

Bimanual irrigation-aspiration for ectopia lentis and use of a small incision for 4-point scleral-sutured foldable intraocular lens and anterior vitrectomy in patients with Marfan syndrome

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Purpose: This study evaluated bimanual intracapsular irrigation-aspiration for ectopia lentis with use of a small incision for 4-point scleral fixation of a foldable posterior-chamber intraocular lens (IOL) and anterior vitrectomy in patients with Marfan syndrome. **Methods:** We performed a retrospective study of 18 eyes from 10 patients with Marfan syndrome who underwent surgical intervention for ectopia lentis at our clinic between July 2012 and September 2018. In this study, intraoperative and postoperative complications, uncorrected visual acuity, best-corrected visual acuity, spherical equivalent, intraocular pressure, and endothelial cell density were evaluated. **Results:** No intraoperative complications were reported. In all cases, early postoperative evaluation revealed a clear cornea, round pupil, and well-centered IOL. Mean logMAR uncorrected visual acuity improved from 1.09 preoperatively to 0.56 postoperatively ($P < 0.05$). Mean logMAR best-corrected visual acuity improved from 0.45 preoperatively to 0.17 postoperatively ($P < 0.05$). Aside from transient ocular hypertension, no postoperative complications were reported. **Conclusion:** The combined surgical technique presented above yields excellent visual outcomes with an extremely low incidence of complications. This approach is simple, safe, and effective in the treatment of ectopia lentis in patients with Marfan syndrome.

Key words: Anterior vitrectomy, ectopia lentis aspiration, foldable 4-point scleral fixation IOL, Marfan syndrome, small incision

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Marfan syndrome, which was first reported by Antoine Marfan in 1896,^[1] is also known as spider-like toe syndrome or congenital mesoderm dysplasia. The condition is an autosomal dominant disorder of connective tissue that affects many body systems, including the skeleton, eyes, and cardiovascular system. Marfan syndrome is caused by heterozygous mutations in fibrillin-1 (FBN1) gene located on chromosome 15q that affects synthesis of FBN1, an important protein in connective tissue.^[2] The clinical phenotype of this disease varies greatly among individuals.^[3] The incidence of Marfan syndrome in the general population is 1 in 5,000–10,000,^[4] and there is no sex or ethnic predilection.^[5]

Marfan syndrome is the most common congenital cause of ectopia lentis, occurring in 80%–90% of affected individuals. The condition typically develops between birth and 30 years of age.^[6,7] Subluxation of the lens is typically bilateral, symmetric, and superotemporal. In patients with Marfan syndrome, the zonular fibers are few and often attenuated and broken.^[6] Axial myopia, myopic degeneration of the retina, retinal detachment, and megalocornea may also occur.^[8–10]

Indications for surgery include uncorrectable poor visual acuity (VA), excessive induced astigmatism, dislocation

of the lens to the anterior chamber, and complications of lens subluxation, including secondary glaucoma, uveitis, corneal endothelial dysfunction, and monocular diplopia.^[11] To manage lens subluxation, most surgeons perform intracapsular cataract extraction (ICCE) or pars plana lensectomy (PPL) combined with 2-point transscleral fixation of a large polymethyl methacrylate (PMMA) intraocular lens (IOL), which requires a larger incision. Use of a larger corneal incision may result in significant surgically induced astigmatism, intraoperative hypotony, and detachment of the retina, and 2-point transscleral IOL fixation had a higher incidence of IOL dislocation.^[12]

Here we describe a novel combined surgical technique in which the dislocated lens is removed by bimanual intracapsular irrigation/aspiration. A foldable hydrophilic acrylic Akreos IOL (Bausch and Lomb) was inserted through a small (2.85-mm) incision to facilitate 4-point scleral fixation with no tilt and effective centration. Anterior vitrectomy and peripheral iridectomy were also performed in order to decrease risk for the complications listed above.

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Methods

Eighteen eyes from 10 patients (5 males and 5 females) who had undergone surgery for ectopia lentis associated with Marfan syndrome at our institution during the period from July 2012 to September 2018 were retrospectively included in the present study. The mean age was 14.2 ± 7.7 years (range, 5–33 years). The study was approved by the Medical Ethics Committee of our hospital and adhered to the tenets of the Declaration of Helsinki. All patients provided their informed consent.

All treated eyes were examined preoperatively to determine uncorrected visual acuity (UCVA), best-corrected visual acuity (BCVA) ("E" chart), and spherical equivalent (SE) [Table 1]. Slit-lamp microscopy was performed after dilation of the pupil to determine the direction and amount of decentration. Intraocular pressure (IOP) was measured by non-contact tonometry (Canon, Tokyo, Japan). Endothelial cell density (ECD) was measured using a Specular Microscope (SP-3000P Topcon, Tokyo, Japan). The fundus was examined with direct ophthalmoscopy and B-scan ultrasonography. Axial length was measured using the IOL Master (Carl Zeiss Meditec Co Ltd). IOL power was calculated using the SRK/T formula. A one-piece hydrophilic acrylic-posterior chamber Akreos IOL was implanted into the eye using the corresponding injector system.

Surgical technique

All surgeries were performed by a single fully qualified and highly experienced surgeon (Z.S.B.). General anesthesia was used in pediatric patients; local anesthesia was used for adult patients. Pupils were dilated perioperatively with 0.5% (wt/vol) tropicamide.

Conjunctival peritomy was performed: one extended from the 2 o'clock to the 4 o'clock position; the other extended from

the 10 o'clock to 8 o'clock positions. A 2.85-mm superior corneal incision was made and corneal side ports were created using a 15° blade at the 2 o'clock and 10 o'clock positions [Video 1].

After the anterior chamber was injected with viscoelastic material Sodium Hyaluronate, continuous circular capsulorhexis [Fig. 1a] was performed on the anterior lens capsule [Video 1]. The lens was removed using bimanual intracapsular irrigation/aspiration (I/A) under low-bottle height with 60 cm H₂O and a low-vacuum setting with 100 mmHg [Fig. 1b]. The lens was completely removed, and the capsular bag could be seen clearly [Fig. 1c]. The empty capsular bag was removed with microforceps [Fig. 1d and Video 2].

A one-piece foldable hydrophilic acrylic Akreos IOL with 4 hollow haptics was implanted through the main superior incision and positioned in front of the iris [Fig. 2a and Video 2]. The Akreos IOL has a 6.0-mm optic zone and overall length of 11.0 mm.

A 10-0, double-armed polypropylene suture (Alcon, Dallas–Ft. Worth, TX, USA) was affixed at end with a straight needle. One of the straight needles was inserted through the inferonasal sclera (approximately 1.5 mm posterior to the limbus, between the 8 and 9 o'clock positions), then passed through one hollow haptic of the Akreos IOL [Fig. 2b]. A bent 27G needle was inserted through the opposite inferotemporal sclera (1.5 mm posterior to the limbus, between the 3 and 4 o'clock positions), then passed through the hollow haptic of the IOL on the opposite side of the cornea [Fig. 2c]. The 27G needle was then used to guide the straight needle to the appropriate exit point [Fig. 2d]. The other straight needle was similarly passed through the other 2 hollow haptics of the Akreos IOL [Fig. 2e and f, Video 3]. A Sinsky hook was used to pull the sutures out via the main corneal incision [Fig. 3a]. Sutures were then cut in half with

Table 1: Descriptive characteristics of eyes included in the study

Pt	Gender/ Age	OD/ OS	AXL (mm)	Preop UCVA/ BCVA (logMAR)	Postop UCVA/ BCVA (logMAR)	Preop/Postop IOP (mmHg)	Preop/ SE (D)	Postop/ SE (D)	FU (Mo)
1	M/16	OD	26.5	0.5/0.3	0.4/0.1	12/12	6.38	-3.75	70
2	M/16	OS	27.6	0.5/0.3	0.4/0.1	16/16	4.75	-4.63	70
3	F/10	OS	26.9	1.7/0.9	0.8/0.0	11/16	-6.0	-5.25	73
4	F/11	OD	28.2	2.0/0.7	0.5/0.4	15/19	-18.0	-2.75	73
5	F/11	OS	29.8	1.7/0.7	0.7/0.4	16/19	-17.88	-6.13	73
6	M/20	OD	28.2	0.5/0.2	0.2/0.2	16/13	4.75	1.38	70
7	M/20	OS	27.2	0.5/0.2	0.4/0.2	18/18	5.88	-1.88	70
8	M/11	OD	26.9	1.4/0.7	1.0/0.3	12/10	-14.5	-3.0	73
9	M/6	OD	23.6	1.1/0.6	0.2/0.1	10/10	-7.0	-0.9	33
10	M/6	OS	23.9	1.0/0.6	0.2/0.0	12/15	-5.5	-1.9	33
11	F/33	OD	22.9	1.0/0.7	0.2/0.0	13/18	-1.5	+0.5	39
12	F/33	OS	23.7	1.0/1.0	0.2/0.0	12/17	-0.4	-0.4	39
13	F/17	OD	28.6	1.7/0.9	1.1/0.7	21/20	8.3	-0.75	54
14	F/17	OS	22.9	1.1/0.5	0.2/0.0	21/15	-13.8	-2.38	54
15	F/13	OD	28.4	0.5/0.5	0.4/0.1	19/10	6.25	2.75	55
16	F/13	OS	27.0	0.7/0.4	0.1/0.0	16/17	-14.0	-0.5	57
17	M/5	OD	28.1	1.7/0.5	0.7/0.2	15/18	4.5	-0.5	50
18	M/5	OS	23.7	1.0/0.3	0.3/0.2	16/15	-0.75	-4.6	49

Pt=Patient; CF=Counting fingers; AXL=Axial length; Preop=Preoperative; Postop=Postoperative; UCVA=Uncorrected visual acuity; BCVA=Best-corrected visual acuity; IOP=Intraocular pressure; SE=Spherical equivalent; FU=Follow-up

scissors [Fig. 3b and Video 4], and the two ends of each suture were knotted homolaterally in front of each hollow IOL haptic [Video 5]. After 4 haptics had been reimplanted behind the iris, the sutures surrounding the haptic were tightened. After the Akreos IOL had been centered in the posterior chamber [Video 5], sutures across temporal sclera extending from a point between the 2 and 3 o'clock positions to a point between the 3 and 4 o'clock positions were knotted together [Fig. 3c]. Sutures across nasal sclera extending from a point between the 9 and 10 o'clock positions to a point between the 7 and 8 o'clock positions were similarly knotted together [Fig. 3d and Video

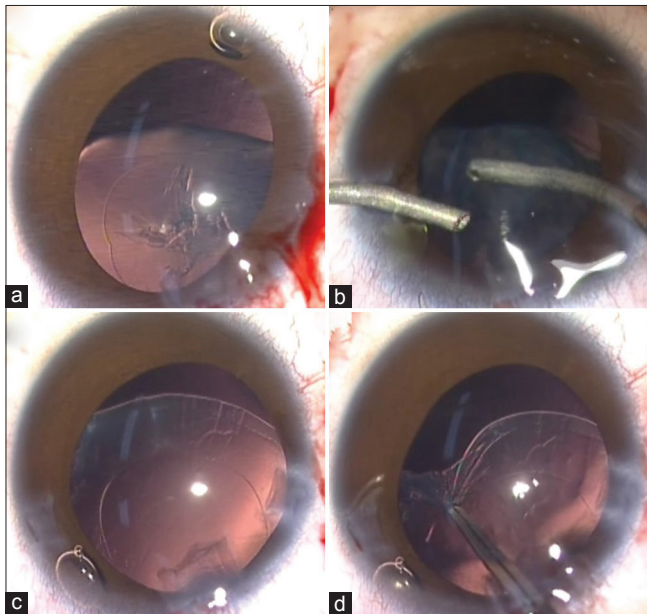


Figure 1: (a) Capsulorhexis was performed. (b) The lens was removed by bimanual I/A. (c) The empty capsular bag can be seen clearly. (d) The capsular bag was removed with a microforceps

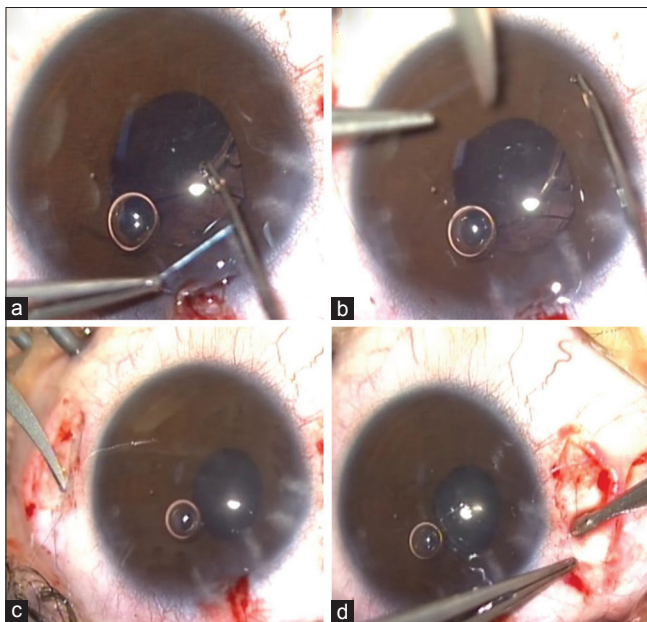


Figure 3: (a) Sutures were pulled out with a Sinsky hook. (b) The sutures were cut in half. (c) Each suture stitch across the temporal (c) and nasal sclera (d) is tied to the sclera

6]. Suture knots were buried under the conjunctiva, and the incised conjunctiva was irrigated with balanced salt solution. After I/A was performed to remove any residual viscoelastic material, anterior vitrectomy [Fig. 4a] was performed to remove vitreous that had penetrated the anterior chamber. A superior peripheral prophylactic peripheral iridectomy [Fig. 4b] was created. balanced salt solution was injected from the corneal side port to control IOP. The 3 corneal incisions were hydrated with balanced salt solution [Fig. 4c and Video 7].

Postoperatively, patients were treated with 0.3% (wt/vol) dexamethasone/tobramycin eyedrops 4 times daily for 4 weeks and 0.3% (wt/vol) dexamethasone/tobramycin ophthalmic ointment once before sleep for 4 weeks.

Postoperative examinations consisted of VA testing with manifest refraction, slit-lamp examination, IOP measurement, ECD detection, and retinal evaluation. Postoperative complications were addressed by the primary surgeon. The results of the final postoperative examination are presented in Table 1.

Statistical analysis

Preoperative vs. postoperative data were analyzed using the paired Student's t-test. Statistical measurements were presented as mean ± standard deviation. *P* < 0.05 was taken as the threshold of statistical significance. Statistical analysis was performed using SPSS software (Version 16, SPSS, Inc.).

Results

The study population comprised 10 patients with dislocated lenses. All eyes were successfully treated without intraoperative complications. The mean duration of follow-up was

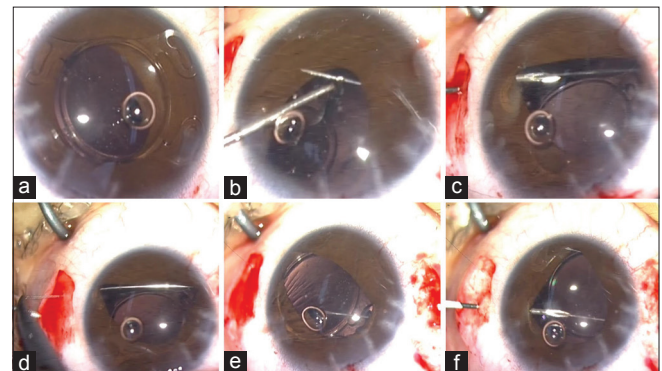


Figure 2: (a) The Akreos IOL was placed in front of the iris. (b) A straight needle with 10-0 polypropylene was passed through one hollow haptic of the IOL. (c) A 27G needle was passed through the opposite haptic of the IOL. The 10-0 needle was then guided to the exit point (d). (e and f) The other straight needle was similarly passed through the other 2 hollow haptics of the IOL

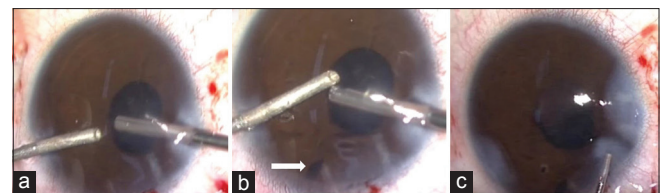


Figure 4: (a) Anterior vitrectomy and (b) iridectomy (white arrow) were performed. (c) Stromal hydration was performed to close the 3 corneal incisions

57.5 ± 14.2 months. Table 1 shows the duration of follow-up for all patients as well as mean preoperative and final postoperative UCVA, BCVA, and SE. Mean preoperative axial length was 26.34 ± 2.18 mm. Manifest refraction was determined for the 18 eyes presented for follow-up examination. At the final follow-up, mean logMAR UCVA had increased from 1.09 ± 0.49 to 0.56 ± 0.24 ($P < 0.05$). Mean logMAR BCVA improved from 0.45 ± 0.29 to 0.17 ± 0.18 ($P < 0.05$). No eye had a reduction in BCVA. Mean SE preoperatively was -3.25 diopters (D). Mean SE postoperatively was -1.93D. Mean preoperative ECD was 3660.39 ± 529.23 cells/mm² (range, 5434–2890 cells/mm²). ECD was observed to have decreased to 3583.72 ± 519.22 cells/mm² (range, 5262–2700 cells/mm²) at the final follow-up examination, indicating mean cell loss after surgery of 2.1%; however, this difference was not statistically significant. Postoperative slit-lamp microscopy showed a clear cornea, a well-centered IOL, a round pupil, and negligible anterior chamber reaction in all cases. In 2 eyes of one patient, transient ocular hypertension observed on the first postoperative day resolved without treatment. There were no cases of postoperative retinal detachment, iris atrophy, secondary glaucoma, uveitis, bullous keratopathy, or suture erosion. At the last follow-up examination, IOL centration was stable in all eyes.

Discussion

In the early stages of lens subluxation secondary to Marfan syndrome, refractive error and amblyopia may respond well to nonsurgical treatment. However, surgery is needed if patients develop secondary glaucoma, corneal endothelial dysfunction.

Various surgical techniques for lens aspiration have been described for the treatment of subluxated lens in Marfan syndrome. These include ICCE, PPL, limbal lensectomy and intralenticular bimanual I/A through two small anterior capsulorhexes (1.5–2.0 mm).^[13–16]

ICCE or PPL is preferred by many surgeons for management of a severely dislocated lens. However, the ICCE method requires a large incision for lens extraction. Use of a large incision may result in significant surgically induced astigmatism, intraoperative hypotony, and detachment of the retina.^[12] PPL increases risk for iatrogenic retinal breaks just posterior to the site of insertion used for vitrectomy probe.^[17] The eyes of patients with Marfan syndrome are more prone to develop retinal detachment,^[18] especially eyes with severe lens dislocation or high axial myopia.^[19]

With use of the novel combined surgical technique presented above, after continuous circular capsulorhexis was performed through small corneal ports, the dislocated lens was removed using bimanual intracapsular I/A under low-bottle height with 60 cmH₂O and a low-vacuum setting with 100 mmHg. The use of bimanual I/A facilitated the aspiration of lens matter in the capsule without causing damage to the posterior capsule or allowing lens matter to drift posteriorly into the vitreous cavity. The anterior chamber was also well maintained due to the use of small corneal incisions for the combined surgical technique presented here.

Lens aspiration with implantation of an IOL for postoperative refractive correction may be performed using a scleral-fixated posterior chamber IOL,^[20,21] anterior chamber IOL,^[22] or

iris-fixated IOL.^[23] Use of scleral-fixated IOLs is associated with low risk for complications.^[15] Posterior chamber IOLs are considered preferable to anterior chamber lenses. Nearly all published reports of transscleral IOL fixation describe use of a large PMMA lens,^[12,13] which requires a larger incision. As mentioned above, use of a large incision may result in intraoperative hypotony, expulsive hemorrhage, and/or retinal detachment. In recent years, the use of foldable IOLs has increased because of the wide acceptance of small-incision cataract surgery.

Kim *et al.*^[6] treated ectopia lentis secondary to Marfan syndrome with transscleral 2-point foldable IOL fixation, using a small 2.8-mm incision. This approach preserved the anterior vitreous face and resulted in satisfactory visual outcomes. However, a high rate of postoperative complications including glaucoma and pupillary capture were reported. In contrast, the combined surgical technique presented here, for the first time, is associated with a low rate of postoperative complications.

Clinically successful transscleral fixation requires effective IOL centration and no IOL tilt. The holes in the haptics of the Akreos IOL used in our study allow for 4-point fixation. Furthermore, creation of a symmetric loop suture at each haptic ensures good IOL positioning. Four-point fixation of the Akreos IOL minimizes IOL tilt^[24] and thus decreases postoperative lens-induced astigmatism. Over the course of 57.5 ± 14.2 months of follow-up, all eyes in our study showed effective IOL centration, without any instance of IOL dislocation or pupillary capture. Use of this technique may also decrease risk for IOL dislocation over the long term. Future studies should include a longer follow-up period to investigate this issue.

The techniques presented herein incorporate the use of a sutureless, 2.85-mm incision and one-piece lens injection. The Akreos IOL is foldable and easily fits into a small 2.85-mm incision. Preservation of a closed, anterior-chamber system through use of a 2.85-mm incision allowed for stable maintenance of IOP. This surgical technique may therefore reduce complications associated with intraoperative hypotony. As mentioned above the use of a sutureless, 2.85-mm incision may reduce the likelihood of postoperative astigmatism and improve visual outcomes. Fass^[24,25] and Liu^[26] have reported their use of Akreos IOLs in eyes without sufficient capsule support. The authors created limbus-based scleral flaps and fixed the IOLs at 4 points under the sclera. An exhaustive search of the literature indicates that the Akreos IOL has not previously been used without scleral flaps. This novel approach reduced the frequency of surgically induced trauma and hemorrhage.^[26]

Anterior vitrectomy was performed to remove vitreous that had penetrated the anterior chamber through side corneal ports. Because no additional incision was required for the vitrectomy, the risk for trauma induced by surgical instrumentation was decreased. Furthermore, because anterior vitrectomy decreases the strength of tractional forces on the vitreous base,^[6] our approach decreases long-term risk for retinal detachment. Prophylactic peripheral iridectomy was performed to further decrease risk for pupillary-block glaucoma. Transient ocular hypertension observed in 2 eyes of one patient on the first postoperative day resolved without treatment. No postoperative complications (e.g. glaucoma,

retinal detachment) occurred in any of the patients throughout the follow-up period.

The results of using this novel technique in 18 eyes show that the procedure is effective and leads to good visual rehabilitation and fewer postoperative complications.

Conclusion

Combined surgical technique that includes bimanual intracapsular I/A for a subluxated lens, implantation of a 4-point scleral-fixated posterior chamber foldable Akreos IOL, anterior vitrectomy, and peripheral iridectomy is a safe and effective way to treat ocular issues in patients with Marfan syndrome. This approach allows for globe stabilization during surgery and the creation of stable and simple loop sutures. These techniques minimize lens tilt,^[24] result in decreased incision size, thus minimizing risk for surgical complications. This technique offers the advantages of a small incision and the benefits of using a 4-point scleral-fixated IOL, thereby avoiding many of the complications previously associated with surgery for ectopia lentis. Although further long-term follow-up is necessary to evaluate the safety of the present technique, it appears to be an excellent technique for the management of ectopia lentis in patients with Marfan syndrome.

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Conflicts of interest

There are no conflicts of interest.

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