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Case Report

Delayed Perforation of an Intrastromal Corneal Ring Segment into the Anterior Chamber: A Case Report and Review of the Literature

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Keywords
Intrastromal corneal ring segments · Keratoconus · Corneal ectasia · Intacs · Corneal perforation · ICRS

Abstract
Intrastromal corneal ring segments (ICRSs) are an effective treatment for stabilizing and normalizing corneal shape in patients with keratoconus and other corneal ectasias. Intraoperative segment perforation through the corneal endothelium into the anterior chamber (AC) is an uncommon but known complication. However, perforation into the AC postoperatively is an exceedingly rare complication with only 3 reported cases in the literature. One case was due to Descemet membrane detachment and another due to ocular trauma. In the third case, the mechanism for perforation was unclear. We present the fourth case of delayed ICRS perforation due to silent migration through the endothelium into the AC. We also present all reported cases in the literature of intraoperative and postoperative perforation into the AC.

Introduction

Intrastromal corneal ring segments (ICRSs) are thin plastic rings inserted into the cornea to stabilize the cornea in patients with corneal ectasias, including keratoconus [1]. In keratoconus, ICRSs are inserted after nonsurgical treatments such as spectacles or contact lenses have proven insufficient or inviable [2]. Additionally, ICRS insertion has been shown to delay

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and, in some cases, prevent the need for corneal transplant [3]. Although the insertion of ICRSs to treat keratoconus has yielded favorable postoperative outcomes, perforation of the ring segment into the anterior chamber (AC) is possible [4]. Usually, perforation occurs intraoperatively and is believed to be due to excessive force or inaccurate depth estimation [5]. Delayed or late perforation is much less common, with only 3 reported cases in the literature [6–8]. We report a delayed intrusion of an inferior corneal ring segment into the AC within 1 week after surgery in a patient with keratoconus.

Case Report

A 35-year-old woman with Amsler-Krumeich stage II keratoconus and previous bilateral cross-linking presented to the clinic for worsening visual acuity in her right eye. After cross-linking, best-corrected visual acuity (BCVA) was 20/30 OD and 20/20 OS. Two years later, BCVA had worsened to 20/40 OD, and she began complaining of worsening blurry vision. Due to worsening visual acuity, the patient elected for Intacs (Addition Technology, Inc, Sunnyvale, CA, USA) insertion in order to delay potential transplant. The medical history was significant for asthma controlled on a long-acting beta-2-agonist and inhaled corticosteroid. Uncorrected distance visual acuity (UDVA) was 20/300+ in the right eye and 20/70 + 2 in the left eye, respectively; BCVA was 20/40-2 OD, 20/15- OS. Manifest refraction was −1.25 to 2.00 × 067 OD and −1.00 to 1.75 × 121 OS. The keratometric reading obtained with Pentacam HR (Oculus, Wetzlar, Germany) was 45.7 D at 51°/51.4 D at 141° with the thinnest part of the cornea measuring 439 μm in the right eye and 42.8 D at 127°/44.6 D at 217° with the thinnest part of the cornea measuring 488 μm in the left eye, respectively.

On the day of surgery, the axis of the steep meridian was identified at 145°. The site was then measured 4 times intraoperatively with ultrasound pachymetry. The corneal thickness was measured to be an average of 604 μm at the planned incision site, and the 70% depth was calculated to be 422 μm. The guarded diamond blade was set to the appropriate depth, and an initial incision was made. A stromal separator was then used to create a tunnel within the cornea mechanically. Per the surgeon's personal nomogram, a 0.45-mm segment was positioned inferiorly, and a 0.25-mm segment was placed superiorly. The incision was closed with a 10-0 nylon suture. After surgery, ofloxacin 0.5% and prednisolone 1% drops were prescribed.

Both ring segments were in place on postoperative day one. On her 1-week follow-up, the patient returned to the clinic with no complaints. However, UDVA had declined to 20/400 OD. On slit-lamp examination (SLE), the inferior segment had dislocated into the AC, while the superior segment had remained in place. Interestingly, SLE revealed a quiet AC with no obvious corneal edema or endothelial contusion (Fig. 1). Upon further questioning, she denied preceding ocular trauma or eye rubbing. She also wore an eye shield at night. Subsequently, the inferior segment was explanted. UDVA on the first day after explantation was 20/300 OD, and SLE revealed trace inferior stromal edema and AC cells. The patient was treated with prednisolone acetate 1% and ofloxacin 0.3% drops 4 times a day. On postoperative week one, after explantation, significant improvement of stromal edema was noted. On week 3, stromal edema and inflammation had resolved. BCVA improved to 20/70 OD. Keratometric readings were 46.6 D at 28°/50.3 D at 118° and the thinnest aspect of the cornea measured 448 μm in the right eye. An anterior segment optical coherence tomography (OCT) revealed the inferior tunnel pocket at a depth of 468 μm, which was 90% of the corneal thickness and 46 μm deeper than intended. There were no signs of Descemet membrane folds or detachment. The superior stromal ring was in place at a depth of 535 μm, which was 84% of the corneal thickness and 113 μm deeper than intended (Fig. 2). On a follow-up visit 3 weeks later, the superior segment

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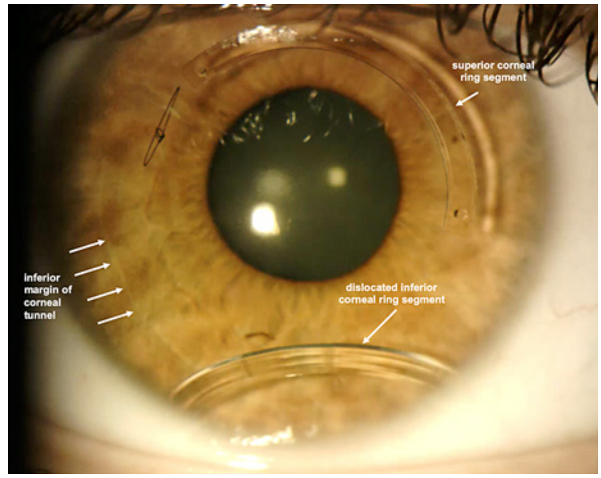

Fig. 1. On postoperative day 1, slit-lamp photograph demonstrating a dislocated inferior corneal ring segment in the AC. The inferior margin of the corneal tunnel is outlined with 4 arrows. No corneal edema, AC cell, or other signs of inflammation were noted. The superior corneal ring segment is in place without signs of migration or extrusion. AC, anterior chamber.

Fig. 2. OCT of the cornea OD. On the left, the tunnel of the inferior segment is still visible, despite the absence of the segment. There is no evidence of Descemet folds, detachment, or corneal edema. On the right, the superior segment is in place at a depth of 90%. OCT, optical coherence tomography.

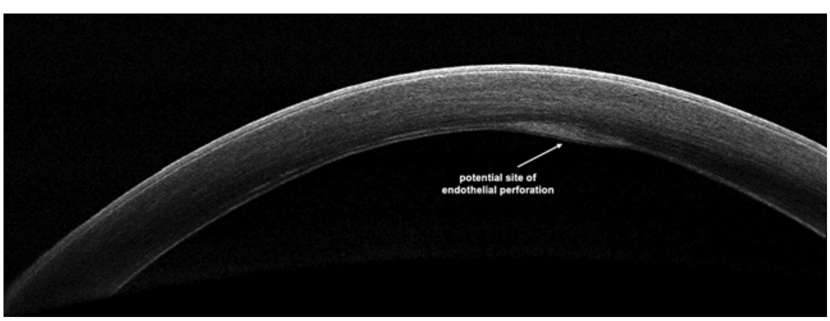

Fig. 3. OCT of the collapsed tunnel of the displaced inferior segment. The site of the potential perforation is marked. OCT, optical coherence tomography.

had remained in place on OCT. Additionally, the potential site of perforation was noted (Fig. 3) corresponding to an area with a corneal thickness of approximately 490 microns. Keratometry had improved to near preoperative values (Fig. 4). Due to concern for endothelial cell damage, cell counts were obtained and reported 2,924 cells/mm^3 in the right eye and 3,175 cells/mm^3 in the left. Whether the patient will elect for observation, scleral contact lens fitting, reimplantation of ICRS at a more superficial depth, or more invasive surgery such as corneal transplant has yet to be determined.

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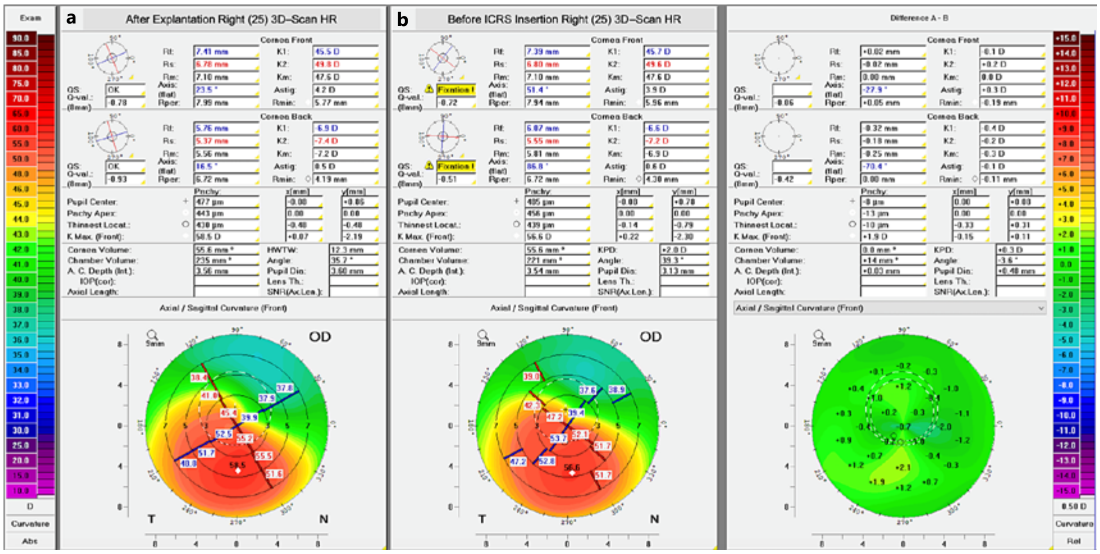

Fig. 4. Difference maps of corneal topography. **a** Keratometry after the explantation of the inferior segment. **b** Keratometry prior to inferior segment insertion.

Literature Review

Methods

PubMed, Google Scholar, Embase, Asian Digital Library, and Scopus were searched from January 1997 to September 2020. The search terms were *intrastromal corneal ring segment*, *ICRS*, *Ferrara ring*, *Keraring*, *Intacs*, *perforation*, *intrusion*, *extrusion*, and *dislocation*. Only publications that described perforation into the AC were included in this review. ICRS perforations were reported in 17 publications consisting of 5 retrospective case series, 5 prospective case series, 3 clinical trials, and 4 case reports. The articles were written mainly in English; however, one Portuguese publication was also included.

Discussion

Before our case presentation, ICRS perforation through the endothelium into the AC had only been reported in 30 eyes [5–21]. A summary of each publication can be found in Table 1. ICRS implantation was mainly indicated for keratoconus, occurring in 25 of 30 eyes [8, 9, 11, 13–21]. Most of the perforations into the AC, 26 of 30 eyes (87%), occurred intraoperatively. One case did not specify the timing of perforation [18]. Only 3 cases reported delayed ICRS perforation through the endothelium into the AC, all of which differ significantly from our report. Ghajarnia et al. [6] described a case of postoperative intrusion due to a microperforation of Descemet membrane during surgery that led to the progressive detachment of the membrane over 1 week. The detachment presented as severe corneal edema, and both segments had to be explanted. Park et al. [8] reported a case of perforation through the endothelium into the AC 3 weeks after insertion. The perforation was thought to be due to the silent migration of the superior segment into the AC due to eyelid blinking or perhaps eye rubbing. However, the cause could not be confirmed as OCT was not performed. The third case of perforation through the endothelium into the AC occurred 7 years after insertion and was attributed to aggressive eye rubbing. Interestingly, the segment had also extruded through the corneal epithelium creating a fistula requiring emergent surgical intervention [7].

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Table 1. Summary of existing literature of ICRS perforation into AC

Author	Reason for implant	Eyes with ICRS perforation, n (%)	Ring type	Ring thickness, µm	Minimum corneal thickness, µm	BCVA	Surgical technique	Surgery to perforation interval	Reason for perforation
Schanzlin et al. [5]	Myopia	1 of 102 (0.98)	Intacs	250–450	–	20/20	Manual	Intraoperative	Incorrect knife setting
Ruckhofer et al. [10]	Myopia	1 of 163 (0.61)	Intacs	250–450	–	–	Manual	Intraoperative	Incorrect knife setting
Schanzlin et al. [12]	Myopia	2 of 454 (0.44)	Intacs	250–350	–	20/20	Manual	Intraoperative	Incorrect knife setting
Moreira et al. [13]	Keratoconus	2 of 10 (20)	Ferrara	200–350	–	20/112	Manual	Intraoperative	–
Kanellopoulos et al. [16]	Keratoconus	1 of 20 (5)	Intacs	250–450	>300	20/37	Manual	Intraoperative	Blunt dissection
Ghajarnia et al. [6]	PMD	1 of 1 (100)	Intacs	350	398	20/80	Femtosecond laser	1 week	Descemet membrane detachment
Piñero et al. [11]	Keratoconus	1 of 146 (0.68)	–	250–450	–	20/66	Manual	Intraoperative	–
Coskunseven et al. [15]	Keratoconus	5 of 850 (0.59)	Keraring	150–300	>350	–	Femtosecond laser	Intraoperative	Inaccurate depth estimation or deeper channel creation
Park et al. [8]	Keratoconus	1 of 1 (100)	Intacs	250	467	20/40	Manual	3 weeks	–
Ferrer et al. [18]	–	1 of 58 (1.72)	Intacs	–	–	–	Femtosecond laser	2 weeks*	–
Al-Amry et al. [9]	Keratoconus	1 of 2 (50)	Intacs	–	–	20/50	–	Intraoperative	–
Hamdi et al. [17]	Keratoconus	3 of 100 (3)	Ferrara	150–350	–	20/37	Manual	Intraoperative	Inaccurate depth estimation
Rho et al. [19]	Keratoconus	1 of 51 (1.96)	Intacs	–	441.6	20/30-20/300	Femtosecond laser	Intraoperative	Inaccurate depth estimation
Moshirfar et al. [7]	LASIK ectasia	1 of 1 (100)	Intacs	–	1062	20/500	–	7 years	Blunt trauma
Torquetti et al. [14]	Keratoconus	2 of 45 (4.44)	Ferrara	150–300	430	20/100	Manual	Intraoperative	–
Mounir et al. [21]	Keratoconus	1 of 623 (0.16)	Keraring	150–300	>350	20/123	Femtosecond laser	Intraoperative	Excessive force during insertion
Monteiro et al. [20]	Keratoconus	4 of 265 (1.51)	Keraring	150–300	454.5	20/54	Manual	Intraoperative	–
Monteiro et al. [20]	Keratoconus	1 of 111 (0.9)	Keraring	150–300	460.8	20/62	Femtosecond laser	Intraoperative	–

ICRS, intrastromal corneal ring segment; D, diopters; BCVA, best-corrected visual acuity; Intacs (Addition Technology, Sunnyvale, CA, USA); Ferrara ring (Mediphacos, Belo Horizonte, Brazil); Keraring (Mediphacos, Belo Horizonte, Brazil); PMD, pellucid marginal degeneration; LASIK, laser-assisted in situ keratomileusis; AC, anterior chamber.
*Ferrer et al. [18] reported time to explantation but did not record when the perforation had occurred.

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To prevent complications such as perforation, investigators have focused on understanding the risks associated with the technique chosen to create the corneal tunnel for ICRS insertion. Currently, there are 2 options for tunnel creation: mechanical/manual tunneling or through the use of the femtosecond laser [20]. Perforations into the AC have been reported in both manual (60%) and femtosecond laser tunneling techniques (33%), while 7 percent of cases reported in the literature did not specify the tunneling method [5–21]. The manual technique involves making an initial incision at a predetermined corneal depth, usually 70%. A stromal separator is then inserted to manually dissect the corneal tissue creating a pocket at the intended depth. It is assumed that the manual dissection creates the tunnel at a proportional depth throughout the cornea as it separates lamellar tissue.

In contrast, the femtosecond laser method creates a tunnel at a uniform depth as measured from the corneal epithelium. Thus, when using the femtosecond laser, depth must be set at 70–80% as measured from the thinnest part of the cornea [22]. As corneal thickness is irregular in keratoconus, ICRS insertion may often be shallower than intended with the femtosecond laser. Additionally, the charge per utilization of the laser is cost-prohibitive in many areas of the world. However, femtosecond laser is the preferred method of tunnel creation by many surgeons as several studies suggest the technique has fewer complications of infection, channel opacification, epithelial plugs, neovascularization, chronic pain, segment migration, extrusion through the corneal epithelium, and perforation through the endothelium into the AC [18, 20, 23, 24].

Often recognized intraoperatively, signs of AC perforation include observing bubbles in the AC and decreased chamber depth [13, 15]. Causes of intraoperative perforation into the AC may include forceful manipulation of tissue during insertion, incorrect depth settings, and inaccurate pachymetry at the site of insertion [5, 10, 17, 25]. Even with correct measurements and accurate calculations, ICRS may not be placed at the desired depth of 70–80% of the corneal thickness [3]. Studies by Barbara et al. [26], de Sanctis et al. [27], and Naftali et al. [28] have reported that the actual depth of ICRS are, on average, 90–153 μm more superficial than the intended depth. Interestingly, Barbara et al. [26] observed that all 30 segments implanted were more superficial than expected. Kouassi et al. [29] observed that superficial ring placement could be observed, regardless of the tunneling technique. Manual and femtosecond laser techniques placed segments more superficially than intended by 76 μm and 86 μm, respectively. Since superficial ring placement is a major risk factor for extrusion, these studies could explain, in part, the much higher rates of ICRS extrusion through the corneal epithelium (0.5–3%) as compared to perforation through the endothelium [17, 22, 30, 31]. However, in our case report, OCT demonstrated the inferior and superior segments had been placed deeper than expected, not shallower. In the literature, only one study has suggested that ICRS can be inserted deeper than planned, and it was still significantly less common than superficial placement. Monteiro et al. [32] recently reported that manual and femtosecond laser tunneling could lead to ring placement deeper than intended in 27.6% and 9.44% of eyes, respectively. When segments are placed at increasing stromal depths, the risk of perforation into the AC also increases. Theoretically, these results could explain intraoperative perforations into the AC, despite accurate pachymetry and depth calculations. Additionally, if a microperforation without signs of endothelial penetration were to occur intraoperatively, segment perforation into the AC might present days to weeks later.

It should be noted that ICRS thickness selection may have had an impact on this patient's clinical course. According to the Addition Technology nomogram for selection of segment thickness, this patient's recommended surgical plan involved the placement of a 0.35-mm segment inferiorly and a 0.21-mm segment superiorly. Based on the surgeon's personal nomogram, a 0.45-mm segment was positioned inferiorly, and a 0.25-mm segment was placed superiorly. It is possible that the presumed excessive thickness of the inferior segment contributed to the eventual intrusion, though we believe that a silent microperforation occurred distal and inferior to the initial incision intraoperatively and was ultimately due to deeper than intended ICRS placement. We postulate that the initial hole in the endothelium was small enough to prevent the segment from perforating into the AC during surgery. As the patient was supine immediately after surgery, the segment was affected minimally by gravity and mechanical stress, potentially explaining the desired position within the corneal stroma when examined on postoperative day one. However, in the following week, the repeated mechanical stress of blinking and activities of daily living could have been sufficient to cause the segment to migrate into the AC silently.

Due to the significant time between implantation and perforation (7 years) as well as the traumatic cause (eye rubbing), we suggest that the case presented by Moshirfar et al. [7] be classified as a late perforation, instead of delayed. All 3 cases of delayed perforation likely were caused by a microperforation intraoperatively that was not clinically apparent until one to 3 weeks after surgery. Unlike the case reported by Ghajarnia et al. [6], our patient had no visual complaints, no signs of Descemet membrane detachment, and no corneal edema. While Park et al. [8] proposed a similar mechanism for delayed perforation of an ICRS, without an OCT, they were unable to confirm depth. Thus, to the best of our knowledge, we may be the first to present a confirmed case of delayed silent microperforation into the AC. Additionally, we propose microperforation as the predominant cause of delayed perforation of ICRS through the endothelium into the AC.

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Statement of Ethics

Written informed consent was obtained from the patient for publication of the case report as well as all accompanying images. This case report was reviewed and approved by the Hoopes Vision Ethics Board on September 4, 2020. The study complies with the guidelines for human studies and in accordance with the World Medical Association Declaration of Helsinki.

Conflict of Interest Statement

None of the authors have any conflict of interest related to this work.

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Author Contributions

Majid Moshirfar: conceptualization, methodology, writing – original draft, and supervision. Dallin Milner: conceptualization, methodology, writing – original draft, project administration, resources, and investigation. Tanisha Martheswaran: investigation, resources, and writing – original draft. Shannon McCabe: investigation, resources, and writing – original draft. Yasmyne Ronquillo: conceptualization, writing – original draft, and editing. Phillip Hoopes: supervision and funding acquisition.

Data Availability Statement

There is no dataset required for this case report.

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