ELECTRONIC CLINICAL CASE

A traumatic corneoscleral epithelial cyst

Un kyste cornéoscléral d’origine traumatique

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MOTS CLÉS
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Summary We describe a case of a patient with a corneoscleral epithelial cyst originating from a traumatic scleral rupture. Ultrasound biomicroscopy (UBM) and in vivo confocal microscopy (IVCM) were used to diagnose this rare condition. A lamellar corneoscleral graft was performed with histopathological examination of the excised cyst. The treatment of corneoscleral epithelial cysts is discussed.

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Résumé Nous décrivons le cas d’un patient qui présentait un kyste épithélial cornéoscléral provenant d’une rupture sclérale d’origine traumatique. Le diagnostic a été réalisé grâce à une analyse en UBM et en microscopie confocale in vivo. Le patient a bénéficié d’une greffe lamellaire cornéosclérale avec un examen anatomopathologique de la paroi du kyste. Les traitements des kystes cornéoscléraux sont discutés.

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Corneoscleral cyst is a rare clinical condition that can occur in the corneal or scleral stroma, and irido-corneal angle tissues [1]. Although developmental and traumatic etiologies have been postulated, the exact pathophysiology of corneoscleral cysts remains unclear [1,2]. Several surgical procedures have been reported but recurrences within a short time after surgery are frequent [3–5].

We describe a case of a patient with a corneoscleral cyst originating from a traumatic scleral rupture. Complete excision of the cyst and a corneoscleral lamellar graft were performed. The diagnosis of an epithelial lined cyst was confirmed by histological examination. There was no recurrence after 3 years of follow-up.

Case report

A 24-year-old man presented to Beijing TongRen Eye Center complaining of painless decreased vision in his left eye for more than 20 years. His history revealed a possible fireworks trauma just before the onset of symptoms. On ocular examination, best-corrected visual acuity was limited to counting fingers in his left eye and 20/20 in the right eye. Intraocular pressure was 13 mmHg and 9 mmHg in the right and left eye, respectively. Slit lamp examination showed a subconjunctival cyst measuring 12 × 8 × 5 mm and extending into the anterior stroma of the inferior cornea of the left eye (Fig. 1). The cyst was renitent and immobile. The central and lower corneal regions were edematous and thickened (approximately 1.5 times the normal corneal thickness). A white infiltration was observed in the superior and inferior peripheral area of the corneal cyst. Although difficult because of the corneal cyst, fundus examination of the left eye was unremarkable. Slit-lamp examination of the right eye was normal.

To further determine the relationship between the subconjunctival cyst, the cornea and the anterior chamber, a cyst puncture was performed under topical anesthesia for cytology examination. Smears of cyst fluid showed many degenerated epithelial cells, lytic goblet cells, several white blood cells and lymphocytes (Fig. 1). Ultrasound biomicroscopy (UBM) showed that the episcleral cyst was connected to the intracorneal cyst in the nasal region (Fig. 2) and revealed an inferonasal scleral rupture, 3 mm from the limbus, resulting in a communication between the cyst and the vitreous cavity (Figs. 2 and 3). In vivo confocal microscopy (Heidelberg Retina Tomograph Rostock Cornea Module, Heidelberg, Germany) showed epithelioid cells in the anterior and posterior wall of the corneal cyst (Fig. 4).

Under general anesthesia, a complete excision of the corneoscleral cyst followed by irrigation of the wound area with 95% ethanol was performed. This procedure was associated to a lamellar corneoscleral allograft covering the scleral perforation and the corneal cyst. Histopathological examination of the excised cyst wall showed a fibrous tissue lined by squamous epithelial cells (Fig. 5). Two weeks after surgery, corneal edema decreased, visual acuity of the

Figure 1. A. Anterior segment photograph of the right eye showing a large corneoscleral cyst. B. Central and inferior corneal edema. C. Cytology analysis of the cyst fluid showing many degenerated epithelial cells and lytic goblet cells (× 1000). D. Corneal cyst.
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Figure 2. A. UBM image showing the corneal cyst connecting with the episcleral cyst in the temporal limbus region. B. UBM image showing an inferonasal scleral rupture creating a passage between the episcleral cyst and the vitreous cavity.

Figure 3. UBM and slit-lamp photograph composite image showing the entire cyst, from the scleral rupture (green arrow) to the corneal cyst.

left eye was 0.08 and IOP was 19 mmHg. After 3 years of follow-up, there was no recurrence of the corneoscleral cyst (Fig. 6).

Discussion

Corneoscleral cysts were first described by Appia in 1853 and a series of eight cases was presented by Reed and Dohlman in 1971 [1]. The majority of reported cases were congenital and occurred in children. Corneoscleral cysts may also have a traumatic origin and may occur after cataract surgery with corneal incision, lamellar keratoplasty or strabismus surgery [6]. In all cases, corneal cysts are resulting from the migration of epithelial and goblet cells into the corneal stroma [2].

Sclera is an avascular fibrous tissue with few inactive fibroblasts resulting in a slow wound healing process that provides the conditions for intrascleral cyst formation. Traumatic scleral inclusion cysts are usually located in the anterior sclera near the limbus, and then extend into the cornea forming corneoscleral cysts. The anterior limit of the cyst usually lies into the cornea, whereas the posterior limit is diffusing. UBM is a useful tool to rule out differential diagnosis and, in the present case, showed the scleral rupture as well as the distribution of the cystic space. Typical UBM images of corneoscleral cyst are well-circumscribed structures with a hyperechogenic wall and low internal reflectivity [7]. In addition to UBM examination, in vivo confocal microscopy may be useful to analyze cell populations within the cyst wall.

To avoid recurrences of the corneoscleral cyst, several surgical procedures including excision of the cyst wall, chemical cautery using iodine or ethanol have been proposed [5,8]. Cyst excision associated to peripheral lamellar keratoplasty may also represent an effective treatment [9]. Considering the location of the scleral rupture in our case (3 mm from the limbus) and the corneal cyst size, we performed a corneoscleral lamellar allograft after excision of the cyst wall. During the surgical procedure, surgeons should carefully remove the cyst wall in order to eliminate residual epithelial cells that may be responsible for recurrences. For that purpose, the cyst area was also irrigated repeatedly with 95% ethanol. After a long follow-up, no recurrence was observed in the presented case.

The treatment of corneoscleral cyst remains difficult in order to avoid recurrences and to preserve corneal transparency. In case of large cysts, the use of a corneoscleral lamellar keratoplasty associated with ethanol irrigation might be an interesting surgical option.
Epithelioid cells were observed in the posterior wall (depth 477 μm) of the intracorneal cyst using in vivo confocal microscopy (400 × 400 μm, Heidelberg Retinal Tomograph Rostock Cornea Module, Heidelberg, Germany).

Histology section of the corneoscleral cyst. The cyst wall consisted of a fibrous tissue lined with squamous epithelial cells (hematoxylin-eosin stain × 200).

Disclosure of interest

The authors declare that they have no conflict of interest concerning this article.

References