may be unnecessary in patients for whom immediate macular hole closure can be documented intraoperatively by OCT. This may be an important patient satisfaction consideration for surgery.

Macular holes have been shown to close either by simple closure or by bridging inner retinal tissue with slow reabsorption of residual foveal subretinal fluid.[15] The mechanisms of these differences in closure are unclear but could be related to the health of the subfoveal retinal pigment epithelium. The patient reported here developed bridging tissue on intraoperative OCT with subretinal fluid reabsorption within 2 weeks. This case demonstrates the potential of intraoperative OCT as a powerful tool to elucidate the mechanisms of successful macular hole repair and guide our clinical decision making.

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Sequential traumatic and spontaneous corneal rupture in patient with osteogenesis imperfecta

Osteogenesis imperfecta (OI) is a rare collagen synthesis disorder that is caused by mutations in genes that encode type I collagen.[1] Given that type I collagen is an important structural component of the cornea and sclera, OI patients can have structural problems in the anterior segment and be vulnerable to trauma.[1–5] Here, we report a unique OI patient, who sequentially experienced a corneal rupture by minor trauma in one eye and a presumed spontaneous corneal rupture in the other eye.

An 18-month-old male with type I OI was referred to the authors' clinic just after trauma to the left eye. While his father showed him a video on a smartphone, the smartphone was dropped accidently on his left eye from a height of approximately 30 cm. Medical history revealed that he had experienced multiple episodes of injuries, such as a humerus fracture, subdural hematoma, and a pulled elbow from major or minor trauma.

On initial examination, the patient was too irritable to check the fixation and follow of each eye. Slit-lamp examination of the left eye under sedation showed a peripheral and circumferential full-thickness corneal rupture from 2 to 9 o'clock with iris prolapse through a corneal wound. The anterior chamber was totally collapsed and filled with a dense hyphema (Fig. 1A). The lens and posterior segment were not visualized, and thus ultrasonography was performed and showed a shrunken eyeball with total hemophthalmos (Fig. 1B). Under general anaesthesia, he emergently underwent repair of ruptured cornea. The cornea was so thin and fragile that it was easily broken when pinched by forceps. To achieve watertight sealing, histoacryl glue was applied along the whole length of the ruptured wound after it was cautiously adapted with

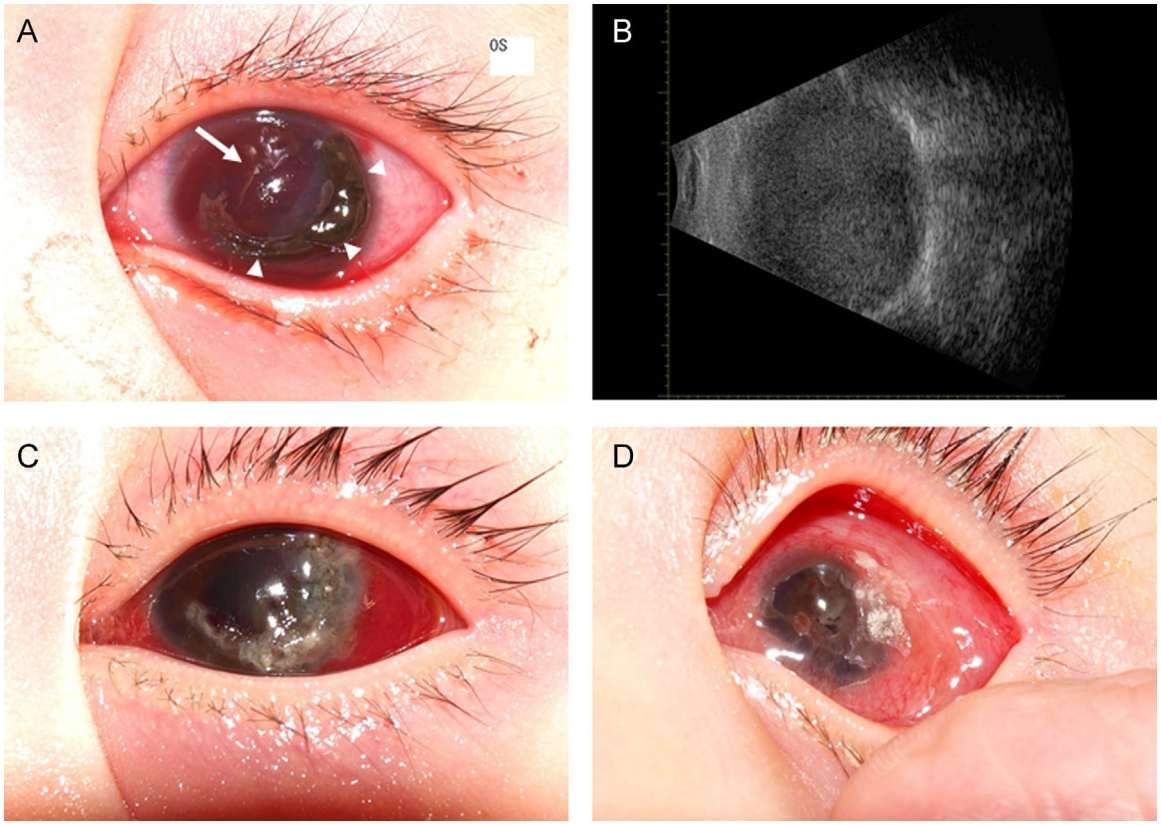

Fig. 1—Corneal rupture by minor trauma in the left eye. (A) Circumferential corneal rupture with iris prolapse (arrow heads) and dense hyphema (arrow) were observed after minor trauma. (B) B-scan ultrasonography showed shrinkage of eyeball contour with total hemophthalmos. (C) The ruptured corneal wound was successfully repaired with long-bite 10-0 nylon sutures and tissue adhesive (postoperative 1 day). (D) The eye quickly resulted in phthisis bulbi (postoperative 2 months).

long bites of 10-0 nylon sutures (Fig. 1C). During the follow-up, the left eye quickly resulted in phthisis bulbi despite the corneal wound being well adapted without any leakage (Fig. 1D).

Meanwhile, complete ophthalmic inspection of the right eye performed during primary repair of the left eye showed diffuse stromal thinning and 360° peripheral new vessels on the corneal surface (Fig. 2A, B). The central corneal thickness (CCT), measured by an ultrasonic pachymeter (Pocket Pachymeter; Quantel Medical, Clermont-Ferrand, France), was 271.7 μm (mean of 3 measurements). Therefore, we cautioned his parents to avoid additional trauma to the right eye because the cornea of the right eye was also likely to be broken even by minor trauma.

Five months after surgery in the left eye, the patient revisited our emergency room for evaluation of abnormality in the right eye. His mother denied any ocular trauma history and said that he had simply frequently rubbed his right eye before visit. Comprehensive ophthalmologic examination under general anaesthesia revealed slightly shallow anterior chamber and corneal endothelium–iris adhesions along the irregular white line (extending from 10 to 1 o'clock), which implied self-sealed wound after spontaneous rupture (Fig. 2C). Histoacryl glue was applied to the corneal wound for tectonic support despite no leakage being evident. During 4-month follow-up, the depth of the anterior chamber was well maintained without any leakage of aqueous humour, and no additional abnormality was observed in the right eye.

In cases of corneal or scleral rupture after even minor trauma, OI, Ehlers–Danlos syndrome type VI, and brittle cornea syndrome should be considered as underlying conditions.[4–9] OI can be differentiated from the other conditions by the typical signs of blue sclera, deafness, and bone fractures, and the definite diagnosis can be made by genotyping. The current patient experienced typical multiple episodes of bony fractures and was eventually found to have type I OI by genotyping, although blue sclera and deafness were not prominent.

In addition to blue sclera, other ocular findings also can be found in OI patients, especially in the cornea and the sclera. It has been well documented that OI patients have a low ocular rigidity and a reduction in the thickness of the corneal/scleral collagen fibres. Other possible findings include congenital absence of Bowman's layer, thin cornea, small corneal diameter, megalocornea, corneal opacity, and keratoconus.[1–3] However, despite the structural abnormalities just described, there have been only a few reports of corneal/scleral rupture in OI patients.[4,5]

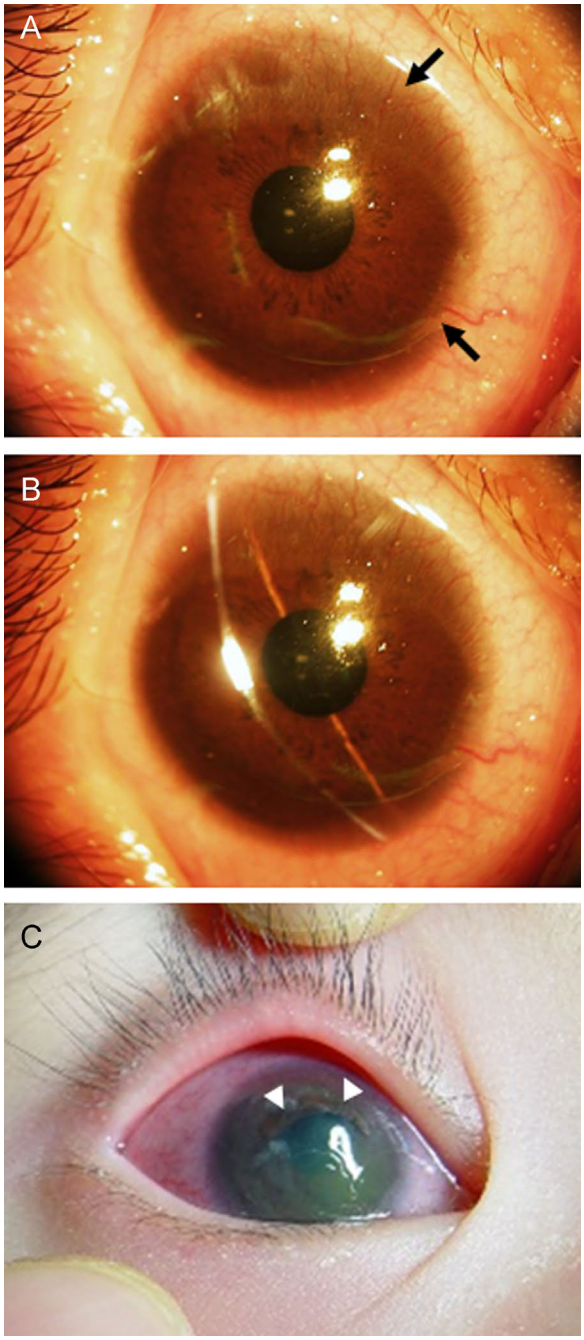

Fig. 2—Presumed spontaneous corneal rupture in the right eye. (A, B) Diffuse corneal thinning and 360° peripheral superficial new vessels (arrows) were found. (C) Presumed spontaneous corneal rupture showed an endothelium–iris adhesion along the corneal irregular white line (arrow heads).

Furthermore, we could not find any report on spontaneous corneal rupture in OI patients through the extensive literature search. In this patient, we also confirmed the markedly fragile nature of the cornea during surgery and an extremely thin cornea through a measurement of CCT. This directly resulted in corneal rupture by minor trauma in one eye and spontaneous corneal rupture in the contralateral eye. With regard to unusual prominent limbal vasculature, further investigation should be necessary to find a significance in OI patients.

Regarding surgical treatment for a thin and fragile corneal laceration, in contrast with the repair of the usual cornea, special strategies have been suggested to minimize tissue loss and to achieve watertight sealing. These include longer-bite sutures with 11-0 nylon, C_3F_8 gas tamponade in the anterior chamber, primary penetrating keratoplasty, and onlay epikeratoplasty followed by penetrating keratoplasty.[4,6–9] In this case, the lacerated corneal wound was successfully repaired by long-bite sutures with 10-0 nylon and application of cyanoacrylate adhesive as donor corneas or 11-0 nylon were not available at the time.

In summary, this case highlights that OI patients are vulnerable to the ocular trauma that may result in unwanted permanent visual loss. Therefore, all physicians who deal with OI patients should be aware of the importance of ophthalmic screening, including CCT measurements, and advise the patients to undergo regular ophthalmic examinations. Moreover, the patients and guardians should be warned of the possibility of ocular injury even by minor trauma and be encouraged to use protective eyewear particularly during physical activities.

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Eyebrow madarosis reflecting an intradermal neoplasm: pleomorphic adenoma, a rare brow tumour

Pleomorphic adenoma is a neoplasm named for its mixture of disparate tissue elements, including branching double-layered ductules within a fibroid, myxoid, cartilaginoid, and (rarely) adipocytic stroma.[1] A rare cutaneous tumour presumably arising from apocrine or eccrine sweat ducts, it has been described in the eyelid, where it may occupy various locations, including the sub-brow region.[1,2] Well-recognized tumours with identical histology may arise from the ductules of the major salivary glands and all portions of the lacrimal gland.[3] We report a pleomorphic adenoma of the brow that presented with the unusual symptom of focal brow loss.

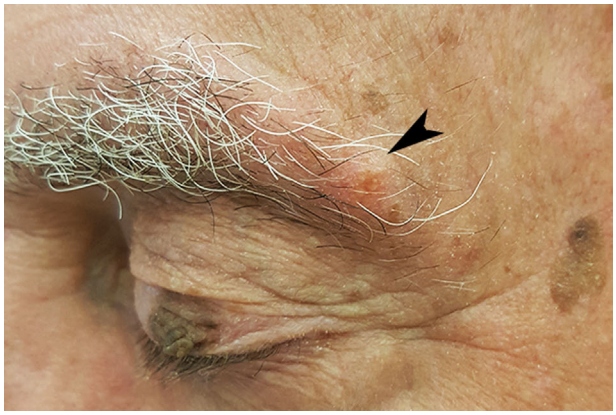

Fig. 1—Hair loss and tumour of the temporal portion of the left brow (arrow). The pigmented eyelid lesion is a seborrheic keratosis.

A healthy 77-year-old male complained of hair loss on the temporal third of the left eyebrow. Examination disclosed a subtly elevated, nontender, faintly erythematous subcutaneous mass in that location (Fig. 1). Excisional biopsy including overlying skin yielded a mass measuring 0.7 cm × 0.2 cm × 0.3 cm. Histology showed a well-circumscribed, nonencapsulated intradermal tumour consisting of branching and nonbranching strands of tubuloalveolar structures arranged among chondroid and adipocytic zones (Fig. 2A). The tubules were composed of a double layer of epithelial cells, the inner layer of which stained positively with epithelial immunostains AE1/AE3, EMA, and CAM5.2 (spotty) as well as gross cystic fluid disease protein (GCDFP-15), an apocrine marker. The outer layer was positive for S100 and NSE but not SMA or GFAP. Ki67 showed a low proliferative rate (<5%).

Formerly designated "chondroid syringoma" and "cutaneous benign mixed tumour," the current preferred terminology of this rare cutaneous neoplasm is "pleomorphic adenoma" implying morphologic variety that includes epithelial and mesenchymal portions.[1] Histology

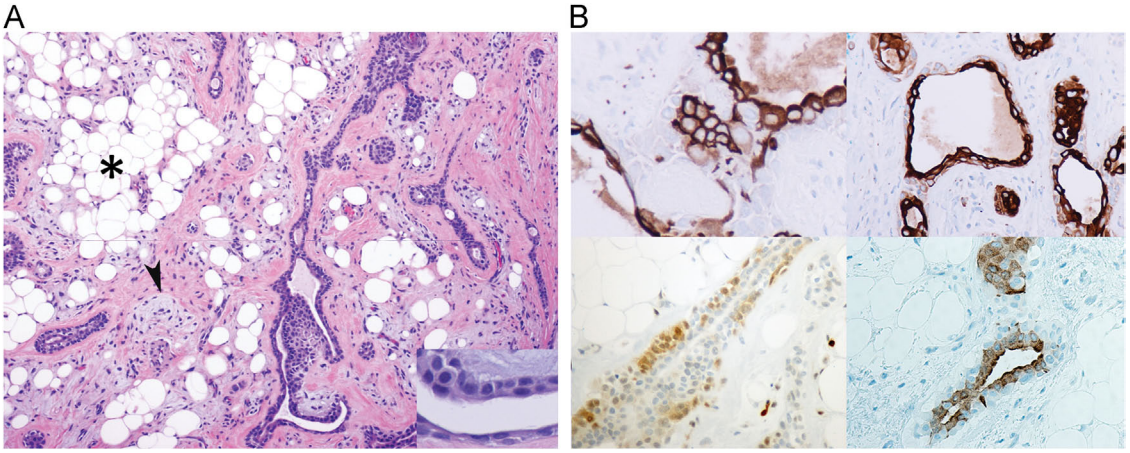

Fig. 2—(A) Tumour consists of branching, double-layered tubuloalveolar structures amidst adipose tissue (asterisk) and chrondroid tissue (arrowhead). Inset shows a double layer of cuboidal cells (hematoxylin–eosin, original magnification ×200, inset ×400). (B) Immunohistochemistry highlights adluminal layer of tubules for AE1/AE3 pancytokeratin stain (upper left) and CAM 5.2 low-molecular-weight keratin (upper right). Outer layer of cuboidal cells stains positively for neuron-specific enolase (NSE) (lower left). Inner layer is positive for gross cystic disease fluid protein-15 (GCDFP-15) (lower right) (immunperoxidase reaction, diaminobenzidine chromogen, ×400).