Surgical excision and distilled water lysis of a congenital corneal intrastromal cyst

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ARTICLE INFO
Keywords: Intrastromal Corneal Cyst

ABSTRACT
Purpose: To describe imaging findings and encourage the use of distilled water as an adjuvant osmolytic in the surgical management of corneal intrastromal cysts.

Observations: A five-year-old female with no history of ocular trauma presented with a visually significant corneal opacity of the left eye. She was diagnosed with a presumed corneal intrastromal cyst and underwent surgical excision with distilled water osmolysis of the cyst cavity.

Conclusions and Importance: Optical coherence tomography (OCT) can confirm diagnosis of intrastromal cysts. Presumed epithelial cell nests remain visible at post-operative month eight, with no evidence for cyst recurrence. The authors propose that OCT findings are pathognomonic for corneal intrastromal cysts and that cyst excision combined with distilled water osmolysis at the time of debridement may be beneficial in conserving tissue integrity.

1. Introduction
Intrastromal corneal cysts of epithelial origin are rare, non-inflammatory lesions which tend to be slowly progressive and classically occur after penetrating traumatic injury in which epithelial cells are displaced into the corneal stroma.1-4 A congenital form, arising without trauma, can occur spontaneously or after a scleral cyst extends into the corneal stroma.5-6 Surgical intervention is indicated for those lesions which threaten the visual axis or create significant astigmatism.3 A variety of approaches have been described, although no consensus has been reached on the appropriate surgical management.2-9 Some patients require multiple surgical interventions before resolution.1,2 Intra-operative destruction of the epithelial cells lining these cysts is considered imperative to avoiding their recurrence. Previously suggested irrigation solutions include 10% acetic acid, 1% iodine, cocaine, 5-fluorouracil, 20% trichloroacetic acid, 96% ethanol, distilled water, and balanced salt solution.2,7,10 The authors present a case of an intrastromal cyst successfully treated with surgical drainage, curettage, and distilled water irrigation.

2. Case report
2.1. History
The patient was a 5-year-old female with normal development and no significant past medical history who presented for second opinion of a left corneal opacity which was noted in the first year of life and gradually enlarged. There was no reported history of trauma. An outside cornea specialist diagnosed Salzmann’s nodular degeneration and recommended superficial keratectomy. She was poorly compliant with patching treatment for amblyopia.

2.2. Examination
Spectacle correction for hyperopic astigmatism of both eyes (OD: +0.50 + 0.50 × 080, OS: +1.00 + 2.00 × 108) allowed for a corrected visual acuity of 20/20 (OD) and 20/60 (OS). Pre-operative photographs, Scheimpflug tomography, and anterior segment optical coherence tomography (OCT) can be seen in Fig. 1. Scheimpflug measured 14.3 diopters of anterior corneal astigmatism.
2.3. Surgical management

Her family elected to pursue drainage, curettage, and distilled water rinse of intrastromal corneal cyst. Under general anesthesia, a crescent blade was used to make an entry into the cyst at the nasal limbus with drainage of milky fluid. There was no apparent scleral involvement of the cyst. The lining of the cyst was scraped thoroughly using a Terry Descemet’s scraper and sterile water was flushed repeatedly inside the cavity. Material recovered from the cyst lining was insufficient for pathology. The incision was sutured to prevent epithelial ingrowth.

2.4. Follow-up

At the patient’s one-week follow-up appointment, sutures were removed and neomycin/polymyxin/dexamethasone ointment was stopped. Spectacle-corrected acuity at month two improved to 20/30, from 20/60 pre-operatively. Tomographic astigmatism improved from 14.3D to 4.0D (Figs. 1 and 2). Although she maintained stromal haze in the region of the former cyst, there was no evidence for cyst recurrence at month eight (Fig. 2). Nests of presumed intra-stromal epithelial cells were visible in the region of the former cyst. At fourteen months post-procedure, there was again no evidence for cyst recurrence and she achieved best corrected 20/20 vision in each eye (OD: +1.00 + 0.25 x 090, OS: +0.25 + 3.25 x 112).

3. Discussion

Congenital intrastromal corneal cysts have mostly been described with scleral involvement, but cases without scleral involvement have been reported, as seen in our patient. Histopathology of the cyst wall lining typically demonstrates non-keratinized, stratified squamous epithelium with goblet cells which proliferate slowly. Intrastromal corneal cysts characteristically lack inflammation, enlarge over time, demonstrate a “pseudo-hypopyon” of white proteinaceous material at the dependent side of the cyst, and induce high amounts of astigmatism. The lesions can be confused clinically with Salzmann’s nodules, corneal keloids, or dermoid cysts. OCT of the lesion demonstrates a pathognomonic finding of space-occupying intrastromal lesion which is well-demarcated from surrounding tissues and highly uniform in reflectivity, lacking the speckled pattern of corneal stroma.

Because of the high risk for recurrence with drainage alone, surgical approaches focus on eradication of epithelial cell wall components through scraping of the cyst lining, irrigation with cytolysing solutions, cautery, or direct excision. Compared with lamellar or penetrating keratoplasty, drainage with cytolysis is preferred due to its simplicity, shorter recovery, and decreased risk of amblyopia. Keratoplasty should be reserved for severe central scarring or if anatomy prohibits a less-invasive procedure.

The use of distilled water for lysis of epithelial cells within corneal cysts has been described for its value in minimizing postoperative opacity and corneal tissue damage. Ethanol may be more effective in...
lysing epithelial cells, but does carry a theoretical risk of toxicity to adjacent tissues,\(^1\) which was our justification for preferring distilled water. Our patient demonstrates small epithelial nests without recurrence at 14 months. Recurrence of these cysts more than one year postoperatively have been noted and our patient is being monitored regularly.\(^2\)

4. Conclusions

In summary, intrastromal cysts can be congenital or sequelae of corneal trauma. They can impose a threat to the visual axis through either opacification or astigmatism induction. The authors support the use of a cytolytic agent, such as distilled water or ethanol, to reduce recurrence. Amblyopia management and long-term monitoring for recurrence are essential in caring for these patients.

Patient consent

The patient’s legal guardian consented to the publication of the case in writing and orally.

Funding

No funding or grand support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no conflicts of interests.

Acknowledgements

None.

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Fig. 2. Post-operative assessment. Eight-month post-operative slit lamp exam, Scheimpflug tomography, and anterior segment OCT of the left eye, demonstrating resolution of the cystic fluid with remaining nasal sub-epithelial haze. Tomography demonstrates 4 diopters of residual astigmatism.