6. Brotanek JM, Gosz J, Weitzman M, Flores G. Iron deficiency in early childhood in the United States: risk factors and racial/ethnic disparities. *Pediatrics*. 2007;120:568–75.
7. Maguire J, deVeber G, Parkin P. Association between iron-deficiency anemia and stroke in young children. *Pediatrics*. 2007;120:1053–7.
8. Benedict S, Bonkowsky J, Thompson J, et al. Cerebral sinovenous thrombosis in children: another reason to treat iron deficiency anemia. *J Child Neurol*. 2004;19:526–31.

Can J Ophthalmol 2019;54:e108–e111
0008-4182/17/$-see front matter
https://doi.org/10.1016/j.jcjo.2018.08.014

Diffuse siliconoma of the eyelid

Check for updates

Silicone oil is widely used as a vitreous substitute in vitreoretinal surgeries.[1] It is most frequently indicated in complex cases of retinal detachments and proliferative diabetic retinopathy associated with tractional retinal detachment, as well as ocular trauma to stabilize the retina and inhibit proliferative activity.[1] Liquid silicone was first injected into the vitreous cavity of rabbit eyes in 1958, and then utilized for the treatment of retinal detachments in 1962 by Cibis et al.[2] Several complications have been reported: including cataract formation; band keratopathy; rubeosis iridis; optic neuropathy; glaucoma; chronic uveitis; migration into the lateral ventricles of the brain; posterior ciliary artery occlusion; epiretinal membranes; and episcleral, subconjunctival, and orbital foreign body granulomas.[1-6] We report a rare case of silicone oil migration into the upper eyelid that was discovered during levator advancement surgery in a patient presenting with ptosis and an extensive history of retinal procedures.

Case Report

A 56-year-old man presented with ptosis and thickening of the left upper eyelid (Fig. 1A). Three years previously, he underwent numerous procedures in the U.K. for a recalcitrant retinal detachment of the left eye, including laser photocoagulation, cryotherapy, scleral buckling, pars plana vitrectomy and lensectomy, gas injection, infusion of intraocular silicone oil, retinotomies, and partial retinectomy, resulting in anatomic success with reattachment of the retina.

Ocular examination disclosed a best corrected visual acuity of 20/20 in the right eye and count fingers vision in the left eye. The left upper and lower eyelids manifested what appeared to be xanthelasma-like changes associated with yellowish upper eyelid thickening. Four millimetres of ptosis was present in his left upper eyelid. Levator function was 15 mm bilaterally. He demonstrated a 20-prism diopter left hypertropia and a left enlarged, irregular postsurgical pupil. The retina was attached with evidence of laser retinopexy scars and mild inferior retinal scarring.

A left levator advancement procedure was performed with repositioning of the lacrimal gland and graded excisions of

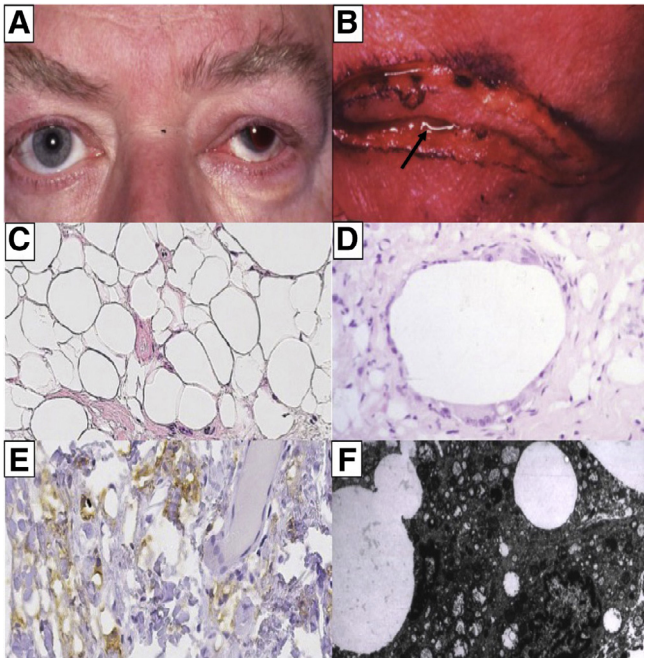


Fig. 1—A. Thickening and ptosis of the left upper eyelid. B. Surgical incision disclosed an oily material (arrow) diffusely seeping from the left upper eyelid. Histopathologic examination showed clusters of variably sized clear vacuoles, presumably silicone oil. C. With surrounding foreign body multinucleated giant cells. D. Hematoxylin and eosin, original magnifications x 400 (C), x 250 (D). E. Positive brown staining with CD68, consistent with macrophages engulfing silicone oil (original magnifications x 500). F. Electron micrograph shows vacuoles of variable size and shape within the cytoplasm of macrophages (original magnification x 4000).

prolapsed orbital fat. Intraoperatively, the entire left upper eyelid contained numerous clear fluid-filled cysts, 0.5 to 1.0 mm in diameter, encountered in the scarred pre-aponeurotic fat, and the levator and Muller's muscles (Fig. 1B). This vitreous-like appearance prompted intraoperative indirect ophthalmoscopy to assure integrity of the globe. The subcutaneous dermal layer had a striking yellow appearance and was grossly thickened to 4 to 5 times normal. The thickened skin was biopsied.

Three weeks postoperatively, residual ptosis was evident. A revision of the levator advancement was performed, and additional biopsy specimens were obtained. Fifteen years postoperatively, eyelid height and contour were satisfactory, and the patient was pleased with the cosmetic improvement.

Pathologic Findings

Histopathological examination of the biopsy specimens obtained from the left upper eyelid disclosed a normal epidermis and the presence of large clusters of tightly packed, variably sized, clear vacuoles in association with numerous histiocytes (Fig. 1C). A fair number of multinucleated giant cells, which contained similar vacuoles, were evident (Fig. 1D). The vacuoles did not show birefringence under polarized light, nor did they stain with periodic acid-Schiff or special stains for glycosaminoglycans. Immunohistochemical studies with CD68 staining the histiocytes (Fig. 1E).

Electron microscopic examination disclosed clear vacuoles of variable size and shape, localized within the cytoplasm of histiocytes (Fig. 1F). A number of the vacuoles, especially the smaller ones, were membrane-bound. Small irregular foci of an amorphous material were evident within the vacuoles. The vacuoles tended to mold and displace the nucleus of some histiocytes. These findings were consistent with a histiocytic response to foreign lipid material, presumably silicone oil.

Discussion

Silicone oil is an established treatment for achieving retinal tamponade to treat complex vitreoretinal diseases. Numerous complications have been associated with this treatment, as described above.

A potential etiology of postoperative ptosis is thought to be due to a dehiscence or disinsertion of the levator aponeurosis during surgery.[7] In our patient, the etiology was likely multifactorial with contributions from surgical manipulation, increased weight of the eyelid, and aponeurotic changes secondary to local infiltration of the silicone oil.

With regard to the mechanism of silicone oil extravasation from the vitreous chamber into extraocular tissues, various routes have been reported. Federman and Schubert[4] reported an outflow of silicone oil through the sclerotomy sites in 3% of eyes receiving this therapy. Lee et al.[6] reported a case of silicone oil leakage through melted sclera in a patient with endophthalmitis and noted other cases with silicone oil leakage through an Ahmed glaucoma valve. We postulate that the silicone oil in our case may have escaped from the interior of the globe through previous scleral vitrectomy wounds and infiltrated the superior rectus-levator complex from the subconjunctival space, which caused the ptosis and thickened eyelid appearance.

Within the English ophthalmic literature, we found 7 other cases of silicone oil migration to the eyelid from intraocular silicone oil use.[6,8-12] Our patient showed more extensive disease than the others published, which was consistent with the diagnosis of diffuse siliconoma. We also are the first to illustrate the associated pathological intracytoplasmic, clear vacuoles engulfed by macrophages using electron microscopy.

Conclusion

In conclusion, we report a case of a diffuse siliconoma of the upper eyelid, secondary to extraocular silicone oil infiltration after retinovitreous surgery. In evaluating ptosis, patients with a history of intraocular silicone oil tamponade and the clinical appearance of a thickened eyelid should suggest silicone oil infiltration as a potential cause.

Disclosure: The authors have no proprietary or commercial interest in any materials discussed in this article.

Alexander K. Soon, MD,[*,†] Seymour Brownstein, MD,[*,†] Steven M. Gilberg, MD,[*] Tina Tang, MD[*,†]
[*]University of Ottawa, The Ottawa Hospital; [†]Ottawa, Ont. Canada

Correspondence to:
Seymour Brownstein, MD; sbrownstein@ohri.ca.

Presented in part at the annual meeting of the Canadian Ophthalmic Pathology Society, Ottawa, Ont. Canada, June 17, 2016.

References

1. Morphis G, Irigoyen C, Eleuteri A, Stappler T, Pearce I, Heimann H. Retrospective review of 50 eyes with long-term silicone oil tamponade for more than 12 months. *Graefes Arch Clin Exp Ophthalmol*. 2012;250:645–52.
2. Cibis P, Becker B, Okun E, Canaan S. The use of liquid silicone in retinal detachment surgery. *Arch Ophthalmol*. 1962;68:590–9.
3. Shin H, Lemke BN, Stevens TS, Lim MJ. Posterior ciliary-artery occlusion after subcutaneous silicone-oil injection. *Ann Ophthalmol*. 1988;20:342–4.
4. Federman JL, Schubert HD. Complications associated with the use of silicone oil in 150 eyes after retina-vitreous surgery. *Ophthalmology*. 1988;95:870–6.
5. Srinivasan S, Singh AK, Desai SP, Talbot JF, Parsons MA. Foreign body episcleral granulomas complicating intravitreal silicone oil tamponade: a clinicopathological study. *Ophthalmology*. 2003;110:1837–40.
6. Lee JH, Kim YD, Woo KI, Kong M. Subconjunctival and orbital silicone oil granuloma (siliconoma) complicating intravitreal silicone oil tamponade. *Case Rep Ophthalmol Med*. 2014: 1–4.
7. Deady JP, Price NJ, Sutton GA. Ptosis following cataract and trabeculectomy surgery. *Br J Ophthalmol*. 1989;73:283–5.
8. Deguchi Y, Maeno T, Hori Y, Hiruta N, Sasai D, Sato Y. Migration of intraocular silicone oil from the vitreous cavity into the upper eyelid causing ptosis. *Case Rep Ophthalmol*. 2014;5:226–30.

9. Dehghani A, Rezaei L, Tavallali A, Dastborhan Z. Upper eyelid silicone oil migration after sutureless 23-gauge vitrectomy. *Adv Biomed Res*. 2017;6:58.
10. Donker DLT, Paridaens D, Mooy CM, van den Bosch WA. Blepharoptosis and upper eyelid swelling due to lipogranulomatous inflammation caused by silicone oil. *Am J Ophthalmol*. 2005;140:934–6.
11. Osaki TH, Osaki MH, Allemann N, Osaki T. Silicone migration: an unusual eyelid complication following intraocular surgery. *Aesthet Surg J*. 2016;36:20–2.
12. Quintyn JC, Genevois O, Ranty ML, Retout A. Silicone oil migration in the eyelid after vitrectomy for retinal detachment. *Am J Ophthalmol*. 2003;136:540–2.

Can J Ophthalmol 2019;54:e111–e113
0008-4182/17/$-see front matter © 2018 Published by Elsevier Inc. on behalf of Canadian Ophthalmological Society.
https://doi.org/10.1016/j.jcjo.2018.08.003

Canalicular melt secondary to *Pseudomonas aeruginosa* infection in a pediatric patient

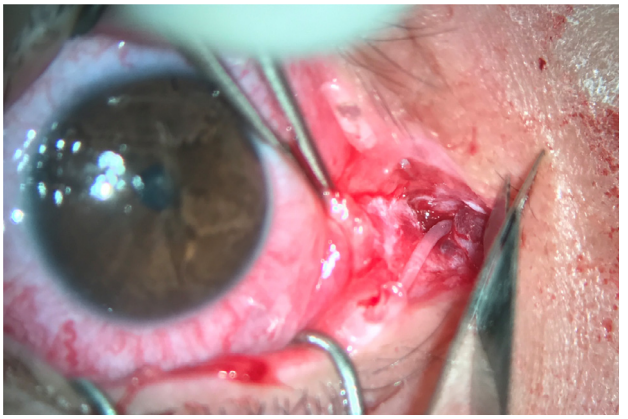
Pseudomonas aeruginosa can cause ocular or periocular infections, such as keratitis, conjunctivitis, and dacryocystitis.[1] There have been reports of periocular and eyelid necrotizing fasciitis secondary to *Pseudomonas* infection and these cases are usually accompanied by systemic illness, malnutrition, alcoholism, or minor ocular trauma.[2] Typically, bilateral ocular involvement and neutropenia is seen. This is the first documented case of unilateral, localized canalicular melting caused by *Pseudomonas* microbial infection.

Case Report

A 14-year-old girl presented with fever, swelling, redness, and pain over the right medial eyelid. She complained of epiphora, foreign body sensation, and mild blurred vision without a history of trauma to the right eye. On examination, she had a visual acuity of 20/20 bilaterally without limitations of extraocular movements. Eye examination was remarkable for congested conjunctiva and tenderness with palpation of the medial right eyelid with purulent discharge from the puncta noted.

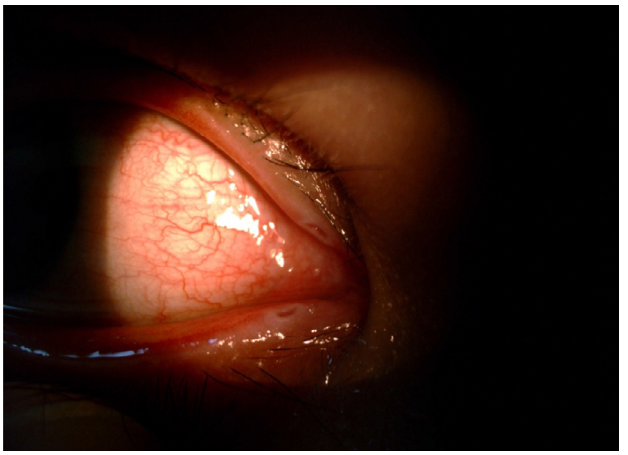
Initially diagnosed with dacryocystitis with culture positive for *Pseudomonas aeruginosa*, a follow-up examination showed upper and lower canalicular melting of the right eye (Fig. 1A, 1B). In her hospital course, the patient developed prolonged neutropenic fever. On physical exam, she developed malar rash, oral mucosal changes, and joint stiffness. Labs revealed low complement levels, high ANA (antinuclear antibody), and high ds-DNA leading to a diagnosis of systemic lupus erythematosus (SLE). She was started on high-dose prednisone and hydroxychloroquine, which resolved her neutropenia.

The patient was treated with cefepime and piperacillin and was taken to the operating room for debridement, repair, and silicone intubation of the upper and lower canaliculi (Fig. 1C). Three months after the operation, the silicone tubes were removed. The upper and lower canalicular structures were



Panel C. Intraoperative silicone intubation of the upper and lower canaliculi of the right eye.

Panel A. Right upper eyelid with probed canalicular disruption and associated purulent drainage and chemosis.



Panel D. Post-operative photo of reconstructed and healed right eyelid canalicular system.