Epiphora as a Presenting Sign of Angioleiomyoma of the Lacrimal Sac
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Abstract: A 50-year-old man presented with progressive enlargement of a right medial canthal mass for 5 years. The patient subsequently noted epiphora of 6-month duration on the affected side. An anterior orbitotomy was performed, which uncovered a well-circumscribed mass in the lacrimal sac. Histologic sections of the mass revealed an angioleiomyoma. Following complete excision, the epiphora resolved and there is no evidence of tumor recurrence at 1 year of follow-up. To the best of our knowledge, this is the first report of an angioleiomyoma in the lacrimal sac.

A ngioleiomyoma or vascular leiomyoma is a benign tumor composed of smooth muscle cells and vascular endothelium.1 The majority of angioleiomyomas have a predilection for the lower extremities and typically presents as a solitary, painful, cutaneous mass.2,3 Approximately 8.5% to 10% of angioleiomyomas present in the head and neck region as a painless mass.4-6 In the orbit, there have been only 2 reported cases of angioleiomyoma.7,8 To date, there have been no cases of angioleiomyoma in the lacrimal outflow system. As far as we are aware, this is the first reported case of angioleiomyoma in the lacrimal sac.

CASE REPORT

A 50-year-old man was referred with right-sided epiphora of 6-month duration. Five years prior to presentation, the patient reported gradual enlargement of a “cyst” located at the right medial canthus. He had a history of an old orbital blowout fracture in the right orbit at the age of 13. The patient denied a history of pain, ulceration, redness, mucoid discharge, or hemorrhagic tearing. His medical history was significant for type II diabetes that was well controlled with metformin and glipizide. There was no prior history of surgery or tobacco use.

On clinical examination, visual acuity measured 20/20 OU. Pupillary examination was unremarkable and ocular ductions were full. Exophthalmometry revealed no asymmetry. Slit lamp examination and dilated fundoscopy were normal. External examination revealed a firm, dark-colored, nonmobile mass at the right medial canthus (Fig. A). The lesion measured approximately 10 mm externally, and with palpation no discharge was noted. Lacrimal irrigation through the right inferior punctum revealed complete reflux through the superior punctum.

A curvilinear incision at the right medial canthus similar to an external dacryocystorhinostomy was performed. A subperiosteal dissection plane was created, and the lacrimal sac was reflected. The lacrimal sac was noticeably distended by the mass. A vertical incision was made in the lacrimal sac and a nodular mass was noted. Blunt dissection was used to completely excise the lesion (Fig. B). Gross examination of the lesion revealed a red-brown, irregular mass measuring 1.0 cm × 0.8 cm × 0.5 cm (Fig. C).

Histopathologic analysis of the lesion showed a confluence of vascular channels among dense bundles of muscle fibers (Fig. D, E). A large thrombosis was present in a central vessel. Figure 1F shows immunostaining with smooth muscle actin that confirms the smooth muscle origin of the tumor, while HMB-45 immunostaining was negative (data not shown). Based on the histopathologic findings, the lesion was most consistent with angioleiomyoma.

After complete excision of the tumor, epiphora resolved and there was no evidence of recurrence at 1 year of follow-up.

DISCUSSION

Leiomyoma is an uncommon tumor of the orbit and has been well described in the literature.9 Angioleiomyoma is a rare subset of leiomyoma and is distinguished by the predominance of endothelium-lined vascular channels among a background of smooth muscle.1 In 1965, Wolter reported the first case of orbital angioleiomyoma in a 41-year-old man with proptosis. In 1970, Henderson and Harrison reported the second case of orbital angioleiomyoma in a 9-year-old girl with upward displacement of the right globe. Our case is the third report in the literature, but is the first report of an angioleiomyoma in the lacrimal sac causing epiphora.

Smooth muscle is relatively rare in the orbit. Given that angioleiomyoma is postulated to arise from the smooth muscle, possible tissues of origin include orbital blood vessels, periocytes, Müller muscle, or the capsulopalpebral muscle of Hes-
It is easy to envisage the origin of the previously reported cases of angioleiomyoma in the orbit, but in our case, the identification of this tumor in the lacrimal sac cannot be easily explained.

In addition to serving as a conduit for tears through the nasolacrimal duct, the lacrimal sac is believed to have the ability to resorb tears. The epithelium of the lacrimal sac is lined by an extensive microvillar network with a vascular plexus that is continuous with the cavernous body of the inferior turbinate. In our case, the angioleiomyoma may have arisen from the smooth-muscle lining of this cavernous vasculature.

Angioleiomyoma is subdivided in 3 histopathologic subtypes: capillary or solid, cavernous, and venous. The presence of a blood-filled central thrombus, exuberant proliferation of smooth muscle, and the well-circumscribed nature (Fig. D, E) classify this lesion as the cavernous subtype. Ferry and Kaltreider reported a case of cavernous hemangioma in the lacrimal sac that demonstrated the typical gross and histopathologic features of this tumor. However, the diagnosis of angioleiomyoma in our case is solidified by the strong immunoreactivity with smooth muscle antigen (Fig. F). In addition, this adnexal variant of angioleiomyoma stained negatively for HMB-45 compared with the positive immunoreactivity with this marker in the retroperitoneal and renal variants of angioleiomyomas.

Standard practice patterns for a medial canthal lesion of the lacrimal system are complete excision confirmed by histopathologic examination. Owing to the potential malignant nature of the patient’s presentation with a medial canthal lesion, our goal was to excise the lesion without reconstruction by dacryocystorhinostomy or silicone stent placement. Interestingly, the lacrimal system remained patent after removal of the angioleiomyoma despite no intubation of the lacrimal system. The prognosis of angioleiomyoma is excellent. The tumor is not radiosensitive and adjuvant external beam radiotherapy after incomplete excision is not advised. Recurrence after incomplete excision has been reported, but in the absence of radiotherapy, there have been no reported cases of malignant transformation.

The treatment of choice remains complete surgical excision.

In summary, this is the first known case of angioleiomyoma in the lacrimal system. Complete surgical excision lead to resolution of all symptoms with no evidence of recurrence. Clinicians should be aware of angioleiomyoma when considering the differential diagnosis of medial canthal masses.

REFERENCES

Retained Silicone Sleeve as a Cause of Dacryocystorhinostomy Failure

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Abstract: Silicone sleeves are 1 method of stabilizing bi-
canalicular silicone intubation near the ostia during
dacryocystorhinostomy surgery to prevent tube prolapse.
Retained sleeve following dacryocystorhinostomy is a
rare and previously unreported complication that can
occur despite endoscopic visualization during tube re-
moval, particularly in narrow nasal passages. It is an
easily reversible cause of dacryocystorhinostomy failure
if identified.

Failure of dacryocystorhinostomy (DCR) surgery is most
commonly due to cicatrizing closure of the rhinostomy or
distal common canaliculus. Silicone intubation in DCR aims
to prevent early membrane formation at the internal canalicular
opening. One of the commonest complications related to can-
icular silicone intubation is tube prolapse in the interpalpebral
space,1,2 and this may be prevented by stabilizing the tubes
near the ostia with knots,1,3 suture ties,2 Ligaclips,4 silicone
sleeves such as the retinal-type Watzke sleeve,7,5 or a combi-
nation of these. Inadvertently retained silicone tubing has been
reported as a cause of persistent epiphora after DCR.5,7 How-
ever, the potential for retained tube-stabilizing sleeves to cause
DCR failure following successful retrieval of the silicone tubes
has not been reported previously.

We report 2 cases of DCR failure due to retained sleeves
following successful tube removal, and highlight the import-
ance of nasal endoscopy in the assessment of such patients.

CASE REPORTS

Case 1. A 35-year-old woman was referred with a 2-year
history of bilateral epiphora, with no improvement following
3-snip punctoplasty. She was found to have bilateral nasolac-
rimal duct stenosis and a narrow left nasal passage because of
septal deviation. She underwent bilateral segmental endoscopic
DCR with bicanalicular intubation and stabilization of tubes
with a size-10 silicone sleeve and Ligaclips. Left epiphora
improved only 50% following retrieval of tubes, and 4 months
postoperatively she was found to have delayed tear clearance and a
delayed fluorescein endoscopic dye test. She was patent to
syringing with no resistance or regurgitation, but found syringing
uncomfortable. Because of nasal septal deviation, no view of the
left internal ostium was possible. She underwent a septoplasty to
allow endoscopic visualization of the left internal ostium and at the
time a retained silicone sleeve was found (Fig. A) and removed
(Fig. B). Her epiphora subsequently improved.

Case 2. An 80-year-old man presented with left-sided epiphora
secondary to complete nasolacrimal duct obstruction. He un-
derwent an uncomplicated left external DCR with bicanalicular
silicone intubation stabilized with a silicone Watzke sleeve. His
epiphora improved significantly, and tubes were removed un-
der endoscopic guidance at 3 months. The internal ostium was
not viewed at the time because of a narrow nasal passage and
scar tissue. He presented 7 months postoperatively with recur-
rent epiphora, and was found to have a positive fluorescein dye
retention test and syringing indicative of distal common cana-
licular stenosis. He subsequently underwent balloon dacryocys-
toplasty, and at the time of surgery was found to have a retained
Watzke sleeve at the internal ostium that was removed along
with scar tissue. Bicanalicular silicone tubes were inserted and
stabilized with Ligaclips. His epiphora had completely resolved
at 6-weeks follow-up.

DISCUSSION

Proximal stabilization of tubes near the ostia helps prevent
lateral tube migration. Knots and silk ties are well-established
means for securing tubes.1,3,5,7,9,10,11 Ligaclips3 and Watzke sleeves2,5
are more recent innovations for this purpose. Removal of tubes
is generally undertaken at 1 month to 3 months after surgery,
after the ostia has fully healed. Unless an endonasal endoscopic
examination is carried out at this stage with visualization of the
internal ostium, it is possible to leave behind a sleeve. The
above cases emphasize the importance of confirming retrieval
of all tube products and disputes the assumption that retained
sleeves may simply become swallowed. The inadvertent reten-
tion of tube products or any foreign stabilization device near

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