Two Cases of Periocular Cutaneous Angiosarcoma
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Abstract: Angiosarcoma is a rare malignancy with only 8 previous reports of eyelid involvement. The authors report 2 further cases, one as a primary lesion and the other as a recurrence from a contiguous area. In both cases, the lesions appeared relatively inconspicuous, and their extent during micrographic excision was considerably larger than anticipated. Although wide surgical margins were obtained and adjuvant radiotherapy and chemotherapy was undertaken, one patient had died from distant metastasis, while the second had distant cutaneous recurrences within 1 year.

Angiosarcomas (AS) are uncommon, highly malignant, vascular endothelial tumors that typically arise in the skin or subcutaneous tissue. The cutaneous variant may be solitary or multifocal and most commonly involves the scalp and upper face in elderly whites. They may present as flat or nodular lesions, may have surrounding edema, and their color may range from bluish-red to skin tones. They are frequently misdiagnosed as benign lesions, and their extent is often underestimated. Chronic lymphedema and previous radiotherapy are predisposing factors for cutaneous AS, but only 10% of tumors have this association. No underlying genetic abnormality has been reported for cutaneous AS. AS involving the eyelid is rare, and we could identify only 8 cases in the literature.

We report 2 additional cases of AS involving the eyelid.

CASE 1
An 82-year-old white woman presented with a painless, erythematous, slightly elevated lesion in the left, medial upper eyelid, increasing in size over the previous 6 weeks. Also noted were small, irregular, poorly defined violaceous papules on the left nasal bridge and lower eyelid and cheek (Fig. 1A). An incisional biopsy confirmed recurrent angiosarcoma (Fig. 2B). A systemic workup found no metastases. She was treated by Mohs excision with wide free margins involving all the multifocal areas with a defect much larger than anticipated (Fig. 1C). Reconstruction with a cheek rotation flap, midline forehead flap, and skin grafts was followed 6 weeks later by focal radiotherapy (Fig. 1D). Although no apparent recurrence developed in the face, within 1 year, she developed a right pleural effusion from a pulmonary metastasis, followed by liver metastases. She died 6 months later.

CASE 2
A 71-year-old white male was referred with a violaceous, painless maculo-papular lesion at the left medial upper eyelid (Fig. 2A). He had been diagnosed with angiosarcoma of the nasal bridge 4 years previously and treated with complete excision of the nose, reconstruction, and adjuvant radiotherapy. An incisional biopsy confirmed recurrent angiosarcoma (Fig. 2B). A systemic workup was negative, and the eyelid lesion was treated with wide excision using frozen section control (Fig. 2C), reconstruction with a modified Tenzel flap, and adjuvant radiotherapy (Fig. 2D).
Bray LC, Sullivan TJ, Whitehead K. Angiosarcoma of the eyelid. A 35-year-old man presented with a recurrent temporal conjunctival mass (25 × 12 mm) involving about 12% to 35%. Some reports suggest that a lesion less than 5 cm carries a favorable prognosis. The current recommended therapy is controlled surgical excision with wide margins and radiotherapy, with chemotherapy using doxorubicin added for systemic involvement. While our cases appeared smaller than 5 cm, they were interpreted to have free margins on surgical excision and were treated with adjunctive radiotherapy (and additional chemotherapy in the second case); one patient died of metastasis within 2 years, and the other has had contiguous and multifocal recurrences over the subsequent 3 years.

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2. Gu¨ndu¨z K, Shields JA, Shields CL. Cutaneous angiosarcoma with orbital invasion of sebaceous carcinoma occurs in 6% to 45% of cases and is associated with poor prognosis. Patients with orbital invasion are usually treated with orbital exenteration.

CASE REPORT
A 64-year-old male presented with a recurrent fungating mass in the right lower eyelid. Histopathologic examination of the slide from a prior surgical excision showed it to be sebaceous carcinoma with involved margins (Fig. 1A). Visual acuity could not be assessed in the right eye and was 20/60 in the left eye. On examination, there was a large fungating mass involving the right lower eyelid. The mass was nontender and firm in consistency, with surface ulceration and eyelash loss (Fig. 1B). The mass was palpable in the superomedial orbit. Axial sections of CT of the orbit showed a large homogenous lesion in the medial orbit with mild contrast enhancement. Indentation and lateral displacement of the eyeball were noted (Fig. 1C). Coronal sections showed the mass to be inseparable from the eyeball. Systemic examination showed a palpable enlarged submandibular node (AJCC TNM classification: T4N1M0). The anterior segment and fundus details in the left eye were unremarkable except for senile cataract. Fine needle aspiration from the submandibular node confirmed tumor infiltration. The patient received one cycle of neoadjuvant chemotherapy with cisplatin 20 mg/m²/day over 2 hours and 5-fluorouracil 500 mg/m²/day over 6 hours for 5 consecutive days.

He developed gastrointestinal toxicity in the form of diarrhea, oral mucositis, and electrolyte imbalance. He missed several subsequent visits. A follow-up visit 7 months after neoadjuvant chemotherapy showed no residual tumor on clinical examination (Fig. 2B). Visual acuity was 20/70 in the right eye and 20/25 in the left eye. The submandibular lymph node was enlarged. CT of the orbit showed a nonenhancing irregular haze in the medial orbit (Fig. 2C). The patient was advised to have radiotherapy to the orbit and neck nodes. The patient did not receive radiation therapy and missed several subsequent visits. Nine months later, he had developed metastasis to the lungs with a 5-year actuarial survival rate reported from 12% to 35%. Some reports suggest that a lesion less than 5 cm carries a favorable prognosis. The current recommended therapy is controlled surgical excision with wide margins and radiotherapy, with chemotherapy using doxorubicin added for systemic involvement. While our cases appeared smaller than 5 cm, they were interpreted to have free margins on surgical excision and were treated with adjunctive radiotherapy (and additional chemotherapy in the second case); one patient died of metastasis within 2 years, and the other has had contiguous and multifocal recurrences over the subsequent 3 years.

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2. Gu¨ndu¨z K, Shields JA, Shields CL. Cutaneous angiosarcoma with orbital invasion of sebaceous carcinoma occurs in 6% to 45% of cases and is associated with poor prognosis. Patients with orbital invasion are usually treated with orbital exenteration.

Abstract: A 35-year-old man presented with a recurrent temporal conjunctival mass (25 × 12 mm) involving about six-clock hours of the limbus in the left eye. The mass encroached onto the temporal half of cornea and showed surface keratin, large intrinsic and feeder vessels. It infiltrated the deep corneal stroma. There were no cells in the anterior chamber. Ultrasound biomicroscopy confirmed infiltration of deep corneal stroma without intraocular invasion. Surgery involved excision of the conjunctival component with 4-mm margin, lamellar sclerectomy and a penetrating sclerokeratoplasty with 3 mm of healthy corneal margin. Cryotherapy (double-freeze-thaw) was done to the conjunctival margins. Histopathology showed it to be invasive sebaceous cell carcinoma. A thin layer of deep corneal stroma and all conjunctival margins were uninvolved. At thirty-six weeks after treatment the left eye recorded a visual acuity of finger counting at 1 meter distance and no recurrence.

Neoadjuvant Chemotherapy in Recurrent Sebaceous Carcinoma of Eyelid With Orbital Invasion and Regional Lymphadenopathy

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Abbreviations: AJCC TNM classification, American Joint Committee on Cancer; ASA, American Society of Anesthesiologists; CT, computed tomography; Cx, chemotherapy; ECOG, Eastern Cooperative Oncology Group; ER, estrogen receptor; ER, estrogen receptor; ERK, extracellular signal-regulated kinase; OS, overall survival; OR, odds ratio; T, tumor node metastasis; TNM, tumor node metastasis; TTV, transient transfection vector; UV, ultraviolet; VEGF, vascular endothelial growth factor; VFA, virtual fundusangiography; XRT, radiotherapy.

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underwent cataract surgery in the left eye. A follow-up visit 18 months after neoadjuvant chemotherapy showed no orbital recurrence. A thorough systemic evaluation, including an x-ray of the chest and ultrasound of the abdomen and pelvis, was normal. He underwent supra-omohyoid lymph node dissection. Histopathologic sections showed only the submandibular node to have metastatic sebaceous carcinoma (Fig. 2A). Thirty months after neoadjuvant chemotherapy, the patient was alive and well with no local-regional or systemic recurrence (AJCC TNM classification: T0N0M0).

**FIG. 1.** A, Histopathology from the excision performed prior to presentation. This shows cells with finely vacuolated, foamy cytoplasm, and nuclear atypia (hematoxylin-eosin) suggestive of sebaceous carcinoma. B, Clinical photograph of the 64-year-old man who presented with a large fungating mass. This seemed to arise from the lower eyelid and involved the medial orbit. The mass was nontender and firm in consistency, with surface ulceration and eyelash loss. C, Axial sections of CT of the orbit shows the large mass in the medial orbit. The mass was indenting and displacing the globe laterally.

**FIG. 2.** A, Histopathology from the lymph node dissection on higher magnification shows vaculated cytoplasm of the tumor cells suggestive of sebaceous carcinoma infiltration of the lymph node. B, Clinical photograph of the man after neoadjuvant chemotherapy shows no residual mass. Cicatrization of the lower eyelid is seen. The anterior segment in the right eye showed the pseudophakic status with a visual acuity of 20/70. C, Axial section of CT of the orbit obtained after neoadjuvant chemotherapy shows an irregular nonenhancing haze in the medial orbit.

**COMMENT**

The promising role of neoadjuvant chemotherapy in down-staging solid tumors of the breast, head, and neck, and of esophageal-gastric tumors is extensively reported in the literature. A case of sebaceous carcinoma with favorable response to neoadjuvant chemotherapy was reported by Murthy et al. In their case, a 55-year-old woman received 3 cycles of neoadjuvant chemotherapy followed by radiation, additional chemotherapy, and finally orbital exenteration for tumor control. In contrast, our case showed a dramatic
response to a single cycle of neoadjuvant chemotherapy. Effective tumor control was achieved without further radiation or exenteration. In addition, useful vision was restored after treatment. Response to neoadjuvant chemotherapy albeit in metastatic sebaceous carcinoma has been reported by Esmaeli et al. We believe that neoadjuvant chemotherapy may have a promising role to play in down-staging sebaceous carcinoma invading the orbit. However, optimal dosage and duration of treatment should be further studied through experience shared in larger number of patients.

REFERENCES


FIG. 1. Preoperative photograph demonstrating bilateral (left greater than right) cicatricial ectropion and lower eyelid retraction with lateral canthal dystopia in association with Kindler syndrome. Tarsal ectropion is present on the left side. Poikiloderma is also evident throughout the facial skin.

**Kindler Syndrome Causing Severe Cicatricial Ectropion**

Gary J. Lelli, Jr., M.D.

*Abstract:* A 32-year-old female with Kindler syndrome presented with a 5-year history of lower eyelid malposition, corneal exposure, and recurrent erosions. Severe anterior lamellar cicatricial changes were noted bilaterally, with bilateral lower eyelid ectropion and retraction. Tarsal eversion was noted on the left lower eyelid. The patient had repeatedly failed conservative treatments for keratopathy and was treated surgically, with resolution of corneal disease and improved lower eyelid position. A review of Kindler syndrome is provided, geared toward the oculoplastic surgeon who may participate in the care of these patients.

Kindler syndrome is a rare autosomal recessive disorder causing acral blisters in infancy and childhood, followed by photosensitivity, diffuse cutaneous atrophy, poikiloderma, and acral keratoses. The disease has been noted to cause keratopathy and corneal ectasia, which in one case was severe enough to require enucleation. Approximately 100 cases of the disease have been reported, but to the best of our knowledge, none in the oculoplastic literature describing the potential cicatricial eyelid changes and subsequent treatment options. Herein, we report a case of Kindler syndrome with cicatricial ectropion requiring surgical repair for treatment of exposure keratopathy.

**CASE PRESENTATION**

A 32-year-old female with Kindler syndrome, diagnosed secondary to a history of acral blistering, poikiloderma, and photosensitivity, and laboratory testing consistent with a mutation in the Kindlin-1 protein presented to the oculoplastic service with recurrent corneal erosions, left more than right, and yellow discharge from each eye. These symptoms were present for 5 years and had been treated by numerous ophthalmologists with a combination of artificial tears, ophthalmic steroids, cyclosporine eye drops, and gels. She had been manually stretching her eyelids for over 1 year, without improvement, and had not undergone prior surgical intervention to her eyelids. Her ophthalmic examination was significant for 20/15 vision in each eye with anterior lamellar cicatization bilaterally causing lower eyelid retraction and ectropion (Fig. 1). The left lower eyelid demonstrated tarsal eversion. Moderate (1 +, modified Oxford scale) superficial punctate keratopathy was present bilaterally.

The patient was treated with staged (left side, followed by right side) surgical repair as follows. A lateral canthotomy and subciliary incision was made across the length of the lower eyelid. A skin-muscle flap was elevated inferiorly to the malar eminence, releasing severe anterior lamellar cicatricial disease. After elevation of this flap, the eyelid was noted to be in good apposition to the globe but with significant horizontal tarsoligamentous laxity. An inferior cantholysis was performed, a tarsal strip fashioned and secured to the inner aspect of the lateral orbital rim with a nonabsorbable horizontal mattress suture, giving the lower eyelid appropriate height, contour, and tension. The remaining skin defect was measured (11 mm vertically × 32 mm horizontally on the left side, 7 mm vertically × 22 mm horizontally on the right side) surgical repair as follows. A lateral canthotomy and subciliary incision was made across the length of the lower eyelid. A skin-muscle flap was elevated inferiorly to the malar eminence, releasing severe anterior lamellar cicatricial disease. After elevation of this flap, the eyelid was noted to be in good apposition to the globe but with significant horizontal tarsoligamentous laxity. An inferior cantholysis was performed, a tarsal strip fashioned and secured to the inner aspect of the lateral orbital rim with a nonabsorbable horizontal mattress suture, giving the lower eyelid appropriate height, contour, and tension. The remaining skin defect was measured (11 mm vertically × 32 mm horizontally on the left side, 7 mm vertically × 22 mm horizontally on the right side).
Kindler syndrome patients with eyelid malposition. The patient reported herein demonstrated reversal of exposure keratopathy without additional erosions. The decision to treat this patient with retroauricular skin grafts, as opposed to upper eyelid skin grafts, was prompted by the patient’s disease process and the presence of diffuse cicatricial changes throughout her face. Because the disease is associated with lifelong diffuse cutaneous atrophy and photosensitivity, there is concern that even after successful surgical repair, these patients may be at risk for recurrent lower eyelid retraction and ectropion. Additionally, in many patients with the disease, cicatricial ectropion may present earlier in life, further increasing the potential need for additional revision surgeries with disease progression. Patients with Kindler syndrome should be advised to avoid sun exposure, as this is known to worsen cutaneous scarring. Additionally, depending on the extent and severity of the cicatricial eyelid and midfacial changes, midface lifting may be an appropriate correlative to eyelid repair, especially in refractory cases. While conjunctival scarring was not present in this patient, assessment for posterior lamellar scarring is important in Kindler syndrome patients, as the disease frequently affects other mucosal surfaces.

**REFERENCES**


**Eyelid Fistula Caused by a Scleral Buckle**

Omar K. Ozgur, B.S.*, Sara P. Modjtahedi, M.D.*, and Lily Koo Lin, M.D.*

**Abstract:** Complications of scleral buckle procedures for retinal detachments are rather uncommon yet may result in a broad scope of problems. The authors report a case of a chronic eyelid fistula caused by a scleral buckle. The patient was an 81-year-old woman who presented with a nonhealing left upper eyelid wound that was repeatedly misdiagnosed as a chalazion, but diagnostic workup revealed an extruded scleral buckle to be the cause. The patient was treated surgically with removal of the scleral buckle and full-thickness fistula repair and her wound healed well. A scleral buckle can erode through conjunctiva and the full thickness of an eyelid, causing an eyelid fistula and necessitate removal of the buckle.

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Scleral buckling material extrusion is a well-recognized complication of scleral buckling surgery. We describe an unusual presentation of an extruded solid silicon band causing a transpalpebral fistula and appearing clinically as a chronic eyelid ulceration rather than a frank eyelid perforation.

**CASE REPORT**

An 81-year-old Asian woman was referred by a general ophthalmologist for an oculoplastics consultation for a 1-year history of a chronic left upper eyelid nonhealing wound (Fig. A) that was unresponsive to antibiotic therapy. She described her left eye as painful, red, and constantly tearing with clear and thick discharge. A prior eyelid biopsy showed nonspecific inflammatory changes. She had a wound repair with a full-thickness skin graft 8 months prior which healed, then returned with redness, tenderness, and pain. Her ocular history was significant for placement of a scleral buckle of the left eye for treatment of retinal detachment 5 years prior. Additionally, she was pseudophakic and status post blepharoplasty bilaterally.

Her medical history was significant for hypertension and dementia. A review of systems was otherwise negative. She reported no known drug allergies. She denied smoking and alcohol use. Her family history was significant for retinal detachments. Her medications included aspirin, memantine, calcium, and a multivitamin.

On examination, her best-corrected visual acuity was 20/30 − 2 in the right eye, and no light perception in the left eye. She had a left afferent pupillary defect. There was restriction of upgaze of the left eye, but the patient and her family were unsure if that was a new problem. The external examination was significant for erythema of a ptotic left upper eyelid, evidence of a skin graft medially, and a focal area of ulceration with overlying crust at the superior aspect of the eyelid, just under the superior orbital rim (Fig. A). On attempted eversion of the left upper eyelid, there was adherence of the superior conjunctiva to the eyelid, suggesting a possible full-thickness ulcerative defect of the eyelid that was communicating with the globe. Furthermore, when the crust and ulcerative material was removed from the eyelid, the defect appeared to be nearly full thickness. No madarosis was noted. There was ptosis of the left upper eyelid with palpebral fissure width measuring 4 mm. Levator function was compromised in the left eyelid with poor (<4 mm) excursion. The slitlamp examination was notable for left eye chemosis and conjunctival injection and pseudophakia. The funduscopic examination was notable for retinal and optic atrophy of the left eye.

An extruded scleral buckle was high on the differential as a cause of the eyelid wound, and a CT scan of the orbit was ordered to further evaluate the area because the eyelid could not be everted or separated from the globe. An aerobic culture swab of the wound was taken and oral amoxicillin and clavulanate 875/125 mg orally twice a day was started. At follow-up 1 week later, her examination was unchanged. Her CT scan demonstrated the presence of a scleral buckle and was suggestive of an adherence to the eyelid (Fig. B). The Gram stain and culture showed no white blood cells and no growth of any organisms.

The patient was taken to the operating room for surgical exploration with removal of the extruded scleral buckle, and full-thickness resection of the eyelid fistula, debridement, and closure of the wound. The scleral buckle, a solid silicone encircling band, was found to have one end extruded through the conjunctiva and in communication with the full-thickness eyelid wound, which incorporated palpebral conjunctiva, levator, orbicularis, and skin. The solid scleral buckle measured 2.5 × 0.2 × 0.1 cm. The procedure was performed without any complications. Postoperatively she did well, and the area of ulceration resolved, with mild improvement of her ptosis and levator function and with improved full motility of the left globe. There was no recurrence of the lesion during 2 years of follow-up.

**COMMENT**

Scleral buckle placement is a common procedure in retinal detachment repair. Rarely, complications may arise requiring the removal of the buckle including scleral buckle exposure, extrusion, migration, intrusion, infection, chronic pain, inflammation, foreign body sensation, strabismus, diplopia, recurrent subconjunctival hemorrhage, macular distortion, impingement of the optic nerve, swelling of buckle elements, granuloma, sudden vision loss, and cutaneous extrusion. Hilton and Wallyn\(^2\) describes 600 cases of retinal detachment repairs with scleral buckles in which 23 (3.8%) were removed because of infection, foreign body sensation, recurrent conjunctival hemorrhages, impingement on the optic nerve, or macular distortion. On diagnosis of infection, immediate removal of the buckle is necessary to prevent further complications.\(^3\)

Extrusion and intrusion of scleral buckling material are recognized complications,\(^4\)–\(^6\) yet our patient is unusual in that her eyelid findings were repeatedly misdiagnosed as a chalazion, despite having undergone an eyelid biopsy to rule out more ominous pathology. It took over 1 year before an accurate
diagnosis was made and her condition was treated. We found 2 other reports in the literature of similar instances in which a silicone sponge implant caused an eyelid fistula.4,5 Our case differs in that the sponge material in the other reports had visibly perforated through the eyelid skin. Our case involved a solid silicone band that eroded through the palpebral tissues and gave the appearance of an ulcerative lesion on the eyelid rather than an obvious perforation. There are a few theories of the pathophysiology of an eyelid fistula formation. According to the mechanic theory, a dislocated silicone implant provides tension and friction against layers of the eyelid, which eventually becomes fragile enough for perforation.5 Another theory is related to aging changes of Tenon capsule in the elderly in which the capsule becomes more atrophic and provides less support to prevent extrusion of the implant.5,6

REFERENCES

Ocular Bacillary Angiomatosis in an Immunocompromised Man
Meltzer A. Murray, M.D.*, Katherine J. Zamecki, M.D.*, Joseph Paskowski, M.D.*, and Gary J. Lelli, Jr., M.D.†

Abstract: An immunocompromised man presented with an inflammatory eyelid lesion. Biopsy was performed; histopathology and special staining confirmed a diagnosis of bacillary angiomatosis. The man was treated with oral erythromycin, and the lesion resolved. The etiologic agents of bacillary angiomatosis are Bartonella henselae and Bartonella quintana, Gram-negative coccobacilli. The organisms stain positively with the Warthin-Starry silver stain. Histopathologic examination of hematoxylin and eosin–stained sections revealed a highly vascular lesion composed of slit-like and focally dilated blood vessels lined by plump endothelial cells. The intervening stroma contained a variety of inflammatory cells with predominantly neutrophils (Fig. 2). There were granular basophilic deposits comprised of pleomorphic bacilli (Fig. 3). The organisms stained positively with the Warthin-Starry silver stain. Gram (Brown-Brenn) and acid-fast stains failed to reveal organisms. Based on the histopathologic findings, the diagnosis of bacillary angiomatosis (BA) was made.

DISCUSSION
The first report of the entity that has become known as BA was made by Stoler et al. in 1983 at which time they used

A 54-year-old Hispanic man infected with HIV developed progressive swelling of his left lower eyelid over the course of several weeks. He had been taking acyclovir for

FIG. 1. A, External color photograph of the lower eyelid mass. B, Mass as it appeared on the palpebral conjunctiva.

FIG. 2. Stroma of the lesion containing inflammatory cells, mainly neutrophils.
There are only a few reports in the literature of such lesions. Subsequent investigators described papular skin lesions clinically resembling Kaposis sarcoma, which showed suppuration and masses of cocobacillary organisms that were Gram-negative and silver positive with the Warthin-Starry stain. LeBoit et al. initially called the process an epithelioid hemangioendothelioma-like vascular proliferation, considering it a manifestation of cat-scratch disease. Subsequently, this pseudoneoplastic lesion has become known as BA.

The etiologic agents of BA are currently classified in the genus Bartonella. The organism was initially classified as Rochalimaea henselae and later named Bartonella henselae, which is also considered the major agent of cat-scratch disease. B. henselae and B. quintana are currently designated as the organisms responsible for BA. Bartonella is a Gram-negative rod. The domestic cat is thought to be a significant reservoir with transmission to humans via a feline-oral route.

While typically affecting the skin of immunocompromised individuals as solitary or multiple lesions, BA has been reported to involve several extracutaneous sites, including the somatic soft tissues, liver and biliary systems, lung, bone, heart, brain, and oral mucosa. Systemic dissemination of organisms can result in bacteremia, sepsis, and death. In immunocompetent patients, the lesions may spontaneously regress.

The florid neovascularization characteristic of BA may lead to an erroneous diagnosis of Kaposis sarcoma, angiosarcoma, or pyogenic granuloma, conditions that merit widely divergent clinical management. The angiogenesis and cellular proliferation of BA have been studied by electron microscopy and immunohistochimistry and found to represent a florid growth of endothelial cells. BA responds well and promptly to oral erythromycin and other antibiotics. Biopsy of an accessible lesion, using silver impregnation of the organisms, should yield the correct diagnosis and permit appropriate therapy, which usually consists of at least 4 months of oral antibiotics.

BA affecting the orbit or conjunctiva is a rare entity. There are only a few reports in the literature of such lesions. However, in the correct patient population, BA should remain on the differential diagnosis list. If diagnosed, it is responsive to antibiotic therapy.

REFERENCES


The Ethmoidal Sinus Roof: Anatomical Relationships With the Intracranial Cavity

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Abstract: A detailed understanding of the relationship between the ethmoidal sinus and the intracranial cavity is essential to prevent intracranial penetration during orbital surgery. The authors analyzed 10 postmortem orbits with their adjacent skull bases of 5 Asian cadavers (3 males and 2 females; mean age of 80 years at death). After removing all orbital contents, skull and brain, the medial orbital wall, ethmoidal cells, and ethmoidal roof were also removed. From the intracranial cavity view, the ethmoidal roof was situated just lateral to the cribiform plate. From the orbital view, the location of the roof was close to the superior border of the medial orbital wall. These anatomical observations may be useful to prevent intracranial penetration and cerebrospinal fluid leakage during medial orbital wall decompression.

MEDICAL ORBITAL WALL DECOMPRESSION IS A COMMON OPERATION THAT IS CONSIDERED SAFE WHEN PERFORMED IN THE ETHMOIDAL SINUS. NEVERTHELESS, IT MAY STILL BE COMPLICATED BY INTRACRANIAL PENETrATION AND CEREBROSPINAL FLUID (CSF) LEAKAGE WHEN SURGERY EXTENDS BEYOND THIS SINUS. THEREFORE, UNDERSTANDING THE PRECISE ANATOMY OF THE ETHMOIDAL SINUS AND ITS CLOSE RELATIONSHIP WITH THE INTRACRANIAL CAVITY IS ESSENTIAL TO ACHIEVE A SAFE MEDIAL DECOMPRESSION.

The general anatomy of the ethmoidal sinus and structures surrounding it is well documented. The medial wall of the...
the ethmoidal sinus is also the lateral wall of the nasal cavity, and the nasal septum is the medial border of the intranasal space. The superior border of the intranasal space is the cribriform plate of the ethmoidal bone. The ethmoidal sinus roof is thin and separates the sinus from the intracranial cavity. The ethmoidal sinus reaches to the cribriform plate only around the superomedial corner of the sinus.

Although the ethmoidal area anatomy was previously studied, the clinical interpretation may vary between different surgical subspecialties. From the point of view of the orbital surgeon performing medial orbital decompressions, it is important to be familiar with the location of the ethmoidal roof as seen from the orbit or from the intracranial cavity. The purpose of our study was, therefore, to examine the relationships between the ethmoidal sinus and the intracranial cavity and to emphasize the implications to the orbital surgeon.

**MATERIALS AND METHODS**

Ten postmortem orbits with their adjacent skull bases of 5 Asian cadavers (3 males and 2 females; mean age, 80 years; range, 68–89 years at death) were studied. All cadavers did not have any history of orbital surgeries, ear-nose-throat surgeries, and neurosurgical operations. All cadavers were registered with Aichi Medical University, and proper consents and approvals were obtained prior to their use. All methods for securing human tissues were humane and complied with the tenets of the Declaration of Helsinki.

In this study, we defined the ethmoidal roof as the part between the lateral border of the cribriform plate and the superior border of the medial orbital wall or the frontal sinus exit. After removing the orbital contents, skull and brain, the medial orbital wall, ethmoidal cells, and mucosa were also removed. For examining the relationships between the ethmoidal sinus and the intracranial cavity, the ethmoidal roof was removed using chisels and a punch. Photographs were taken with a digital camera (CAMEDIA C-8080 Wide Zoom; Olympus, Tokyo, Japan).

**RESULTS**

From the intracranial cavity view, the location of the ethmoidal sinus roof could not be determined before removing the roof (Fig. A). After removing the bony roof, it was shown to be situated just lateral to the cribriform plate. Hence, the ethmoidal roof was not part of the cribriform plate (Fig. B). From the orbital view, the ethmoidal roof was...
obviously close to the superior border of the medial orbital wall. In the anterior part, the ethmoidal roof was adjacent to the superior border of the medial orbital wall through the frontal sinus (Fig. C), but in the posterior part, the roof was directly attached to the medial orbital wall (Fig. C, D). These anatomical findings were demonstrated in all 10 orbits analyzed.

DISCUSSION

The ethmoidal sinus is an important anatomical structure that is frequently encountered by otolaryngologists, neurosurgeons, and orbital surgeons. In this study, we demonstrated the position of the ethmoidal roof in relation to the cribriform plate and the intracranial cavity. Recognizing this position is important for orbital surgeons to avoid CSF leakage during medial orbital wall decompressions.

CSF leakage\(^4\) is an uncommon but severe complication of medial orbital wall decompression. It is thought to be induced by rotational movements that transmit forces to the cribriform plate, leading to fractures, followed by CSF rhinorrhea.\(^4\) As the cribriform plate is not part of the wall of the ethmoidal sinus and only attaches to the superomedial corner of the sinus, the fractures causing CSF rhinorrhea should include both the ethmoidal roof and the cribriform plate. Because only 2 of 3 patients with CSF leakage have symptomatic rhinorrhea,\(^4\) the rest of the patients may have only an ethmoidal roof fracture and an intact cribriform plate. In these cases, other symptoms, such as headache, meningitis, and pneumocephalus,\(^4\) should raise the possibility of CSF.

The fact that the ethmoidal roof is made of thin cortex bone\(^4\) similar to the posterior border of the deep lateral orbital wall,\(^5\) and the anatomical relations described in our study, points to the importance of avoiding strong rotational forces during decompression surgery to reduce the risk of fractures in the ethmoidal roof and the cribriform plate, followed by uncontrolled CSF leakage.

REFERENCES


Abstract: VATER association is the tendency for 5 specific anomalies (vertebral and vascular anomalies [V], anal atresia [A], esophageal atresia and/or tracheoesophageal fistula [TE], and radial and renal anomalies [R]) to occur together in one individual. Recently, malformations and abnormalities, other than those of diagnostic criteria of VATER association, have been considered significant for clarifying the nature of VATER association and for establishing guidelines for the treatment of infants with VATER association because they are supposed to be determinants of prognosis. Malformations associated with the eye have, however, scarcely been highlighted in VATER association, although several occurrences have been reported. The authors describe the first case of congenital blepharoptosis co-occurring in a patient with VATER association. The co-occurrence of several malformations in the ipsilateral face and hand of the patient were indicative of their arising from the common pathogenesis. Surgical repair of the blepharoptosis was carried out by eyebrow suspension with fascia lata when the patient was 5 years old; a favorable outcome was achieved in both function and aesthetics.

The VATER association has been described as the co-occurrence of major malformations: vertebral and vascular anomalies (V), anal atresia (A), esophageal atresia and/or tracheoesophageal fistula (TE), and radial and renal anomalies (R); a co-occurrence of 3 of the 5 malformations constitutes the diagnostic criteria.\(^1\) Although the nature and the pathogenesis of VATER association remain unknown in the majority of cases,\(^2\) numerous etiologies are known to be responsible, such as teratogenic exposures\(^3\) and genetic disorders.\(^4\) Recently, malformations and abnormalities, other than those of diagnostic criteria in VATER association, have been considered significant for clarification of the nature of VATER association and other disorders with multiple malformations,\(^2,5,6\) especially those responsible for mental retardation or other medical or developmental problems that have been highlighted for establishing guidelines for treatment of infants with VATER association. Described here, for the first time, is the case of congenital blepharoptosis co-occurring in a patient with VATER association.

CASE REPORT

A 5-year-old boy was referred to our unit for treatment of congenital blepharoptosis in the right eye (Fig. 1). The diagnosis of VATER association had been established at birth because of the

Congenital Blepharoptosis Co-occurring With VATER Association

Akihiro Ichinose, M.D.*, Koji Nomura, M.D.†,
Hideki Murakami, M.D.*, and Shinya Tahara, M.D.*

Abstract: VATER association is the tendency for 5 specific anomalies (vertebral and vascular anomalies [V], anal atresia [A], esophageal atresia and/or tracheoesophageal fistula [TE], and radial and renal anomalies [R]) to occur together in one individual. Recently, malformations and abnormalities, other than those of diagnostic criteria of VATER association, have been considered significant for clarifying the nature of VATER association and for establishing guidelines for the

FIG. 1. The patient suffered from congenital blepharoptosis with an ipsilateral anomalous long eyebrow.

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CASE REPORT

A 5-year-old boy was referred to our unit for treatment of congenital blepharoptosis in the right eye (Fig. 1). The diagnosis of VATER association had been established at birth because of the
The only child born of healthy nonconsanguineous parents, he was of short stature and with hypotrophy (~2 standard deviation), weight 1,680 g, height 42 cm; his psychomotor development was normal. The right eyebrow was anomalously long, and right microtia and alopecia of the right temple were noted. CT showed no anomaly in cranial or facial bones, and no other malformation was detected. At 1 month of life, he had undergone surgery for esophageal atresia. At 3 years of life, pollicization of his right index finger had been carried out.

The blepharoptosis was surgically repaired at the age of 5 years. Preoperatively, the margin-to-reflex distance was -1 mm in the right eye and 3 mm in the left eye; excursion of the upper eyelids was 3 mm in the right eye and 14 mm in the left eye; visus was 0.63 and 0.8; and the optic nerve and retinas were normal. Eyebrow suspension with autogenous fascia lata was selected because the function of the levator muscle was poor while that of the frontalis muscle was excellent. Two strips (width, 2 mm; length, 40 mm) of fascia lata were removed from the thigh. They were transferred in 2 tunnels made through the retroorbicularis fibroadipose layer from an incision near the margin of the upper eyelid to an incision 5 mm superior to the eyebrow (Fig. 2). The lower ends of both strips were fixed to the tarsal plate, and the upper ends were fixed to the subcutaneous tissue superior to the eyebrow. The shape and the height of the upper eyelid were also checked by physically pressing the eyebrow upward. Under-correction is preferred during operation because the fascia lata tends to shorten several weeks postoperatively.

Postoperatively, the eyelid height increased, reached its maximum height (margin-to-reflex distance: 3 mm), and achieved symmetry with the other eyelid 3 months postoperatively; no changes were observed thereafter (Fig. 3A–C). A natural shape and contour of the eyelid was obtained without lagophthalmos or conspicuous eyelid lag in the down gaze.

**DISCUSSION**

VATER association is named for the tendency of 5 specific anomalies to occur together in a single individual more commonly than would be expected.\(^1\) It is considered to arise from abnormalities during mesodermal differentiation\(^1,3,4\); however, its nature and pathogenesis remain unknown. Recently, several malformations and abnormalities, other than those of diagnostic criteria in VATER association, have been observed. First, some patients with VATER association are reported to have overlapping features of many disorders with multiple malformations.\(^7-10\) Clarifying the nature of VATER association and other disorders with multiple malformations is a challenging task.\(^5\) Second, some investigators suspect that the malformations and abnormalities, other than the criteria of VATER association, might be determinants of the prognosis.\(^6\) Long-term follow-up would be conducive to determine what features of VATER association or of features other than those of VATER association would put an individual at risk of mental retardation or other medical or developmental problems and would contribute to determine guidelines for the treatment of infants with VATER association.

Malformations around the eye have scarcely been highlighted in VATER association; however, it is anticipated that the frequency is, in fact, not low according to recent reports.\(^3,6\) More than a few occurrences have been observed, such as aniridia,\(^5\) optic disc coloboma,\(^6\) retinal anomaly,\(^6\) and strabismus.\(^9\) Such nonfatal malformations, which had been considered less important, seem to become a more crucial issue to the patients of VATER association who now have demonstrated a tendency to

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**FIG. 2.** Two strips (width, 2 mm; length, 40 mm) of autogenous fascia lata were transferred in 2 tunnels of the right eyelid and were made deep to the orbicularis oculi between the small incisions near the eyelid margin and the suprabrow. One end of both strips was fixed to the tarsal plate, and the other end was fixed to the subcutaneous tissue of the suprabrow.

**FIG. 3.** Nine months after correction by the eyebrow suspension: straight gaze (A), down gaze (B), and closed eyes (C).
Hypertrichosis of the Upper Cheek Area Associated With Travoprost Treatment of Glaucoma

Santiago Ortiz-Perez, M.D., and Jane M. Oliver, F.R.C.Ophth.

Abstract: Travoprost is a prostaglandin analog used in the treatment of open-angle glaucoma. This drug is safe and efficacious and has a low incidence of systemic and local side effects. Common local side effects are conjunctival hyperemia, iris pigmentation, and hypertrichosis of the eyelashes. The authors present a case of a patient who developed marked hypertichosis of the cheek vellus 3 months after starting treatment with travoprost.

REFERENCES


CASE REPORT

We report an 80-year-old white female with a medical history of anxiety treated with diazepam. During routine ophthalmologic examination, diagnosis of open-angle glaucoma was made, and treatment with daily topical travoprost 0.004% (Travatan; Alcon Laboratories, Fort Worth, TX, U.S.A.) was initiated. The drug was well tolerated during the first week. After 3 months of follow-up, intraocular pressure (IOP) was within normal range in both eyes. The standard static automated perimetry, optical coherence tomography, and clinical examination remained stable. Trichomegaly with mild ptosis of the eyelashes in both eyes was evident. Although this side effect was expected, she also complained of increased growth of the vellus hairs on her upper cheek (Fig. 1). We noted marked hypertichosis in the region of the upper cheek malar eminence, including greater thickness and length of the cheek hair, and an increase in the hair pigmentation (Fig. 2). Travoprost treatment was suspended, and a nonprostaglandin analog IOP-reducing agent was started. At last follow-up, 6 months later, the hypertichosis has disappeared, and the eyelashes have returned to a normal length, showing that these side effects are reversible after stopping the treatment (Fig. 3).

DISCUSSION

Prostaglandin analogs are some of the most recent additions to the list of ocular hypotensive medications available for the treatment of glaucoma. The 3 commercial drugs at present are latanoprost 0.005% (Xalatan; Pharmacia & UpJohn, Kalamazoo, MI, U.S.A.), bimatoprost 0.03% (Lumigan; Allergan, Irvine, CA, U.S.A.), and travoprost. The main mechanism of action of these agents in the reduction of IOP has been shown to be increased uveoscleral outflow of aqueous humor.3

In clinical use, prostanoids are known to cause iris and eyelid pigmentation, eyelash growth, and conjunctival hyperemia. In general, the incidence of iris and eyelash pigmentation increases with the treatment period and differs depending on the iris, skin, and hair color. This may explain why the incidence differs in

FIG. 1. Appearance of 80-year-old woman after 3 months of treatment with travoprost. Note trichomegaly with ptosis of the eyelashes in both eyes.
different countries and the psychological impact that these side effects can cause in the patients. Furthermore, it has been reported that prostaglandin analogs can affect not only the eyelashes but also the adjacent adnexal hair.2–5

The prostaglandin E2 and prostaglandin F2a have been described as possible modulators of hair growth. Topical use was reported to increase human hair growth and terminal hair counts. Minoxidil, a potent trichogenic agent, was found to enhance prostaglandin endoperoxide synthase-1 activity, suggesting a link between prostaglandin synthesis and hair growth. Furthermore, topical prostaglandin analogs used in the treatment of glaucoma have been reported to cause eyelashes growth. Nevertheless, this effect occurs only in the application area of the drug, proving that prostaglandins work in a paracrine way naturally.2,6

Our patient presented with hypertrichosis not only of the eyelashes but also of the cheek hair. This event has a more significant aesthetic impact than the eyelashes growth, especially in women, and it can be due to unnoticed drops in this area or by applying too many drops in the eye, resulting in spillage over the cheek.

Most glaucomatous patients are old, sometimes with visual impairment due to glaucoma and other possible pathologies. Sometimes, they are unaired in administering the drops. It is common for these patients to put the drops outside the eye or to use an excessive amount. This unnoticed use of prostaglandin analogs over the skin of the face by the patients can cause hypertrichosis and a marked change in their appearance.

With the widespread and increasing use of topical prostaglandin analogs, physicians must be aware of this possible side effect. Patients must be informed about this effect, which can be reversible but also can cause a considerable amount of discomfort to them.

REFERENCES


Large Particle Hyaluronic Acid Gel for the Treatment of Lower Eyelid Retraction Associated With Radiation-Induced Lipoatrophy

Jeffrey L. Peckinpaugh, M.D., Harsha S. Reddy, M.D., and Robert N. Tower, M.D.

**Abstract:** A 42-year-old female with a remote history of a left maxillary sinus tumor treated with excision and radiation therapy was referred for dry eye symptoms. Ophthalmic examination revealed left-sided exposure keratopathy, lagophthalmos, lower eyelid retraction, and fat atrophy of inferior periorbital tissue with associated hollowing. Large particle hyaluronic acid gel was injected to expand and reinforce the lower eyelid. After treatment, there was significant improvement in lagophthalmos, inferior scleral show, and periorbital hollowing. Excellent symmetry with the fellow eye was achieved. The patient reported markedly reduced dry eye symptoms. No adverse side effects were observed. The use of large particle hyaluronic acid gel shows promise as a novel nonsurgical therapy in the management of lower eyelid retraction associated with radiation-induced lipoatrophy. Additionally, large particle hyaluronic acid gel may provide better soft tissue expansion and retention than small particle hyaluronic acid gel, increasing the longevity of treatment.

Lower eyelid retraction with associated lagophthalmos and exposure keratopathy can result from multiple etiologies. These include thyroid-associated orbitopathy, trauma, age-related involution, and postsurgical changes, most often following blepharoplasty. Less commonly, retraction may result from external beam radiation of paranasal masses, which results in periorbital lipoatrophy and loss of vertical eyelid volume.1 Standard surgical treatments for lower eyelid retraction include posterior lamellar spacers, skin grafts, and myocutaneous advancement flaps. However, these procedures risk

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complications, including fibrosis, eyelid contraction, tissue rejection, and the need for further surgery. A nonsurgical alternative using small particle hyaluronic acid gel injections has previously been described by Goldberg et al. and Goldberg and Fiaschetti. We describe a similar technique using recently available large particle hyaluronic acid gel for a novel indication, the treatment of lower eyelid retraction associated with radiation-induced lipoatrophy.

**CASE REPORT**

A 42-year-old African American female was referred for lagophthalmos, lower eyelid retraction, and exposure keratopathy of her left eye. Her medical history was significant for a left maxillary sinus neuroblastoma treated 18 years prior with excision, chemotherapy, and local radiation. She reported ocular irritation and increasing difficulty in closing her left eye over the past several years. She was using artificial tears with minimal relief of her symptoms.

External examination of the left eye was notable for lower eyelid retraction with moderate inferior periorbital hollowing. No enophthalmos or periorbital scarring was noted. Slit-lamp examination showed inferior corneal punctate epitheliopathy in the left eye. The right eye examination was normal. External measurements were obtained (Table). Standard pretreatment external color digital photographs were taken (Fig. 1A, B).

A diagnosis of lower eyelid retraction with associated lagophthalmos and exposure keratopathy from suspected radiation-induced lipoatrophy was made. After full discussion of all options with the patient, we decided to proceed with injection of large particle hyaluronic acid gel tissue filler (Perlane, Medicis Corporation, Scottsdale, AZ, U.S.A.) to treat her lower eyelid retraction and periorbital hollowing.

**DISCUSSION**

Successful treatment with small particle hyaluronic acid gel of lower eyelid retraction has been described by Goldberg et al. and Goldberg and Fiaschetti. They reported a mean reduction of inferior scleral show of 1.0 mm and filler longevity of 3 to 4 months, with an average interval of 3.6 months between the first and repeat injection. This case demonstrates similar efficacy of large particle hyaluronic acid for the treatment of lower eyelid retraction, especially in the reduction of inferior scleral show and lagophthalmos (2.0 mm each). We report good functional and cosmetic success for the treatment
of inferior hollowing and lower eyelid retraction associated with radiation-induced lipoatrophy, a previously undescribed indication for hyaluronic acid fillers. External beam radiation targeting orbital and periorbital tumors is known to cause degenerative changes to periocular skin and soft tissue, which pose therapeutic challenges. Advantages of this method include minimal invasiveness in poor surgical candidates, potential reversibility, and the ability to adjust eyelid position with additional applications.

We report an adequate treatment effect for approximately 5 to 6 months in comparison with 3 to 4 months described with small particle materials. Theoretically, large particle hyaluronic acid will retain better soft tissue expansion and prolonged duration. This may enable permanent tissue responses that reduce the frequency or volume of subsequent filler injections, although no definitive evidence exists currently. Further longitudinal studies with larger sample sizes are needed to better define the frequency of maintenance injections and potential long-term complications.

REFERENCES

Enhanced Enlargement of Silicone Ocular Prostheses Achieved by Silicone Gel (MED-361)
Carrie Lynn Morris, M.D.*, Matt Singer†, and James C. Fleming, M.D.*

Abstract: Recently, silicone prostheses have been used instead of acrylic ocular prostheses. The authors sought to assess the ability of silicone gel to facilitate the growth of an ocular silicone prosthesis and to determine the best application modality to achieve maximal possible growth. Two ocular prostheses with similar baseline dimensions and contour were used for comparison of growth environment. One prosthesis was placed in an eyelid mold, while the other was placed in an open Petri dish. Silicone gel drops (MED-361; NuSil Technology, Carpinteria, CA, USA) were administered 3 times daily to both prostheses. Over the course of 41 days, an equal amount of enlargement occurred in all dimensions in both prostheses. Enlargement of a silicone prosthesis can be achieved by various modes of application of topical silicone gel.

Enhanced Enlargement of Silicone Ocular Prostheses Achieved by Silicone Gel (MED-361)
Recently, silicone prostheses have been used instead of conventional acrylic ocular prostheses in anophthalmic patients. These offer an alternative to an acrylic prosthesis, which may rarely cause an allergic tissue reaction.\textsuperscript{1,2} While refining the silicone prosthesis design, patients were instructed to use silicone gel drops to lubricate the prosthesis in an effort to maintain a polished appearance. Serendipitously, it was observed that the silicone prosthesis began to enlarge with administration of the silicone gel drops. Given this information, we postulated that it might be possible to intentionally enlarge a silicone prosthesis by application of silicone gel drops. If one could controllably manage the growth of ocular or orbital prostheses, then socket expansion of fornices could be achieved.

Therefore, a prospective, comparison, nonblinded trial was designed to evaluate whether growth of a silicone prosthesis could be achieved by application of silicone gel drops. The best application modality, drop form versus direct topical lubrication, was explored.

METHODS

Two silicone ocular prostheses with similar initial dimensions and contour (25 mm × 20.50 mm × 11.50 mm [prosthesis A]) and (25 mm × 21.00 mm × 12.50 mm [prosthesis B]) were measured at baseline. Over the course of 41 days, the prostheses were applied with silicone gel drops at a frequency of 1-2 times per day. Measurements were taken at baseline and at day 41.

FIG. 3. A, Specimen A at baseline measures 25 mm × 20.5 mm at width and height, respectively (top). Specimen A is pictured at day 41, showing a 2-mm increase in width and 1-mm increase in height, measuring 27 mm × 21.5 mm, respectively (bottom). B, Specimen A is shown to note a baseline thickness of 11.5 mm (top). On day 41, a 1-mm enlargement in thickness was noted of 12.5 mm (bottom). C, Specimen B was pictured on grid for comparison at baseline (top). Specimen B measures 25 mm × 21 mm, width and height, respectively. Specimen B grew an equivalent amount as specimen A, a 2-mm increase in width and 1-mm increase in height (27 mm × 22 mm) was observed, respectively (bottom). D, Specimen B is measured to be 12-mm thick at baseline (top). On day 41, a similar growth of 1 mm in thickness was observed for specimen B to be 13 mm (bottom).
mm × 21 mm × 12 mm [prosthesis B]) were used for comparison of growth environment (Fig. 1). Prosthesis A was placed in an orbital mold to mimic the epithelium of the bulbar conjunctiva and palpebral conjunctival fornices and eyelids. The mold was created using 300-bloom gelatin with glycerin material, which is commonly used in theatrical masks and molds because it has properties similar to human skin. Prosthesis B of the same initial size was placed within a plastic Petri dish (Fig. 2). Prosthesis volumes were not measured.

Two drops (0.05 ml) of 350 cps silicone oil gel (MED-361; NuSil Technology, Carpinteria, CA, U.S.A.) were placed on prosthesis A while it was within the eyelid enclosure 3 times a day at regularly spaced intervals. Two drops (0.05 ml) of 350 cps silicone oil gel were used to lubricate prosthesis B by direct placement with a finger. Digital photographs were taken at baseline and twice per week on a millimeter grid to assess enlargement in all dimensions (thickness, width, and height). Enlargement of each prosthesis was compared with each prosthesis’ baseline dimensions and between each prosthesis.

RESULTS

After 41 days of silicone oil gel administration to the prostheses, a 2 mm (8%) increase in width occurred in both prosthesis A and B (25–27 mm) compared with baseline parameters (Fig. 3A, B, respectively). This was a 0.05 mm/day rate of growth in terms of width. A 1-mm enlargement in height and thickness of each prosthesis A (height: 20.5–21.5 mm; thickness: 11.5–12.5 mm) and prosthesis B (height: 21–22 mm; thickness: 12–13 mm) was observed (Fig. 3A–D). This was a 0.025 mm/day rate of growth in regard to both thickness and height. Prosthesis A had a 4.9% increase in height versus 4.8% for prosthesis B. Prosthesis A became 8.7% thicker compared with prosthesis B, which became 8.3% thicker by applying silicone gel in various forms. A continuous, steady, linear growth pattern occurred in all dimensions for both prosthesis A and B during the silicone gel application.

DISCUSSION

Enlargement of silicone prosthesis is achieved by placement of topical 350 cps silicone gel in drop form or direct application. Use of 350 cps silicone gel may increase the size of a silicone prosthesis. We observed a relatively linear growth pattern of both prostheses in all dimensions with continued administration of silicone oil. One may extrapolate these data and postulate that growth would have indeed continued. Silicone polymers are thought to lay over each other in a scaffolding-like fashion, achieving enlargement.

As expected, we noted that the orbital mold was stretched by prosthesis A. In addition, it became increasingly difficult to set the prostheses in a grid position due to their growth. These expansive forces noted may not correlate to in vivo conditions induced by a contracted socket or phimotic eyelids of congenital anophthalmos. While the gelatin mold closely mimics the epithelium of human skin, it does not have the dynamic nature of a socket or includes contractile forces, nor does it have the advantage of elastic malleable properties of epithelium and dermis, which may allow for socket molding. Limitations of this study include a small sample size, application and measurement error, and lack of volume measurement. Future study goals include: (1) continued administration of silicone drops until a plateau in growth is reached, (2) cessation of drop administration to assess whether regression occurs, (3) volume analysis, (4) comparison of silicone gel and a control material, (e.g., normal saline or water), and (5) human trials involving microophthalmic and anophthalmic patients.

Silicone gel application may not be ideal for every patient in need of socket expansion due to lack of controlled growth in one dimension, but it may serve as a noninvasive alternative for those in need of overall enlargement of the socket.

REFERENCES


Pseudodacryocystitis and Nasolacrimal Duct Obstruction Secondary to Ethmoiditis

Bülent Yazıcı, M.D.*, and Zeynep Yazıcı, M.D.†

Abstract: A 23-year-old woman presented with clinical symptoms suggestive of acute dacryocystitis. She had no history of epiphora, and her lacrimal drainage system was patent on irrigation. CT findings were consistent with anterior ethmoiditis and maxillary sinusitis. Although the symptoms responded to antibiotic treatment, they recurred 2 times within the following 5 months. During the last episode, a complete obstruction of the nasolacrimal duct developed. The condition did not recur after external dacryocystorhinostomy and anterior ethmoidectomy during a follow-up of 21 months. Ethmoiditis may rarely cause a localized infection in the lacrimal sac region mimicking dacryocystitis. Recurrent infections may progress to complete obstruction of the nasolacrimal duct.

The orbital soft tissues are separated from the anterior ethmoid sinuses by thin bones, lamina papyracea, and lacrimal bone. Because of this close anatomical relationship, ethmoiditis may cause several orbital infections. Anterior ethmoiditis may rarely lead to a localized infection in the lacrimal sac region mimicking dacryocystitis (“pseudodacryocystitis”).1 We describe an adult patient with anterior ethmoiditis manifesting as recurrent pseudodacryocystitis. In this case, the repeated peri-saccal infections progressed to complete obstruction of the nasolacrimal duct, which was different from the previous cases.

CASE REPORT

A 23-year-old woman was referred with a diagnosis of recurrent acute dacryocystitis. On examination, a tender, erythematous, fluctuant swelling was noted in the lacrimal sac region under the right medial canthal tendon. Digital pressure over the swelling did not produce any discharge through the puncta. She had experienced a similar attack once previously. She denied any history of tearing. We performed lacrimal irrigation through the upper lacrimal canaliculus, after which the fluid could pass easily in the nasopharynx without reflux. The ocular examination findings were otherwise normal, and she did not have a history of a systemic disease.


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Digital subtraction macrodacryocystogram showed the patency of the lacrimal drainage path, but a slight extrinsic compression of the nasolacrimal sac and duct was noted laterally (Fig. 1A). No diverticula formation or filling defect, which could be due to a dacryolith or tumor, was seen. CT dacryocystography showed a soft tissue mass in the right medial canthal region and opacification of the anterior ethmoid sinus (Fig. 2). Mucosal thickening and air-fluid level were also noted in the right maxillary sinus. Extrinsic compression of the mass led to a thinning of the column of contrast media in both sac and nasolacrimal duct. A culture of the purulent material aspirated from the perisaccal mass yielded *Staphylococcus aureus*. Based on these findings, a diagnosis of pseudodacryocystitis arising from anterior ethmoiditis was made.

The symptoms disappeared with treatment with peroral amoxicillin clavulanate and nasal decongestant spray. However, the same clinical picture recurred 2 times within the following 5 months. During the last episode, the patient reported constant tearing in her right eye. There was no fluid passage on lacrimal irrigation. Dacryocystography demonstrated complete obstruction at the junction between the lacrimal sac and the nasolacrimal duct (Fig. 1B).

External dacryocystorhinostomy and anterior ethmoidectomy were performed. Macroscopically, the lacrimal bone seemed to be intact at the time of surgery. No foreign body was noted in the sac. The histopathologic examination showed nonspecific inflammation, vascular congestion, and edema in the sac wall. No complication or recurrence of epiphora developed during a postsurgical follow-up period of 21 months.

**DISCUSSION**

Acute dacryocystitis is mostly secondary to acquired idiopathic nasolacrimal duct obstruction. Other conditions that can cause acute dacryocystitis are lacrimal sac tumors, sac dacryoliths, diverticuli, and ethmoidal mucocele. There are few reports of pseudodacryocystitis secondary to anterior ethmoiditis in the literature. Remulla et al. described 2 pediatric patients who were 4 and 11 years old. In these cases, ethmoiditis recurred several times, which led to abscess formation in the sac region masquerading as chronic dacryocystitis. One of these children was successfully treated with systemic antibiotics and the other with external ethmoidectomy plus an antibiotic. Although these cases had epiphora during the acute exacerbations, they did not progress to obstruction of the nasolacrimal duct during the follow-up period. Shvero et al. reported a case of ethmoiditis in an adult who presented with bilateral fistulae above the medial canthus, with no clinical signs of nasolacrimal duct obstruction.

Several studies have suggested a relationship between sinonasal disorders and acquired idiopathic nasolacrimal duct obstruction. In their CT study, Kallman et al. reported a significantly higher incidence of ethmoidal opacification, agger nasi cell opacification, and nasal septal deviation in patients with nasolacrimal duct obstruction compared with control subjects. Francis et al. reported anterior ethmoidal soft tissue opacification in 15 of 107 cases of dacryostenosis on preoper-
Anterior ethmoid air cells extend beyond the sutures of the ethmoid bone in most normal orbits. In 93%, the cells are anatomically adjacent to the lacrimal sac fossa, with 40% entering the frontal process of the maxilla.\(^7\) In our patient too, the ethmoid cells extended to the level of the lacrimal sac in both sides. Ethmoidal sinus infections may gain access to the orbit by direct extension through the thin ethmoidal or lacrimal bones, congenital bony dehiscence, or infected thromboemboli along the valveless venous connections between the orbit and paranasal sinuses.\(^8\) Recent clinicopathologic findings indicate that osteitis of the sinus walls may be responsible for both the spread of the local sinuses and the cases refractory to medical or surgical treatments.\(^9\) The periosteal sheets surrounding the lacrimal sac circumferentially, and possibly the lacrimal fascia, prevent the lacrimal sac from orbital infection in most cases. However, as the small dehiscences or holes in the lacrimal fascia allow the transmission of infection from the sac to the other tissues of the orbit, the reverse of this is also possible.\(^10\)

In the present case, the direct compression of the local abscess to the sac and nasolacrimal duct, extension of the inflammation from the adjacent sinusitis and perisaccal infection to the nasolacrimal duct or invasion of microorganisms in the duct, or a combination of these factors could play a role in the development of lacrimal obstruction. During episodes of pseudodacryocystitis, defects may develop in the lacrimal fascial barriers secondary to stretching and inflammation, which may allow passage of the microorganisms to the sac.

In patients who present with clinical features of acute dacryocystitis and patent lacrimal drainage system on irrigation, the differential diagnosis should include pseudodacryocystitis due to ethmoiditis. In these patients, the infection may recur despite systemic antibiotic treatment. Early ethmoidectomy should be considered to prevent recurrence of the infection and progression to nasolacrimal duct obstruction.

**REFERENCES**


**Presumed Primary Orbital Neuroblastoma in a 20-Month-Old Female**

Nanfei Zhang, M.D., and Lily Koo Lin, M.D.

**Abstract:** Neuroblastoma is the most common malignant disease of childhood, and it often arises from either the adrenal gland or along the sympathetic chain. The authors report a case of a 20-month-old female with a presumed primary neuroblastoma of the orbit. Radiologically, the tumor showed a brightly enhancing mass lesion of the left side with intraorbital, temporal extraorbital, transspheenoid, and intracranial components. Histopathologically, the tumor was composed of small round blue cells. Immunohistochemical staining was positive for neuron-specific enolase. To the best of the authors’ knowledge, primary orbital neuroblastoma has only been previously reported once in children, although it has been reported twice in adults.

Neuroblastoma is the most common extracranial solid malignancy of childhood. The tumor is thought to arise from the neural crest and can occur in the adrenal gland or anywhere along the sympathetic chain in the retroperitoneum or mediastinum. Secondary orbital involvement occurs in approximately 20% of patients, and it is sometimes the initial manifestation of tumor.\(^1\)

**CASE REPORT**

A 20-month-old female presented for a work-up of child abuse with a 2-week history of periocular ecchymosis and swelling of her left eye. There was an inconsistent history of left eye trauma against a sharp object. On eye examination, she was able to fix and follow with each eye. The left eye showed significant proptosis with resistance to retropulsion and limitation of motility in superior and lateral gazes (Fig. A). The pupils were equally round and were reactive to light. No relative afferent pupillary defect was present. Intraocular pressures were 16 mm Hg OD and 22 mm Hg OS. Subconjunctival hemorrhage was present supersonally and inferotemporally in the left eye. In addition, diffuse punctate epithelial keratopathy was present in the interpalpebral fissure area, consistent with chronic exposure keratopathy due to the lagophthalmos resulting from the degree of proptosis.

The MRI of the orbit and brain showed a brightly enhancing mass of the left side with intraorbital, temporal extraorbital, transspheenoid, and intracranial components. There was enhancement of the dura extending over the lateral aspect of the left frontal lobe and proptosis of the left globe with downward and medial deviation of the muscle cone. The mass in the coronal plane measured approximately 3.4 cm in horizontal and vertical dimensions (Fig. B). The patient underwent a lateral orbitotomy for biopsy of the most easily accessible mass.

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The hematoxylin-eosin stain revealed sheets and nests of small blue cells with high nuclear-to-cytoplasmic ratios and hyperchromatic nuclei infiltrating through bone and skeletal muscle (Fig. C). Immunostained slides revealed positive cytoplasmic staining for neuron-specific enolase, consistent with neuroblastoma (Fig. D).

A bone marrow biopsy showed abnormal hypocellular bone with trilineage hematopoiesis and small clusters of foreign cells, consistent with metastatic neuroblastoma. Peripheral blood smear showed mild normocytic anemia and mild left shift in the granulocytes.

Abdominal and chest CT failed to reveal any other masses. Urine homovanillic acid and vanillylmandelic acid levels were within normal limits. Meta-iodobenzylguanidine scintiscan scan and bone scan were negative for sites of metastasis.

Due to the unfavorable factors, including age at diagnosis, clinical stage 4 disease, and histopathologic classification in the unfavorable histology group, the patient was treated according to the high-risk protocol. Subtotal resection of the tumor was performed by neurosurgery with removal of orbital roof, including dura, sphenoid bone, orbital groove, supraorbital rim, lateral orbit, zygoma, and the soft tissue tumor involving the orbit and temporalis muscle. Pathology specimens again demonstrated neuroblastoma cells of poorly differentiated subtype and a failure to identify a primary tumor site. The patient received 5 courses of chemotherapy consisting of cisplatin, etoposide, vincristine, cyclophosphamide, doxorubicin, and mesna. Repeat bone marrow biopsy was free of malignant cells. She underwent autologous stem cell bone marrow transplantation. At 12-month follow-up, no primary site other than the orbit has been identified. She continues to be followed by the pediatric hematologist and oncology, neurosurgery, and ophthalmology services.

**DISCUSSION**

Like other cases of neuroblastoma in the literature, the initial focus of the work-up was suspected child abuse. The team had not recognized the concerning degree of proptosis until it was brought to their attention by the ophthalmology service. Even then, there was reluctance in the team to drop the child abuse work-up. Once the results of the MRI, which ophthalmology had ordered, were available, the management took a different turn.

The intriguing aspect of this case was the primary site being the orbit in a 20-month-old female. The bone marrow biopsy showed malignant cells consistent with neuroblastoma, but no other primary site other than the orbit could be identified through CT chest, CT abdomen, metaiodobenzylguanidine scintiscan, and bone scan during her follow-up period of 1 year. This tumor did not secrete high levels of vanillylmandelic acid or homovanillic acid,
Isolated Cavernous Hemangioma of Conjunctiva

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Abstract: A 70-year-old Indian female presented with a painless mass in the left eye medially since 5 years. It increased gradually to its present size and was constant over the past 2 years. There was no change in color and no bleeding from the mass. Cosmesis was the only concern.

In the conjunctiva and caruncle. Because of this rarity, an interesting clinical report of this condition is reported.

Case Report

A 70-year-old female presented with a painless mass in the left eye medially since 5 years. It increased gradually to its present size and was constant over the past 2 years. There was no change in color and no bleeding from the mass. Cosmesis was the only concern.

On examination, a smooth, lobulated, oval, violet-colored mass measuring 5 × 4 × 3 mm appeared to involve both the caruncle and bulbar conjunctiva (Fig. 1). It was soft, vascular, compressible, and freely mobile. The swelling was neither reducible nor pulsatile and did not cause any proptosis or dystopia. Any syndromic associations and orbital extension of the lesion were ruled out. Excisional biopsy was performed, and histopathologic examination confirmed the diagnosis of cavernous hemangioma of conjunctiva involving the caruncle.

Hemangioma is a developmental malformation of blood vessels, rather than a true tumor, and is an example of a hamartoma. It may be capillary, venous (cavernous), or arterial. Cavernous hemangioma can appear at any age, consists of multiplicity of venous channels of varying caliber, shows no tendency to involution, and may become larger and troublesome by bleeding after remaining stationary for years. It is mostly seen in the orbit and eyelid and is relatively uncommon in adults twice and in a child once, to the best of our knowledge, this is the first report in the U.S. literature of such a tumor in this age group.

Our case reinforces the important role of the ophthalmologist in recognizing proptosis, which thwarted significant delay in diagnosis in this case, and in monitoring response to treatment and managing ophthalmic complications.

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REFERENCES


FIG. 1. A round, vascular, freely mobile, compressible mass arising from the bulbar conjunctiva and involving the caruncle is shown.
on the lids healed. The patient underwent cataract surgery 2 months later with good gain in vision. Since then, the patient has been followed up with no recurrence at 1 year of follow-up, and her cosmetic appearance is normal.

**DISCUSSION**

Cavernous hemangioma is the most common benign orbital tumor in adults. Eyelid is a common site, but conjunctival cavernous hemangiomas are a rarity, and to our knowledge, no case report of cavernous hemangioma of the caruncle is reported. In a series of 1,643 conjunctival tumors, less than 1% were cavernous hemangiomas. Conjunctival cavernous hemangioma arises often from the conjunctival vessels and rarely from the scleral, muscular, or orbital vessels. It is asymptomatic and stationary for many years but gives poor cosmetic appearance and sometimes bleeds, giving rise to bloody tears.

The present case was asymptomatic, except for the cosmetic factor. Total excision was possible, and histopathologic examination confirmed the diagnosis. There was no recurrence on 1 year of follow-up.

Only 3 cases of isolated cavernous hemangioma of the conjunctiva have been reported. Rao et al. reported a similar case that was confirmed on excisional biopsy, although caruncular involvement has not been commented upon. A young male with a bulbar cavernous hemangioma was reported by Koo et al. Another report of isolated pedunculated hemangioma of the conjunctiva exists in literature. A case of blue rubber bleb nevus syndrome was noted in an infant who had cutaneous, conjunctival, retinal, and intestinal cavernous hemangiomas. This is uncommon in adults.

Our patient had isolated cavernous hemangioma of the caruncle and conjunctiva, without orbital involvement. To our knowledge, no reports of caruncular cavernous hemangioma have been reported. Orbital involvement should be ruled out to confirm the extent of the lesion. Treatment in patients with orbital involvement includes orbitotomy or transconjunctival approach, and treatment in patients with isolated caruncular involvement includes excisional biopsy.

**REFERENCES**


**Effects of Reflected CO₂ Laser Energy on Operative Field Materials: Risks to Patients and Operating Room Personnel**

**Abstract:** This study investigates the effects of specularly and diffusely reflected CO₂ laser energy on operative field materials. The CO₂ laser was reflected off the sandblasted and polished surfaces of an eyelid plate and a wet cotton gauze pad. The laser was aimed at a surgical glove, operative gown, laser safety goggles, and endotracheal tube, each positioned 5 cm, 10 cm, and 15 cm from the plate. Primary outcomes were time to initial effect and description of result. There was no reflection off wet gauze and no effect on the glove itself. When reflected off sandblasted and polished surfaces, the laser created a hole in the glove and a flame in the surgical gown. When targeting safety goggles and an endotracheal tube, the laser created a surface divot at short distances and surface irregularity at 15 cm. While the CO₂ laser is an excellent surgical instrument, reflected laser energy can affect operative field materials.

The CO₂ laser has been used in multiple surgical specialties for decades, including oculoplastic, plastic, dermatologic, and gynecologic surgeries. Advantages of laser use include improved intraoperative hemostasis, clearer view of relevant anatomy, decreased operating time, and improved appearance in the immediate postoperative period. The safety of CO₂ laser for medical procedures has been extensively explored.

Instruments are made “laser safe” by altering their surfaces through sandblasting, ebonization, and anodization. Driver and Taylor compared the power densities of reflections off samples of stainless steel that were polished, roughened, and/or chemically blackened. Polished steel reflected most energy, followed by blackened surface, then shot-blasted surface, and finally a combination of blackened and shot-blasted materials.

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This study investigates effects of CO₂ laser reflections off a "laser-safe" surgical instrument on several operative field materials.

**METHODS**

A Sharplan 40c CO₂ laser (Lumenis, Inc., Santa Clara, CA, U.S.A.) was set at the typical 6.0 W in continuous-wave mode for incisional surgery. The hand piece was fixed at 30° from the normal of the following reflective surfaces: wet cotton gauze, polished handle and sandblasted working area of a used but sterilized Jaeger eyelid plate (Oculo-Plastik Inc., Montreal, Canada). A Jaeger eyelid plate is generally used to protect ocular/periocular tissues from misdirected direct laser energy. The instrument was inspected under 2.5× magnification after each trial to assess for surface changes. The laser was kept in pinpoint focus on the reflective surface. Target materials included a surgical glove, gown, CO₂ safety goggles (OD6 at 200–360 nm, 10,600 nm), and endotracheal tube, each fixed at 5 cm, 10 cm, and 15 cm from the reflective surface. For each material, the time to observable change and a description of the change were recorded for 3 trials at each test distance. Analysis of variance techniques were used to assess the significance of the variation of the time to effect off the different reflective surfaces.

**RESULTS**

No change was observed on the surface of the wet gauze after 30 seconds of continuous laser firing. No alterations in the sandblasted or polished surfaces of the Jaeger plate were observed throughout the experiment.

For all surfaces except the goggles, the polished reflective surface created visible changes more quickly (Table). No change was appreciable after 15 seconds of continuous laser firing for both the goggles and endotracheal tube at 15 cm. Neither reflective surface created a consistently larger defect. Effects of the CO₂ laser energy on target materials are displayed in the Figure.

**DISCUSSION**

This study demonstrates a clear risk of damage to operative field materials from reflected laser energy off both the sandblasted and the polished surface of Jaeger eyelid plate. At 5 cm from a "laser-safe" reflective surface, the reflected beam burned a hole in a surgical glove in less than 2 seconds of laser firing. The time to initial damage was shorter with the polished reflective surface for all materials but the goggles at 10 cm. Initial changes were difficult to visualize on the goggles' clear surface. This difficulty may have contributed to observer error in timing. Future investigations may use thermometric monitoring to minimize this error. Because no damage to or reflection from dampened gauze was noted after more than 30 seconds of laser firing, these materials may offer protection in some clinical situations.

**Results of analysis of variance comparison of time to initial visible change in target material with laser reflected off the polished versus the sandblasted surface**

<table>
<thead>
<tr>
<th>Target material</th>
<th>Distance (cm)</th>
<th>Polished surface Mean time to initial change (seconds)</th>
<th>Sandblasted surface Mean time to initial change (seconds)</th>
<th>Difference</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Glove</td>
<td>5</td>
<td>1.09</td>
<td>1.64</td>
<td>0.55</td>
<td>1.000</td>
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<tr>
<td></td>
<td>10</td>
<td>1.88</td>
<td>3.95</td>
<td>2.07</td>
<td>0.004</td>
</tr>
<tr>
<td></td>
<td>15</td>
<td>5.39</td>
<td>7.50</td>
<td>2.11</td>
<td>0.003</td>
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<tr>
<td>Surgical gown</td>
<td>5</td>
<td>0.73</td>
<td>1.25</td>
<td>0.52</td>
<td>1.000</td>
</tr>
<tr>
<td></td>
<td>10</td>
<td>1.31</td>
<td>3.02</td>
<td>1.71</td>
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<tr>
<td></td>
<td>15</td>
<td>2.83</td>
<td>9.24</td>
<td>6.41</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Goggles</td>
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<td>1.37</td>
<td>2.54</td>
<td>1.17</td>
<td>0.702</td>
</tr>
<tr>
<td></td>
<td>10</td>
<td>4.64</td>
<td>4.44</td>
<td>-0.020</td>
<td>1.000</td>
</tr>
<tr>
<td>Endotracheal tube</td>
<td>5</td>
<td>1.00</td>
<td>1.11</td>
<td>0.11</td>
<td>1.000</td>
</tr>
<tr>
<td></td>
<td>10</td>
<td>3.10</td>
<td>12.31</td>
<td>9.21</td>
<td>&lt;0.001</td>
</tr>
</tbody>
</table>

A, Surgical glove with full-thickness holes from the CO₂ laser. B, Surgical gown material with full-thickness holes and charred areas from the CO₂ laser. C, Plastic safety goggles with divots on the surface from the CO₂ laser. D, An endotracheal tube showing a large surface divot from the CO₂ laser.

seconds of continuous laser firing, we recommend replacing the Jaeger plate with a nonspecular wooden tongue blade wrapped in damp gauze. This implement may be used to protect the cornea, globe, or any underlying or adjacent tissues during any procedure that involves laser energy.

No correlation was found between reflective surface and damage size. The surface of the Jaeger eyelid plate was curved, which could create unpredictable diffuse reflections and explain variability in defect size. This effect was minimized by aiming the laser at a fixed location on both the polished and sandblasted surfaces. The curvature of target materials could affect defect size. The gown and gloves were stretched to create flat target surfaces, while the goggles and endotracheal tube were both curved. Although care was taken to orient targets perpendicular to the angle of incidence, any variation could lead to differences in the angle of impact and thus in the size of the created defect. These curved surfaces may be flattened to minimize this effect in future studies.

Our experiment assessed the effect of reflected CO₂ laser energy on common operative field materials. Although “laser-safe” instruments should be used during CO₂ laser procedures, surgeons and support staff should be aware that laser energy reflected from these instruments may still damage operative field materials and personnel. This study supports the use of a tongue blade wrapped in damp gauze to prevent such damage.

REFERENCES