

Case Report

Recurrent and Spontaneous Release of Epiretinal Membrane in a Toddler

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Keywords

Epiretinal membrane · Young children · Spontaneous · Recurrent · Strabismus

Abstract

We report a case of an epiretinal membrane (ERM) in a 3-year-old girl, which was accidentally discovered after a strabismus surgery. The ERM occurred twice in 2 years and spontaneously released within 3 months, which has not been previously reported.

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Introduction

Epiretinal membrane (ERM) is a nonvascular fibrocellular proliferation that occurs on the surface of the retina and causes retinal thickening and wrinkling, leading to visual impairment and metamorphopsia [1]. ERM mostly occurs in patients older than 50 years and is usually associated with posterior vitreous detachment [2]. Idiopathic ERM rarely occurs in young children and is likely underdiagnosed [3]. Here, we report a case of ERM found in a 3-year-old girl who was revisited after strabismus surgery. In this case, ERM appeared twice in 2 years and spontaneously subsided.

Case Report/Case Presentation

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Fig. 1. Fundus image: a large amount of fundus extorsion without ERM at the first visit. ERM, epiretinal membrane.

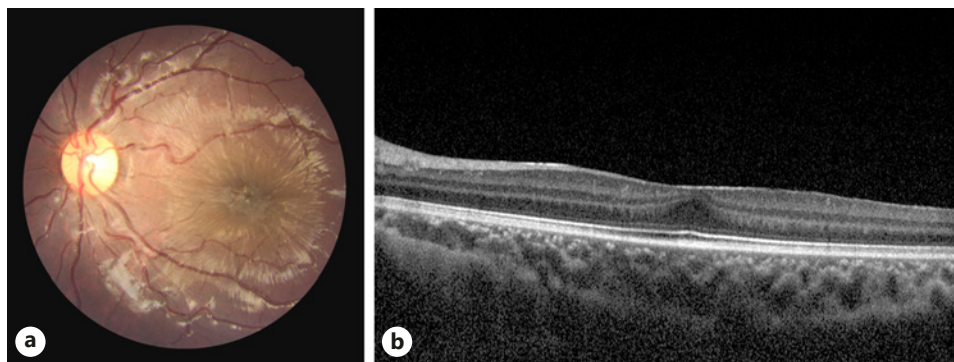


Fig. 2. **a** Fundus image: the ERM appeared at the first time. **b** SD-OCT image: the ERM appeared at the first time. ERM, epiretinal membrane; SD-OCT, spectral-domain optical coherence tomography.

A 36-month-old girl presented to us with apparent torticollis and bilateral exohyperopia (shown in Fig. 1). She received a diagnosis of bilateral congenital superior oblique muscle palsy after careful examination of her eye movements. Evaluation of her anterior segment and fundus showed normal results, except for marked excyclotorsion of both eyes.

Bilateral overaction of inferior oblique muscles was surgically corrected, with the recession of left inferior oblique muscle and the extirpation of right inferior oblique muscle. Two weeks later, we reevaluated her ocular extorsion by fundus photography and found that a grayish ERM occurred in her left macular, which presented as radial striae distributed in the foveal center (shown in Fig. 2a). The right fundi appeared to be normal. No signs of anterior uveitis, such as ciliary congestion, inflammatory cells, or precipitates, were noted. The spectral-domain optical coherence tomography showed the increase of the retinal thickness, the loss of the foveal contour, and a thin hyper-reflectivity at the vitreoretinal interface in her left eye (shown in Fig. 2b). Fundus fluorescein angiography with general anesthesia showed that both vitreous were clear and there was no dye leakage from the retinal blood vessels. Her visual acuity was not evaluated because she could not cooperate very well.

The girl had several transient low-grade fevers (<37.5°C) and rashes before and after the surgery. Therefore, a series of examinations were arranged for her. Antibodies against the Epstein-Barr virus, rubella virus, cytomegalovirus, toxoplasmosis, antineutrophil cyto-

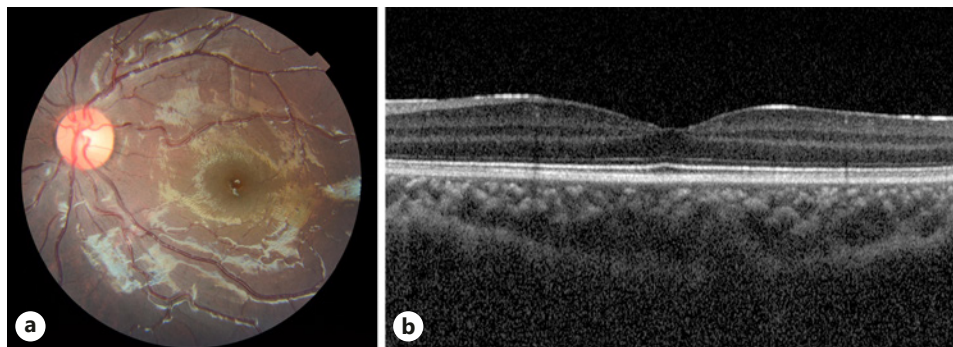


Fig. 3. **a** Fundus image: the ERM spontaneously released. **b** SD-OCT image: the ERM spontaneously released. ERM, epiretinal membrane; SD-OCT, spectral-domain optical coherence tomography.

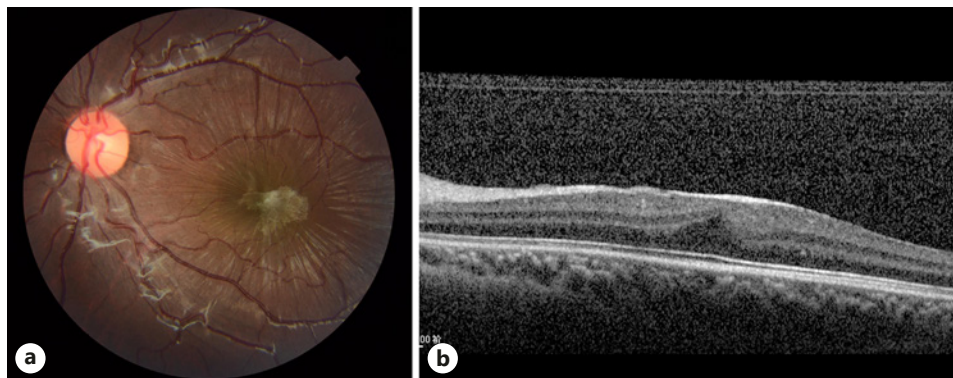


Fig. 4. **a** Fundus image: the ERM reappeared. **b** SD-OCT image: the ERM reappeared. ERM, epiretinal membrane; SD-OCT, spectral-domain optical coherence tomography.

plasmic, and antinuclear, were not detected in the serum. The result of T-SPOT.TB test was negative. Pediatricians diagnosed the girl with only a mild cold and skin allergy.

One week later, the ERM became prominent, and surgical intervention was considered. Two weeks later, the ERM started to retreat. After 2 months, the membrane spontaneously disappeared, and the optical coherence tomography confirmed normal foveal configuration without ERM (shown in Fig. 3a, b).

We followed up the girl every month. Unfortunately, 12 months after the initial presentation, without complaint of blurred vision, the macular lesions in her left fundus reappeared (shown in Fig. 4a, b). Similar to the previous episode, the ERM was released again within 3 months (shown in Fig. 5a, b). After the recovery, her best-corrected visual acuity in the left eye was 20/50 (the cycloplegic refraction was +5.0 DS/−2.5 DC × 110°), meanwhile, the right eye was 20/30 (the cycloplegic refraction was +3.25 DS/−0.75 DC × 70°).

Discussion/Conclusion

ERM rarely occurs in a toddler. The young children did not complain of unilateral blurred vision and the presentation of ERM was insidious both made this condition difficult to accurately diagnose. In this case, the ERM was found incidentally after a strabismus surgery, but there was no evidence of a cause-effect relationship between the emergence of ERM and the

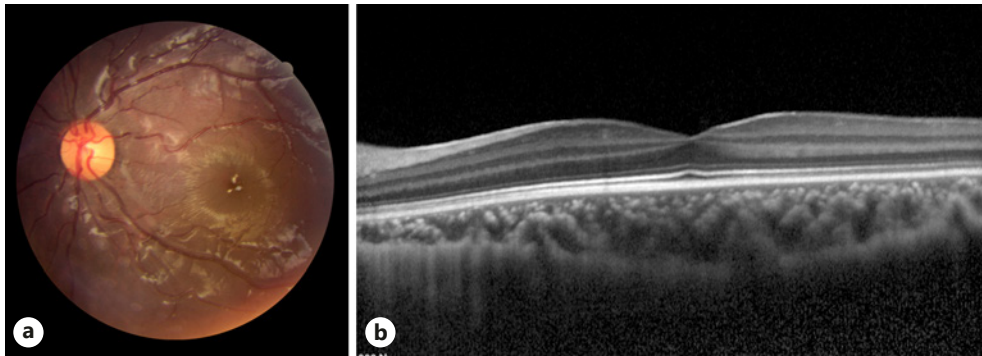


Fig. 5. **a** Fundus image: the ERM spontaneously released again. **b** SD-OCT image: the ERM spontaneously released again. ERM, epiretinal membrane; SD-OCT, spectral-domain optical coherence tomography.

strabismus surgery. It was very certain that no scleral perforation occurred during the operation. The possibility of anterior segment ischemia was not to be considered because no rectus muscle was detached. Neither the symptoms of infection such as swelling, pain, hyperemia nor signs of uveitis were observed. Fundus fluorescein angiography showed no dye leakage from the posterior and peripheral retinal blood vessels. Although the increase in the foveal thickness following inferior oblique muscle had been observed, the change was small [4]. No clinical change of macular or ERM following strabismus surgery were reported before.

Because the girl had a history of repeated low-grade fever and rashes. The pediatricians were organized to evaluate the girl's physical condition. Pediatricians diagnosed the girl with only a mild cold and skin allergy. Mild colds and skin allergies in children will not directly lead to the occurrence of ERM.

The spontaneous resolution of idiopathic ERM in young people has been reported [2]. Conservative observation was advocated if the visual disturbance is mild, as spontaneous peeling of idiopathic ERM in young subjects may occur [2]. However, the idiopathic ERM may reappear after spontaneous or surgical peeling. One of the hypotheses was that the immature internal limiting membrane defects might allow free access to cells moving into the internal retinal surface and proliferating in the foveolar area [5]. Another speculation was that the spontaneous or surgical peeling was incomplete and the residual membrane caused the internal limiting membrane to contract [6].

In this case, the recurrent ERM spontaneously released again, and the interval between the 2 episodes was 1 year, which has not been reported before. This case suggests that doctors should perform fundus examination and fully inform patients of risks before strabismus surgery. The amblyopia, dragged-fovea diplopia syndrome, secondary macular hole, and recurrence of the membrane should be noticed [7, 8], so a closely follow-up is very important.

Statement of Ethics

The patient's parents have given their written informed consent to publish their case (including publication of images).

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Bai Xue-qing: the first author who drafted, revised, and submitted the work. Li Li: the coauthor who contributed to design the work substantially. Cui Yan-hui: the coauthor who contributed to the conception of the work substantially. Li Ning-dong: the corresponding author who agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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