vessel that developed after surgery and originated from the drainage bleb, which allowed good IOP control. These findings appear more likely to be due to lymphatic dilation rather than conjunctival lymphangiectasia given the time course as well as the lack of chemosis and cystic swelling.[5] Successful filtration surgery depends on consistent aqueous humor outflow pathway from the anterior chamber into conjunctival tissue followed by its removal from the conjunctival tissue. Our patient had two previous episodes of failed XEN implantation despite given needling and antimetabolite. Therefore, he was at risk of failure in further surgery due to fibrosis and scarring, which may occlude the outflow pathway. Despite this, his third XEN implant was still functioning well at the time of writing without further intervention with a noticeable dilated lymphatic vessel.

Conclusion

The patient's dilated lymphatic vessel may be an important factor for his currently successful filtration surgery. However, further study and research is needed on conjunctival lymphatics with regards to physiologic function and relation to IOP control. The photographs contained in this case report illustrate that conjunctival lymphatics can occasionally been seen and that quantification of various lymphatic properties may aid drainage surgery outcomes.

Declarations

Ethics approval and consent to participate

Not required.

Consent for publication

The patient provided informed consent for the use of their clinical data in this case report.

Competing interests

None to disclose.

Funding

None to disclose.

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None to disclose.

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