



Implantation iris cysts developing 24 years after penetrating keratoplasty

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ABSTRACT

Purpose: To report a case of iris implantation cysts occurring 24 years after penetrating keratoplasty and its management.

Observations: A 60-year-old man was referred for diagnosis and management of white iris masses of the right eye. He had undergone bilateral penetrating keratoplasty 24 years before without complication. The clinical findings were suggestive of iris implantation cysts and Ultrasound Biomicroscopy (UBM) and anterior-segment optical coherence tomography confirmed the diagnosis. The patient did not develop any ocular complications from the cysts after one-year follow-up from the diagnosis of iris implantation cysts.

Conclusions and importance: Iris implantation cysts are rare benign tumors that develop after the ectopic implantation of epithelial cells within the iris stroma. They can be congenital or secondary to penetrating trauma or surgery. Their diagnosis relies on clinical examination and UBM. In case of intraocular complications, treatment may be required, otherwise observational follow-up is appropriate.

1. Introduction

Iris tumors can be divided in two large groups, solid (~80%) and cystic lesions (~20%).^{1,2} Cystic lesions are composed of one or more layers of epithelial cells without being composed of chorion cells. They may be primary or secondary to a transfixing wound affecting the cornea, leading to corneal epithelial cell implantation and growth in the iris stroma. Epithelial, mesenchymal, and/or conjunctival goblet cells can also be introduced into the anterior chamber as result of trauma, giving rise to a secondary implantation cyst. Cysts are therefore not always composed of epithelial cells and could also have keratinizing epithelium.

Wounds can be due to intraocular surgery, the most common being cataract surgery, or to penetrating trauma. These cysts can be classified by their location in the iris and their tissue origin. Most of them are superficial and derived from iris pigment epithelium (IPE), termed IPE cysts. Some of them are growing inside the iris stroma, secondary to the implantation of epithelial cells, and can be either congenital or acquired. Small iris tumors may remain asymptomatic but when they increase in size they manifest most often with visual loss, raised intraocular pressure, iris deformation, pain, or hyphema.

Here we describe a patient presenting implantation cysts of the iris

that appeared 24 years after a penetrating keratoplasty.

2. Case report

A 60 year-old man was referred for white masses that had developed in the anterior chamber of the right eye. He underwent bilateral penetrating keratoplasty for progressing keratoconus in 1995 and was followed up yearly since the procedure by his attending ophthalmologist. In 2019 (24 years after the surgery), he observed the iris lesion and was referred to our department thereafter. The ophthalmologic examination he had undergone the year before was strictly normal. Best-corrected visual acuity was limited to 20/32 due to the presence of cataract; and intraocular pressure was 19 mmHg. Slit-lamp examination and gonioscopy revealed three amelanotic confluent lesions located superiorly in the anterior chamber (Fig. 1), adjacent to, but not invading the irido-corneal angle (Fig. 2). The lesions were transparent upon transillumination, which was suggestive of cystic lesions. The patient also presented corectopia and uveal ectropion due to iris traction by the lesions. Ultrasound Biomicroscopy (UBM, Aviso, Quantel Medical, Clermont Ferrand France) with a 50-MHz probe confirmed the cystic nature of the lesion with a slightly hypoechogenic mucous content (Fig. 3). High Frequency Color Doppler Imaging confirmed the absence of intralésional or parietal vascularization, consistent with the diagnosis of

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Abbreviations

| | |
|-----|--------------------------|
| UBM | Ultrasound Biomicroscopy |
| IPE | iris pigment epithelium |

liquid-filled cysts. Anterior-segment optical coherence tomography (AngioVue RTx100, Optovue Inc., Fremont, CA) visualized round-shaped lesions with thin hyperreflective walls and isorefective content (Fig. 4). Moreover, the cystic lesions were adjacent to an hyperreflective penetrating structure through the cornea, that corresponded to the penetrating keratoplasty scar.

Overall, clinical and imaging characteristics supported the diagnosis of iris implantation cysts. After one-year follow-up the cysts remained stable and the patient had not developed any complication.

3. Discussion

According to the 4th edition of the WHO classification of eye tumors released in 2018, iris cysts can be divided into two categories, IPE cysts, originating from the IPE on the posterior surface of the iris, and implantation cysts.² These two entities can be primary or secondary, although IPE cysts are more likely to be reported as primary, whereas implantation cysts are more likely to be secondary. IPE cysts are frequent, and most often congenital. They do not usually lead to complications and therefore do not need to be treated.

Implantation cysts of the iris are rarer tumors.³ They are due to the presence of an ectopic epithelium cell inside the iris stroma. Surface epithelial cells from the conjunctiva or the cornea can deposit on the iris and grow inward, forming implantation cysts. They can be primary, acquired during embryologic development and present at birth,⁴⁻⁶ or secondary to penetrating trauma⁷ or intraocular surgery.^{8,9} Most reported cases of secondary implantation cyst occurred after cataract surgery, as it is the most frequently performed surgical procedure.

Few cases are described in the literature, the majority of which lead to complications. Most the reported cases were managed by surgical treatment, as described below.

Histopathological analysis of the cysts has been detailed by some authors following surgical excision.⁸ In these cases, the cyst wall was composed of well-differentiated non-keratinizing squamous epithelium, resembling the normal corneal epithelium, surrounded by a layer of fibrovascular tissue with islands of iris pigment epithelium cells.

The diagnosis of epithelial iris cysts may be challenging, since they may harbor the same clinical and ultrasonographic characteristics as

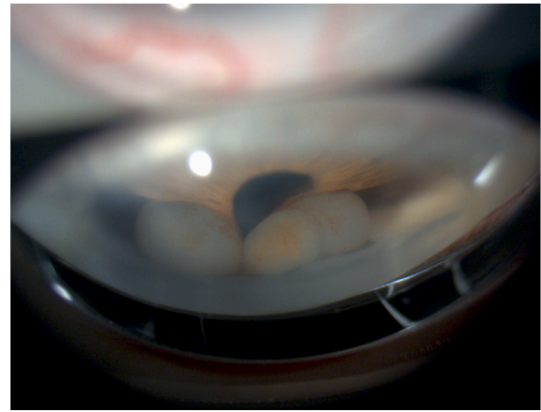


Fig. 2. Right eye gonioscopy showing the iris implantation cysts in the superior quadrant, without invasion of the iridocorneal angle beyond the margins of the lesion.

primary implantation iris cysts.^{10,11} In the present case, the history of penetrating keratoplasty supported the hypothesis of secondary iris cyst. In this patient, the lesions appeared 24 years after the surgery, and a normal ocular examination one year before ascertained the accuracy of the timing of cyst development reported by the patient. To our knowledge, this is the longest period between surgery and development of secondary iris cysts reported in literature.

Possible differential diagnoses of these benign cystic tumors are solid iris tumors, such as achromic melanomas, metastasis or rarely iris lymphomas. Iris cysts should be investigated by UBM to ascertain the diagnosis, before any therapeutic decision.¹²

When they do not induce any anatomical or functional complication, these lesions do not require treatment, because of potential surgical complications, and of the high risk of recurrence if not entirely removed.¹³ However, when threatening visual function, or inducing glaucoma, corneal decompensation or secondary uveitis, or when associated with cataract, several conservative therapeutic options can be considered. They include fine-needle aspiration alone,¹⁴ that carries a high risk of recurrence, or intracystic injection of absolute alcohol.¹⁵ Laser treatment has also been reported, relying on Nd:YAG laser for cystotomy¹⁶ and argon laser for photocoagulation of the collapsed cyst wall.¹⁷ Finally, surgical management is possible if less invasive approaches have failed and consists in the *in toto* excision of the cyst, frequently associated to sectorial iridectomy to avoid recurrences.¹⁸ Regarding the surgical management of cataract in eyes with iris cysts, there is very limited evidence in the literature, and no consensus to date.

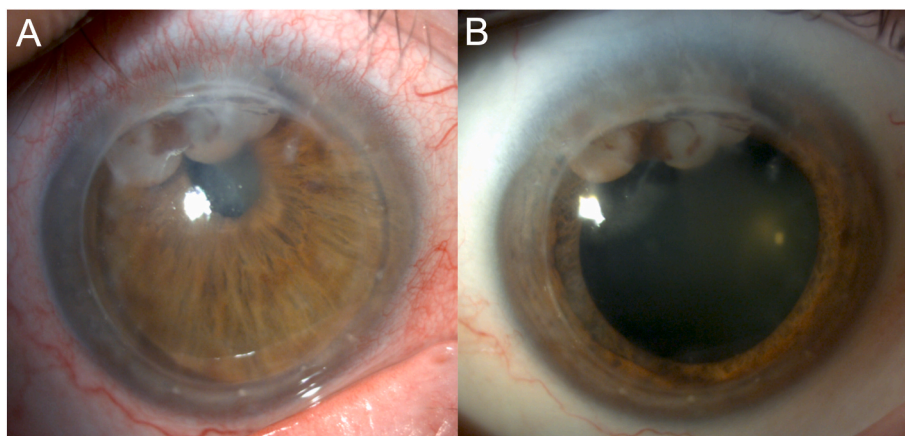


Fig. 1. Right eye biomicroscopy showing epithelial implantation cysts on the iris mimicking an iris tumor, following uneventful full-thickness keratoplasty in a 60-year old male patient. A. Before pupil dilation. B. After pupil dilation (note the regression of conjunctival hyperaemia after topical neosynephrine).

In the present case, our patient has not undergone treatment to date because of the absence of ocular complication, and semestrial observation did not reveal any progression after one-year follow-up.

4. Conclusion

Iris implantation cysts are rare iris tumors, either primary or secondary to transcorneal wound, either traumatic or due to intraocular surgery. History of past ocular penetrating trauma or surgery must be searched for upon anamnesis. Iris implantation cysts are benign lesions, however malignant lesions must be ruled out with ultrasound biomicroscopy. In the present case, the lesions appeared exceptionally more than two decades after intraocular surgery.

5. Patient consent

Patient was informed of this report and oral approval was obtained.

Author declaration

[Instructions: Please check all applicable boxes and provide additional information as requested.]

Conflicts of interest

We wish to draw the attention of the Editor to the following facts, which may be considered as potential conflicts of interest, and to significant financial contributions to this work.

The nature of potential conflict of interest is described below:

No conflict of interest exists.

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

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No funding was received for this work.

Intellectual property

We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property.

Research ethics

We further confirm that any aspect of the work covered in this manuscript that has involved human patients has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

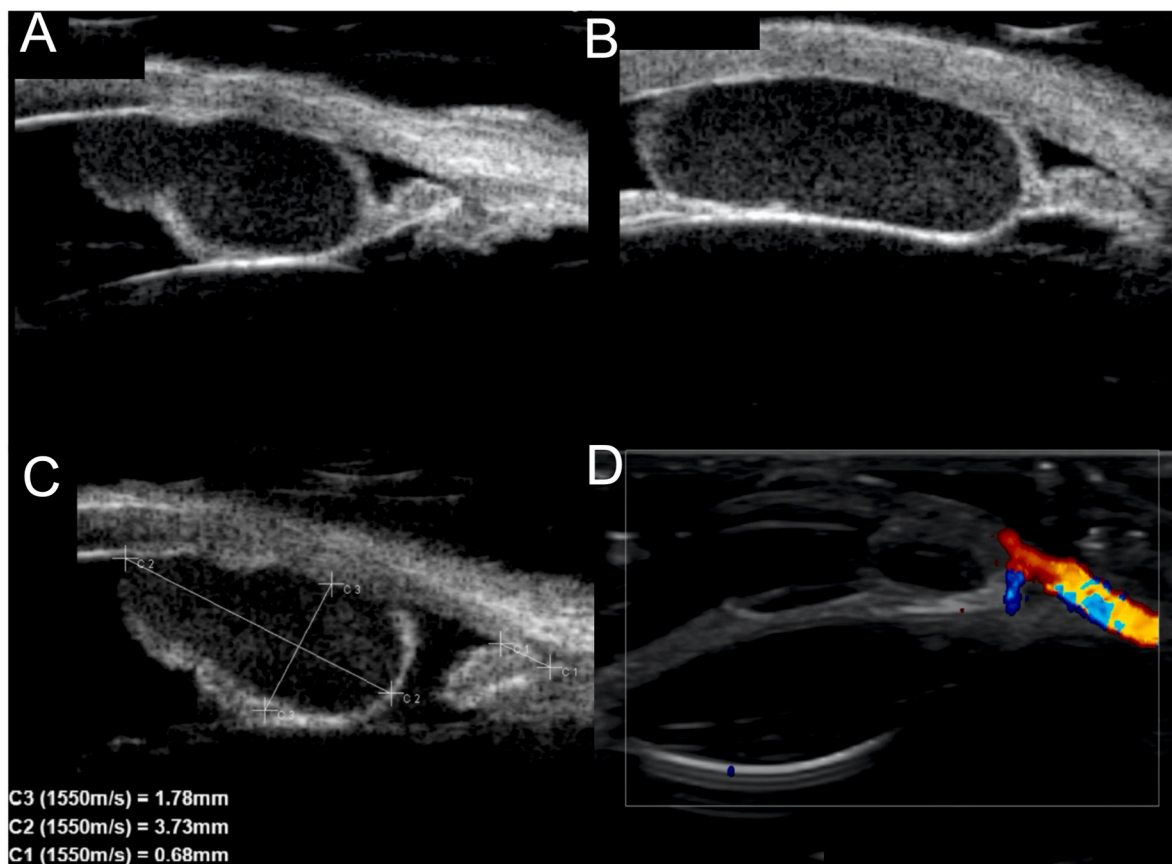


Fig. 3. Right eye ultrasound biomicroscopy (UBM) of the cysts. A and B. Vertical and horizontal sections of the cystic lesion. C. Measurements of the dimensions of the lesion. D. Doppler mode showing the absence of intralesional or parietal vascularization supporting the diagnosis of benign cysts. Note the blood flow detected in the ciliary body and iris adjacent to the lesion (color signal) and note in A, C, and D, the coalescence of two cysts, which, besides, have a very thick wall. (Device: Aviso Quantel with 50-MHz probe). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

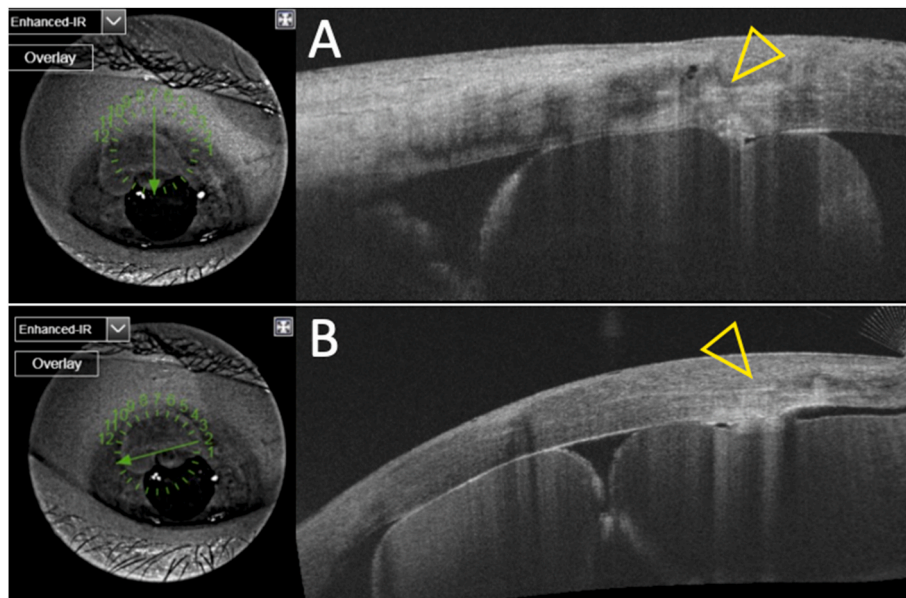


Fig. 4. Anterior-segment optical coherence tomography showing round-shaped lesions with thin hyper-reflective walls, and iso-reflective content, consistent with the diagnosis of iris implantation cysts. A. Vertical section. B. Horizontal section. The cystic lesions were adjacent to an hyperreflective penetrating structure through the cornea, identified as the penetrating keratoplasty scar (arrow).

IRB approval was obtained (required for studies and series of 3 or more cases).

Written consent to publish potentially identifying information, such as details or the case and photographs, was obtained from the patient(s) or their legal guardian(s).

Authorship

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