Management of pellucid marginal corneal degeneration

SUSMITO BISWAS, ARUN BRAHMA, CINDY TROMANS, ALAN RIDGWAY

CLINICAL STUDY

Abstract

Purpose **A retrospective study to ascertain the management of pellucid marginal corneal degeneration (PMCD).**

Method and results **Sixteen patients (average age 42.6 years) presented with PMCD. PMCD was bilateral in 13 and unilateral in 3 patients. Eight eyes underwent surgery. Nineteen eyes were managed non-surgically. Surgery involved corneal wedge excision (WE) (6 eyes), penetrating keratoplasty (PK) (3 eyes) and lamellar thermo-keratoplasty (LTK) (1 eye). Immediate pre-operative average visual acuity (VA) was 6/24, 6/10 and 6/60 with an average pre-operative astigmatism of 11.40 D, 9.75 D and 20.5 D for WE, PK and LTK respectively. After an average post-operative follow-up of 57 months, 66 months and 1 year, the average astigmatism was 8.90 D, 4.63 D and 6.00 D with an average final VA of 6/19, 6/15 and 6/6 for WE, PK and LTK respectively. In the non-surgical group, at presentation, 40% of eyes had a VA of 6/12 or better. After an average follow-up period of 32.3 months, 80% of eyes had a visual acuity of 6/12 or better. Optical correction was achieved with spectacles and or contact lenses.**

Conclusions **Surgical correction for PMCD provides poor long-term reduction of astigmatism. Patients with PMCD may be adequately corrected in the long term by the use of scleral fitted gas-permeable contact lenses.**

Key words Pellucid marginal corneal degeneration, Penetrating keratoplasty, Refractive surgery, Scleral contact lens

Pellucid marginal corneal degeneration (PMCD) is a rare ectasia of the cornea. The usual characteristics of the disease are said to include a 1–2 mm wide arcuate band of thinned cornea 1–2 mm from the limbus.[1] This is often located inferiorly, extending from the 4 o'clock to 8 o'clock position, although thinning may exist at other locations.[2,3] The adjacent clear cornea protrudes markedly and results in high against-the-rule astigmatism. Schlaeppi[4] first employed the term 'pellucid' in 1957, to emphasise the lack of vascularisation, inflammation or lipid infiltration in the pathogenesis of this disorder.

Patients often present in their third to fifth decades with reduced visual acuity due to increasing astigmatism. Management of refractive problems associated with PMCD poses many difficulties. Comfortable spectacle correction or stable contact lens correction is difficult to attain and, due to the paucity of case series, consensus as to the best form of surgical management has yet to be established.

We present our experience with 16 patients diagnosed with PMCD.

Methods

Patients diagnosed with PMCD had typical features as previously described,[1,2,5] together with characteristic corneal topography[6] (Fig. 1). Corneal hydrops, due to rupture of Descemet's membrane underlying thinned cornea, located 1–2 mm from the limbus inferiorly, was also seen in 2 patients.

The age range of the patients at diagnosis was between 22 and 55 years (average 42.6 years). Patients were followed up for an average of 6.5 years (range 10 months to 20 years). Uncorrected and corrected visual acuities were recorded. Refraction, anterior segment slit-lamp biomicroscopy, keratometry and corneal video-topography (TMS-1, Computed Anatomy) were performed at initial assessment and subsequent follow-up.

Optical correction was attempted in all patients by means of either spectacles with toric

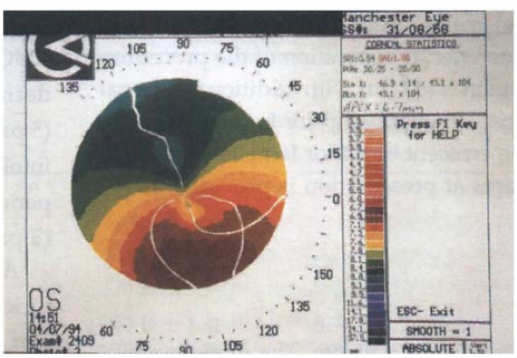

Fig. 1. *Typical topographic appearance of pellucid marginal degeneration. Inferior steepening extends into the infero-oblique meridian with flattening above.*

S. Biswas
A. Brahma
C. Tromans
A. Ridgway
Royal Eye Hospital
Manchester, UK

A. Brahma
Department of Ophthalmology
Manchester University
Manchester, UK

Arun Brahma, FRCOphth ✉
Royal Eye Hospital
Oxford Road
Manchester M13 9WH, UK
Tel: +44 (0)161 276 1234
Fax: +44 (0)161 273 6354
e-mail: brahma@man.ac.uk

Received: 3 March 1999
Accepted in revised form: 20 December 1999

Table 1. *Summary of patient data*

Patient no.	Sex/Age at referral (years)	Affected eye	Vision unaided, R and L	Visual acuity, R and L	Initial refraction of affected eye(s)	Management: initial–final	Visual acuity at last follow-up
1	M/55	Both	3/36	6/18	−7.50/+11.00 × 175 OD	Spectacles	6/9
			3/36	6/18	−4.00/+12.00 × 175 OS	Spectacles	6/12
2	M/34	Both	6/18	6/12	−7.50/+2.75 × 162 OD	Spectacles	6/5
			6/24	6/12	−1.25/+4.00 × 17 OS	Spectacles	6/5
3	M/54	Both	–	6/24	−11.00/+12.00 × 180 OD	CL	6/24
				6/18	−8.00/+12.00 × 180 OS	CL	6/9
4	M/46	Both	–	6/60	Hydrops	CL	6/60
				6/36	−17.00/+8.00 × 170 OS	CL	6/12
5	M/50	Both	3/60	6/18	−16.00/+10.00 × 170 OD	CL–WE	6/12
			3/60	6/6	−9.50/+3.25 × 170 OS	CL	6/9
6	F/34	Both	6/36	6/9	−4.00/+7.00 × 180 OD	CL–WE	6/5
			6/24	6/12	−4.00/+5.00 × 10 OS	CL–LTK	6/6
7	F/45		6/36[a]	6/36	−23.00/+10.00 × 170 OD	CL–WE	6/24
		Left	1/60	1/60	−12.00/+9.50 × 30 OS	CL–WE	1/60
8	F/31	Both[b]	1/60	6/12	−6.00/+4.00 × 155 OD	CL	6/12
			1/36	6/12	−11.75/+1.25 × 165 OS	CL	6/9
9	M/54	Left[c]	–	6/18	−9.50/+10.00 × 180 OS	CL	6/9
10	M/51	Both	2/36	6/5	−8.00/+5.50 × 170 OD	CL–WE	6/12
			6/60	6/6	−9.50/+6.00 × 175 OS	CL–WE	6/9
11	M/33	Left	2/36	6/9	+11.00/+13.00 × 62.5 OS	CL–PK	6/6
12	M/40	Both	–	0.5/60	−6.00/+9.50 × 155 OD	CL	6/18
				6/12	−13.50/+6.00 × 17.5 OS	CL	6/9
13	M/46	Both	6/36	6/18	−11.00/+9.00 × 2 OD	CL	6/9
			6/60	6/60	−14.00/+8.00 × 5 OS	CL	6/9
14	M/33	Both	–	6/18	−3.50/+8.00 × 180 OD	CL	6/9
				6/36	−3.00/+8.00 × 3 OS	CL	6/18
15	M/53	Both	1.5/60	6/9	−11.00/+15.00 × 65 OD	CL	6/12
			2.5/60	6/9	−5.00/+9.50 × 17.5 OS	CL	6/5
16	F/22	Both	–	6/9	−11.00/+4.50 × 35 OD	CL–PK	6/36
				6/6	−13.00/+4.00 × 95 OS	CL–PK	6/5

CL, contact lens; WE, wedge excision; LTK, lamellar keratoplasty; PK, penetrating keratoplasty.
[a]Right eye had keratoglobus.
[b]Right-eye had previous penetrating keratoplasty for presumed keratoconus.
[c]Right-eye had a penetrating keratoplasty for an unspecified corneal ectasia.

lenses or contact lens. The types of contact lenses used included polymethylmethacrylate (PMMA) corneal lenses, haptic and toric PMMA lenses, rigid gas-permeable (RGP) corneal lenses, apex and toric RGP lenses, and preformed RGP scleral lenses.

Where contact lens fitting and spectacle correction failed, surgical correction was undertaken. Surgical procedures included corneal wedge resection (6 eyes), penetrating keratoplasty (3 eyes) and crescentic lamellar keratoplasty (1 eye) with thermal shrinkage. This last surgical intervention was an adaptation of the procedure described by Schanzlin *et al.*[7] but with additional thermal shrinkage of the host lamellar bed prior to the placement and suturing of the crescent of donor lamellar cornea. A summary of the cases at presentation is provided in Table 1.

Results

PMCD was diagnosed in 16 patients. The demographic details are shown in Table 2. The disease was bilateral in 13 and unilateral in 3 patients. Of the 3 patients with unilateral disease, their fellow eyes were noted to have keratoglobus (patient 7), an unspecified corneal ectatic condition (patient 9) and a normal cornea (patient 11). The areas affected by PMCD were inferiorly located in 28 eyes and superonasally located in 1 eye. Against-the-rule astigmatism was present in 18 eyes, oblique astigmatism in 10 eyes and with-the-rule astigmatism in 1 eye. Ten cases were referred with a provisional diagnosis of keratoconus, 3 patients were referred with increasing astigmatism and 3 cases were initially diagnosed with PMCD. The reason for referral in most cases was decreasing visual acuity despite refractive correction (5 patients) or problems with contact lens fitting and intolerance to the lenses (6 patients). The remaining patients were referred because of increasing astigmatism (3 patients) or corneal hydrops (2 patients).

At referral, management of refractive error was by toric spectacles (8 patients), contact lenses (7 patients) and no refractive correction (1 patient). Contact lens correction was by PMMA lenses (6 patients), including one fitted with a scleral PMMA lens (patient 13) and one with a toric PMMA lens (patient 6). One patient wore

Table 2. *Demographic data*

Total no. of patients	16
Male:female ratio	12:4
Age (years)	
Mean	42.56
SD	10.24
Range	22–55

rigid gas-permeable corneal lenses. The initial and subsequent management of these patients are shown in Table 3.

The geometric average unaided vision for all eyes was 6/75 (n = 18, range 1/60–6/18). The best corrected average visual acuity at the time of referral was 6/18 (n = 29, range 0.5/60–6/5). In total 52% of eyes had a visual acuity of 6/12 or better.

The average astigmatism of patients, obtained from refraction, at referral was 8.12 dioptres (D) (range 1.25 D–15.00 D). Individual refractions are shown in Table 1.

Surgical group

The results for those patients who underwent surgery are shown in Table 3. Immediate pre-operative average visual acuity for wedge excision was 6/24, for penetrating keratoplasty, 6/10 and for the eye undergoing lamellar thermo-keratoplasty, 6/60. The average pre-operative astigmatism for wedge excision was 11.40 D. Four eyes had against-the-rule-astigmatism and 1 eye had an oblique axis of astigmatism. For those undergoing penetrating keratoplasty, the average astigmatism was 9.75 D, both with oblique axes. For the patient undergoing lamellar thermo-keratoplasty the astigmatism pre-operatively was 20.5 D, against the rule. After an average follow-up period of 57 months (range 46–65 months) the average astigmatism in the wedge excision group was 8.90 D. Three patients had against-the-rule astigmatism and 2 patients had oblique astigmatism. For patients undergoing penetrating keratoplasty, at final follow-up (average 66 months; range 36–96 months) the average astigmatism was 4.63 D with an average axis of 67.5° (50° and 85°). Corneal graft rejection occurred on two occasions in one patient. This was associated with an eccentric graft position close to the corneal limbus in order to remove the area of corneal thinning. Both episodes were successfully treated with intensive topical steroid drops. The patient who underwent lamellar thermo-keratoplasty had 6.00 D of astigmatism at the last follow-up (1 year).

The final average visual acuity of the wedge excision group was 6/19, of the penetrating keratoplasty group was 6/15 and of the patient who underwent lamellar thermo-keratoplasty was 6/6.

The final optical correction in these patients consisted of rigid gas-permeable scleral contact lenses in 3 patients (5 eyes), PMMA corneal contact lenses in 2 patients (2 eyes) and spectacles in 1 patient (2 eyes).

Non-surgical group

Twenty eyes were managed non-surgically with spectacles or contact lenses. Results are summarised in Table 3. The average visual acuity of these eyes at referral was 6/21, with 40% of eyes seeing 6/12 or better. The

Table 3. *Management and outcomes*

Pre-referral correction (no. of eyes)	Post-referral initial correction (no. of eyes)	Further management (no. of eyes)
Spectacles–14 (*1, 2, 4, 7, 8, 9, 12, 14*)	Spectacles–4 (*1, 2*) Contact lenses–9 (*4L, 7, 8, 9L, 12, 14*) None (affected by hydrops)–1 (*4R*)	WE–2 (*7*)
Contact lenses–14 (*3, 5, 6, 10, 13, 15, 16*)	Contact lenses–12 Surgery (WE)–2 (*5R, 6R*)	WE–2 (*13*), PK–1 (*16R*), LKT–1 (*6L*)
None–1 (*11*)	Contact lenses–1	PK–1 (*11L*)

Non-surgical group outcomes n = 20 eyes	Average visual acuity (geometric mean)	Average astigmatism (dioptres)
At referral	6/21 (n = 20, range 0.5/60–6/6)	8.18 (n = 19, range 1.25–15.00)
At final mean follow-up 32.2 months; range 10 months to 10 years	6/11 (n = 20, range 6/5–6/60)	7.53 (n = 14, range 3.50–15.00)

Surgical group outcomes

n = 9 eyes (*5R, 6, 7, 11L, 13, 16R*)	At referral	Pre-operative	Post-operative 2 weeks	Final post-operative (average follow-up 50.25 months)
Mean visual acuity				
WE	6/18	6/24	–	6/19
PK	6/9	6/10	–	6/15
LKP	6/12	6/60	–	6/6
Mean astigmatism (SD)				
WE (6 eyes)	7.60 D ± 2.04	11.40 D ± 3.32	8.30 D ± 5.03	8.90 D*
PK (3 eyes)	8.75 D ± 6.01	9.75 D ± 3.25	4.75 D ± 1.25	4.63 D
LKT (1 eye)	5.50 D	20.5 D	3.50 D	6.00 D

WE, wedge excision; PK, penetrating keratoplasty; LTK, lamellar thermo-keratoplasty. R, right; L, left – otherwise both eyes.
*$p < 0.05$ (Student's t-test) comparing the magnitude of pre-operative and final astigmatism.

Table 4. *Complications*

	Contact-lens-related complications		Hydrops		Surgical complications	
	Overwear syndromes	Infective keratitis	Pre-existing	New episode	Graft rejection	Retinal detachment
Patient no.	*1, 3, 13*	*10, 15*	*4, 8*	*10*	*11*	*6*
Presenting visual acuity	*1* = 6/18 OD; *3* = 6/18 OD, 6/24 OS *13* = 6/18 OD, 6/60 OS	*10* = 6/5 OD *15* = 6/9 OS	*4* = 6/60 OD *8* = 6/12	6/5 OD	6/9 OS	1/60 OS[a]
Final visual acuity	*1* = 6/9 OD, 6/12 OS *3* = 6/24 OD, 6/12 OS *13* = 6/9 OD	*10* = 6/12 OD *15* = 6/5 OS	*4* = 6/60 OD *8* = 6/12 OS	6/12 OD	6/6 OS	1/60 OS

The patients affected are given in *italic* type.
[a]Pre-existing macular hole.

average astigmatism at referral for was 8.18 D. Eight eyes had oblique astigmatism, the rest had against-the-rule astigmatism. One patient (case 4) had hydrops in the right eye precluding refraction throughout the study period. After follow-up (average 32.2 months) the average visual acuity of patients was 6/11, with 80% of eyes having a visual acuity of 6/12 or better. The average astigmatism at the latest follow-up was 7.53 D. Refractions for 5 eyes were not available. Two patients' visual acuities had dropped in one eye due to development of contact-lens-related microbial keratitis (patient 15) and from progression of the disease (patient 5). These complications and others are summarised in Table 4.

Two patients in the non-surgical group achieved satisfactory optical correction with spectacles alone. The remaining patients were managed with contact lenses. Five patients (9 eyes) were managed with rigid gas-permeable scleral contact lenses. Two patients (4 eyes) wore large-diameter, rigid gas-permeable, and apex contact lenses. One patient (1 eye) wore a rigid gas-permeable toric contact lens and 1 patient (2 eyes) wore standard rigid gas-permeable lenses.

Discussion

Typical PMCD is described as a rare, bilateral corneal ectasia. Crescent-shaped thinning of the cornea is inferiorly located. It is distinguished from other peripheral thinning disorders such as Terrien's peripheral corneal degeneration, Mooren's ulcer, senile marginal degeneration and those associated with systemic connective tissue disorders by the lack of associated inflammation.[1] The cornea protrudes anteriorly, above the area of thinning. The corneal curvature is flat in the vertical meridian. However, thinning may be located elsewhere, as was observed in patient 11, in whom the superonasal quadrant of the cornea was involved. Scarring and vascularisation may be observed in PMCD as a consequence of past episodes of hydrops,[8] such as observed in 2 of our patients (4 and 8), or from long-term contact lens wear. The topographic appearance of PMCD has been described by Maguire *et al.*[6] Though not pathognomonic, corneal topography typically shows flattening in the vertical meridian with marked steepening inferiorly below the site of thinning. Steepening extends into the infero-oblique meridians. This differs from keratoconus where steepening is greatest at the apex of the cone and reduces concentrically towards the periphery. Karabatsas and Cook[9] noted that inferior corneal steepening might not be present in the topographic picture due to limited coverage of the Placido ring target that may not extend to the limbus. Instead the topography may show against-the-rule astigmatism, with a sagging 'bow-tie' configuration, oblique inferiorly. The horizontal meridian is steep resulting in against-the-rule astigmatism. The steepest meridian is generally located 90° to the area of thinning; thus thinning located in the temporal or nasal cornea will result in with-the-rule astigmatism. The simulated keratometry readings obtained from corneal topography may differ substantially from keratometry readings. The reason for this may be due to the inability to ensure a reading from the centre of the visual axis, whilst readings centred elsewhere on the cornea may be markedly different.

Though high astigmatism is a common feature of PMCD, we have found that it is generally 'regular' enabling the patient to achieve good visual acuity with toric spectacle correction. We suggest that this is an additional feature of PCMD distinguishing it from keratoconus. Toric spectacles were the sole management option for 2 patients in our group (1 and 2), who achieved good corrected visual acuity despite high astigmatism. Other patients in the group have spectacles to use when not wearing their contact lens correction. Appropriate selection of spectacle frames and lenses can enhance the tolerance of these prescriptions. We have found that frames with small horizontal 'eyesizes' and of high refractive index lenses with anti-reflection coatings were most commonly dispensed to this group.

Contact lenses are widely used for many forms of corneal ectasia. Many advocate corneal rigid gas-permeable lenses. Contact lens fitting is difficult to perform in patients with PMCD because of the irregularity of the corneal surface and steep corneal curvature inferiorly. This often results in inferior edge lift and unstable centration. This was apparent in our study where, of the 6 patients who wore contact lenses at the time of referral, 4 had contact lens fitting problems or contact lens intolerance as the main reason for referral. Corneal contact lenses were the management option for 7

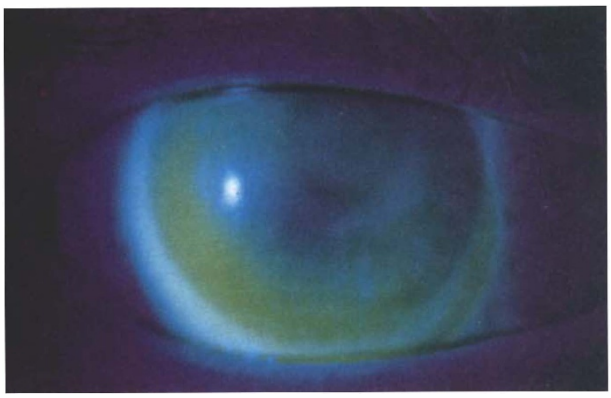

Fig. 2. *Fitting of gas-permeable scleral contact lens with instillation of fluorescein dye. Minimal corneal touch achieves a comfortable fit.*

Fig. 3. *Inferior corneal wedge excision: post-operative appearance. Note vascularisation of the scar due to loosening sutures.*

patients in our group, with 73.3% of patients achieving visual acuities of 6/12 or better. Normal diameter lenses (BZOD < 10.00 mm) were generally used for degrees of corneal astigmatism less than 5.00 D. Large-diameter corneal lenses (BZOD 10.00–12.50 mm) were found to be appropriate in eyes with higher degrees of corneal astigmatism and where centration of normal-diameter lenses proved problematic.

Gas-permeable scleral contact lenses (GPSCL) have been shown to be effective in visually rehabilitating eyes with irregular topography.[10–13] A pre-formed GPSCL was used in this group of patients and has previously been shown to be an effective alternative mode of contact lens correction where fitting with either normal or large-diameter corneal lenses had proved unsatisfactory and where patients had previously worn a PMMA scleral lens.[14] The haptic portion of the lens is supported by the sclera and maintains its positional stability. The posterior curve of the lens forms a sealed fluid compartment. A lens is selected which provides a central vault which, when filled with saline, optically nullifies corneal surface irregularities. Correction of ametropia is provided by the central anterior curve of the scleral lens. In our study the 6 patients in the non-surgical group were managed with scleral lenses (Fig. 2). After the fitting of these lenses 78% of eyes were seeing 6/12 or better compared with 22% at the time of referral. This compares favourably with the study by Tan *et al.*[12] where 72.2% of patients fitted with pre-formed scleral gas-permeable lenses achieved 6/12 or better. No serious complications were encountered with the use of scleral rigid gas-permeable lenses, although in 2 patients both eyes developed inferior corneal vascularisation from overwear. This did not require discontinuation of the lens wear.

Surgical procedures were performed on only 6 patients. Our indication for surgery in all cases was progression of astigmatism uncorrectable by contact lens wear, or contact lens intolerance. Advantages of wedge excision (Fig. 3) over penetrating keratoplasty are said to include a stronger wound, avoidance of the risk of rejection and entry into the anterior chamber being limited to a paracentesis. The disadvantage of this procedure is a tendency for the astigmatism to revert back to pre-operative levels despite overcorrection at the time of surgery. Maclean *et al.*[15] studied long-term astigmatic drift in 10 eyes of 9 patients undergoing wedge excision for PMCD and found a mean of 2.1 D astigmatic drift, although the mean follow-up time for this was not stated. Results from our study suggest that, overall, wedge excision does not provide long-term correction of astigmatism but may enable patients to resume contact lens wear.

Crescentic lamellar keratoplasty is another procedure limited to the area of thinned cornea. In Schanzlin's description of this procedure,[7] the patient's pre-operative refractive data suggested, unusually, with-the-rule astigmatism. Inferior placement of a lamellar graft resulted in flattening of the vertical meridian. Though given as an example of surgery that may be useful for tectonic correction of peripheral astigmatism, such a result would suggest further exacerbation of the flattened vertical meridian seen in the majority of cases with PMCD. A variation of this procedure was performed on one patient in this study (patient 6). In addition to the lamellar dissection of the affected area of the cornea, thermal shrinkage of the affected stroma in this area was performed prior to placement of the graft. The reduction of astigmatism from 20.5 to 6.00 D by 1 year was a dramatic response. Unfortunately more extensive follow-up and patient numbers are required before this can be recommended.

Penetrating keratoplasty has been successfully used in keratoconus. Success rates in this particular instance have been variably quoted as 75–100%.[5] Penetrating keratoplasty for PCMD is less successful because a larger graft would be required with increased risk of rejection. Speaker *et al.*[16] quoted rejection rates of 36% for host corneal diameters of 8.7 and 9.0 mm and rates of rejection of 100% for grafts fitted into host corneal diameters of 9.5 and 10 mm. This compares with rejection rates of less than 20% for grafts performed for keratoconus.[17] Placing a graft eccentrically also increases the rate of rejection, due to its close approximation to the limbus,[18] and also tends to cause increased astigmatism.[17] The outcome of patients undergoing penetrating keratoplasty in our study concurs with the findings of increased rejection risk when grafts are placed eccentrically, although refractive outcome remains good.

Other surgical procedures have been described to correct the astigmatism associated with PMCD, such as lamellar crescentic resection[10] and epikeratoplasty,[20] but lack of patient numbers indicates that no procedure can clearly be recommended as superior to the other.

Progression of PCMD may result in patients requiring gas-permeable scleral lenses to correct large degrees of astigmatism. A good improvement in visual acuity may be achieved in such patients compared with conventional lenses. Surgical correction may achieve only a modest improvement in astigmatism with long-term drift of astigmatism returning back to its original magnitude. However, surgery may enable patients to resume contact lens wear once more. At this stage no particular surgical procedure can be recommended.

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