Pyogenic Granuloma of the Cornea in an Infant With Unilateral Microphthalmia

Pyogenic granuloma is an exuberant proliferation of granulation tissue that typically develops after minor trauma or surgery. This well-known, common inflammatory entity occurs most often on the skin of the face and extremities.1–3 Ocular pyogenic granulomas are usually found on the external surface of the eyelid or the palpebral conjunctiva.4,5 They can also occur at the limbus or on the bulbar conjunctiva, simulating a pterygium or a squamous cell carcinoma.6–8 Few have been reported on the cornea, probably because of its avascularity.1,2,4,9 A constant clinical finding of these reported corneal lesions is either an epithelial defect in the presence of corneal neovascularization and ocular surface disease or chronic chemical and/or mechanical irritation. Abnormal corneal vessels are the source of the newly formed proliferating capillaries.

We report an unusual case of a pyogenic granuloma of the cornea in an infant secondary to mechanical ocular trauma induced by prosthetic fitting for microphthalmia. The granuloma initially was thought to be a sporadic event, but the histopathologic findings implicate an in utero perforation, presumably the result of early midtrimester amniocentesis.

Report of a Case. A healthy 1-week-old girl was referred because, at birth, her left eye was noted to be small and a spot was noted on the left cornea. She had no family history of ocular abnormalities. A transabdominal amniocentesis was performed in the 19th week of gestation because of an elevated α-fetoprotein level. The procedure was performed by an experienced gynecologist in a single attempt under ultrasonographic guidance. It was reported to us that the tap was not bloody, and therefore the procedure was considered to be nontraumatic. It is not known whether the ultrasonographer had the amniocentesis needle in constant ultrasonic view or whether the fetus shifted during the procedure.

Examination at age 1 week showed that the left orbit was slightly shallow. The left palpebral fissure was estimated to be two thirds the size of the right. The left cornea was estimated to be 5 mm in diameter compared with 9 mm for the right eye. There was a small tan corneal opacity, about 2 mm in diameter, just above the pupil. The anterior segment appeared otherwise normal, except for a small pupil that could not dilate well and precluded examination of the left fundus. The right eye was anatomically normal.

The child was referred to an oculist to be fitted with a scleral shell to cover the defect. Because there was concern about sympathetic ophthalmia, a Gunderson conjunctival flap was used to cover the defect. Because there was concern about sympathetic ophthalmia, the left eye was enucleated 1 month later, and a Medpor (Porex Surgical Inc, Newnan, Ga) implant was placed in the left socket.

Histopathologic Findings. Macroscopically, the excisional biopsy...
specimen consisted of a smooth mass of pink soft tissue measuring 6 × 4 × 2 mm. Microscopic examination revealed a multilobed mass whose anterior part was composed of an exuberant proliferation of granulation tissue (Figure 3A and B). Peripherally the surface of the granuloma was partially covered by thickened conjunctival epithelium. Large folds and pseudorosettes of retina adhered to and were incorporated within the base of the granuloma (Figure 3C). The findings were consistent with a mass of granulation tissue (pyogenic granuloma) incorporating neurosensory retina.

Gross examination of the enucleated eye revealed a markedly microphthalmic globe measuring 12 × 10 × 11 mm, with a corneal perforation and a totally detached retina. Microscopic examination revealed lens material and neurosensory retina prolapsing and incorporated within a perforation of the central cornea. Extensive anterior synechiae were noted.

Comment. Since the first description in 1897, pyogenic granulomas have been reported in many sites, including the eyelid skin, conjunctiva, limbus, lacrimal puncta, acquired anophthalnic orbits, and veins of ocular adnexa. The occurrence of pyogenic granuloma on the cornea is relatively rare. Googe et al reviewed the topic in 1984, and a few cases have been reported since then. Of the previously reported corneal pyogenic granulomas, the youngest patient was aged 10 years. Pyogenic granuloma in the cornea has been reported to occur between 1 month to 18 years after the injury. There are some morphologic differences between pyogenic granulomas of the cornea and those of other locations. Corneal lesions appear to be more often sessile. The lack of a vascular stalk may be responsible for the more sessile form, primarily because the cornea is an avascular tissue. The morphological appearance of pyogenic granulomas of the cornea may be also influenced by chronic mechanical stress from blinking, which may limit the exophytic growth. These 2 factors may be responsible for a typically more intense inflammatory response compared with pyogenic granulomas in other locations. Despite its rarity, pyogenic granuloma should be considered in any child with a fleshy, vascular, elevated, rapidly growing corneal mass, especially in the setting of ocular trauma. Such lesions might be misdiagnosed as an anterior segment choristoma, a vascular hamartoma, a teratoma, a viral papilloma, or a squamous cell carcinoma. The age of onset, history of prior trauma, and rapid growth will often point to the correct diagnosis. In our patient we suspected that the lesion was a pyogenic granuloma because it was acquired and grew rapidly. An excisional biopsy was performed to make a definite diagnosis.

Figure 1. The anterior segment of the left eye shows microphthalmia with pyogenic granuloma of the cornea.

Figure 2. Ultrasound biomicroscopy reveals a round, pedunculated mass (top arrow) arising from the cornea and sealing the site of perforation (bottom arrow).
injuries to the skin, which leave only small cutaneous scars. Ocular injuries by amniocentesis are rare. We found 6 reports of ocular perforation. All of them describe trauma to the anterior segment and 2 to the posterior segment. A corneal leukoma may also be a sign of an “old” perforation caused by diagnostic amniocentesis. In our case, the procedure was performed by an experienced gynecologist immediately after ultrasonography, and the tap was considered nontraumatic. The findings of an underdeveloped eye together with evidence of a healed corneal perforation in the presence of a chronically detached retina suggest a perforation during amniocentesis. The partial sealing of the cornea prevented extreme hypotonia and probably allowed further development and differentiation of the ocular structures, including the retina.

In our case, the scleral shell presumably abraded the in utero perforation site. This may have led to an irritated eye and an epithelial defect that appeared to be an ulcer. Corneal infection and ulceration resulted in corneal perforation. To our knowledge, pyogenic granuloma of the cornea in response to perforation from mechanical trauma secondary to the use of a prosthetic scleral shell has not been reported.

Microphthalmia was the initial sign in our patient. Our clinical impression originally was congenital, unilateral, isolated microphthalmia of the left eye. It was not felt that any additional evaluation was warranted. This case report adds support to the notion that microphthalmia can be a rare complication of amniocentesis. We speculate that what had originally been interpreted as a sporadic event may have been caused by unrecognized ocular trauma. It is possible that the previously healed anterior segment injury resulted in a cornea predisposed to another perforation. Perhaps one insult alone is not sufficient to produce a corneal pyogenic granuloma, and the reason that these granulomas are rare, but found in this case, is that in this case the cornea sustained 2 different insults. We are unaware of any similar reports. The presence of a common entity in

Figure 3. Histopathologic findings from the resected corneal lesion are consistent with a mass of granulation tissue (pyogenic granuloma) incorporating neurosensory retina. A, Microscopic examination reveals a multilobed mass whose anterior part is composed of an exuberant proliferation of granulation tissue (top arrow). Peripherally the surface of the granuloma is partially covered by thickened conjunctival epithelium. Large folds and pseudorosettes of retina (bottom arrow) are adherent to and incorporated within the base of the granuloma (hematoxylin-eosin, original magnification ×10). B, Granulation tissue (pyogenic granuloma) composed of radially oriented proliferating capillaries (arrow) and inflammatory cells (hematoxylin-eosin, original magnification ×50). C, Incorporated retinal tissue (arrow) (hematoxylin-eosin, original magnification ×25).
an unusual site was precipitated by mechanical trauma from the use of an ocular prosthesis for unilateral microphthalmia following midtrimester amniocentesis.

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Figure 1. Fundus of the right eye showing a retinal fold extending from the optic nerve through the macula to the vitreous base at the 9-o’clock position.

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