



Orbital and periorbital migration of silicone oil associated with emphysema development after retinal detachment repair – Case report and literature review

Ahmed Algethami^{a,b}, Silvana Artioli Schellini^c, Sahar M. Elkhamary^{d,e}, Mohammad A. Talea^f, Valmore A. Semidey^{a,*}

^a Vitreoretinal Division, King Khaled Eye Specialist Hospital, Riyadh, Saudi Arabia

^b Eye Center, King Abdullah Medical City, Makkah, Saudi Arabia

^c Medical School, State University of Sao Paulo, Sao Paulo State, Brazil

^d Diagnostic Imaging Department, King Khaled Eye Specialist Hospital, Riyadh, Saudi Arabia

^e Department of Diagnostic Radiology - Mansoura Faculty of Medicine, Mansoura, Egypt

^f Saudi German Hospital, Aseer, Saudi Arabia

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ABSTRACT

Purpose: To report a very rare case of silicone oil (SO) migration and emphysema development in the orbit and periorbital tissue, including the lids and subconjunctival space, after a fourth pars plana vitrectomy (PPV) for retinal detachment (RD) treatment.

Observations: A 53-year-old woman with a recurrent rhegmatogenous RD in the right eye underwent a fourth PPV under local anesthesia and 23-gauge vitrectomy with fluid-air exchange and SO injection. Localized choroidal detachment occurred during fluid-air exchange near the end of the surgery. High-pressure infusion of air was used as a temporary control measure prior to SO injection. In the early postoperative period, the patient developed hemifacial and periorbital swelling and the air trapped in the upper lid was associated with lid ptosis and conjunctival chemosis. The emphysema resolved with clinical management, and the mechanical ptosis subsided after partial SO removal from the lid.

Conclusions and Importance: The SO migration and emphysema in our case were presumably related to the multiple previous sclerectomies. Periorbital emphysema can show spontaneous resolution, but the migrated SO requires surgical management.

1. Introduction

Silicone oil (SO) has been used as intraocular tamponade after vitrectomy to repair complex retinal detachment (RD) since the 1960s.¹ This material is very stable, nontoxic, and insoluble in body fluids. However, the use of SO can lead to complications, including cataract, glaucoma, SO emulsification, corneal decompensation, subretinal SO migration, fibrous epiretinal or subretinal proliferations, and migration to the neck, nasopharynx, or through the optic nerve to the cerebral ventricles and subarachnoid space.^{2,3,4}

Subcutaneous emphysema is another complication of vitrectomy. Emphysema is defined as gas or air trapped in the subcutaneous tissue plane. Air can escape from the eye during vitrectomy at the time of fluid-

air exchange. However, orbital emphysema more frequently results from orbital/sinus fractures or can occur also after surgical procedures such as balloon dacryoplasty, orbital decompression, maxillofacial trauma repair, tracheostomy, dental or sinus procedures, positive-pressure mechanical ventilation, bronchopleural fistula, infections, and compressive air injuries.³⁻⁵ Although orbital emphysema can be potentially vision-threatening, periorbital subcutaneous emphysema is usually a self-limiting condition.

In patients who undergo SO injection after fluid-gas exchange in a vitrectomy, the SO may migrate with or without emphysema association. However, migration of the SO to the subconjunctival space reaching the periorbital tissues and lids is a very uncommon complication of retinal procedures. Only 13 previously reported cases have

* Corresponding author. Vitreoretinal Division, King Khaled Eye Specialist Hospital, Al- Oruba Street, PO Box 7191, Riyadh, 12329, Saudi Arabia.

E-mail address: asemidey@hotmail.com (V.A. Semidey).

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described SO migration to the lids, and only two were associated with emphysema-inducing lid mechanical ptosis.²⁻¹³ These cases are summarized in Table 1.

We present a literature review and describe the third case of diffuse migration of SO into the subconjunctival, orbital, periorbital, and upper lid spaces that was associated with orbital and hemifacial emphysema in a patient with a history of multiple pars-plana vitrectomies (PPVs). We also discuss the clinical aspects, pathogenic mechanisms, and outcomes after treatment.

2. Case report

A 53-year-old woman presented to our center, with a history of ocular surgery for phacoemulsification with intraocular lens implantation and three previous pars plana vitrectomies (PPVs) with SO injection for a recurrent RD in the right eye (OD). The best-corrected visual acuity (BCVA) after the third RD repair was 2/200 OD. She subsequently presented to the retina clinic with a recurrent RD in the setting of inferior proliferative vitreoretinopathy (PVR). A fourth surgery was planned for

Table 1
Studies reporting migration of silicone oil placed as tamponade after pars plana vitrectomy.

Author	Age	Sex	Symptoms	Type of retina surgery	Previous eye surgery	Exams	Histological exam	Treatment	Source of tamponade migration
Quintyn et al., 2003 [7]	57	M	Lump in the upper lid 19 years after retina surgery	Vitrectomy with external drainage of subretinal fluid and SO injection	Two previous RD surgeries	CTscan: homogeneous thickening of the eyelid with density identical to the vitreous cavity silicone	Silicone cysts surrounded by collagen fibrosis associated with fibro-collagen reaction	Debulking with ptosis improvement	Sclerotomy site for external drainage of subretinal fluid. Elevated IOP
Donker et al., 2005 [8]	66 68	M	Swelling and redness of the upper lid 6 months after retina surgery Swelling, redness and ptosis of the upper lid starting one year after retina surgery	SO and scleral buckle removal	Vitrectomy with scleral buckle and SO injection 8 years prior Vitrectomy and SO injection twice 12 and 17 months prior	Many cysts in the orbicularis and pre-aponeurotic fat Diffuse swelling, redness and upper lid ptosis. Cysts in the epibulbar conjunctiva	Histiocytic cells with foamy cytoplasm and vacuoles suggestive of histiocytic reaction to SO Lipogranulomatous inflammation in the lid tissues	Ptosis repair Blepharoplasty and debulking	Trapped SO in epibulbar space previously occupied by the buckle Leakage from sclerotomy during or after surgery
Santaella et al., 2011 [9]	48	F	Ptosis, edema, "Xanthelasma"	Phacoemulsification and SO removal	Vitrectomy with scleral buckling and 5000 centistoke SO injection	Cysts in the lid and subconjunctival space	Skin, orbicularis muscle, preaponeurotic fat and conjunctiva with vacuoles, scattered aggregations of histiocytic cells with foamy cytoplasm, suggestive for a histiocytic reaction to SO.	Cysts removal, ptosis repair	Not specified
Damasceno et al., 2014 [4]	55	M	Creptation in soft tissues after retina surgery	23-gauge PPV with fluid-gas exchange	Traumatic orbital floor fracture	CTscan: bilateral emphysema in the face, orbit and mediastinum	Not done	Systemic antibiotics and steroids	High gas pressure during fluid gas exchange associated to orbital fracture
Lee et al., 2014 [10]	30	M	Pain and recidive of endophthalmitis suspected	Pars plana vitrectomy and SO injection	Endogenous endophthalmitis	MRI: shrinkage of the eyeball and large subconjunctival and orbital mass	SO globules with inflammatory cellular infiltration, with SO droplets surrounded by giant cells	Exeresis of subconjunctival and orbital mass	Leakage of SO through melted sclera
Deguchi et al., 2014 [11]	65	F	Swelling of the upper lid, ptosis and SO in the subconjunctival space 2 month after retina surgery	SO removal and cyst excision	Two 20-gauge vitrectomies with Scleral buckling for RRD with PVR.	Cyst in the peribulbar space, upper lid and subconjunctival	Granulomatous reaction. Macrophages with clear vacuoles and SO deposits. Fibrosis.	Removal of SO cysts from the subconjunctival space and lids	Suturing site for scleral buckle fixation
Asnani et al., 2015 [5]	77	M	Crepitation and bilateral face emphysema extending up the chest	Scleral buckle + PPV + SO injection	Globe perforation during peribulbar anesthesia with vitreous hemorrhage and RD	Face, neck and extending up the chest	Not done	Observation	24-gauge needle perforation during peribulbar anesthesia
Osaki et al., 2015 [12]	63	F	Painless mass and ptosis upper lid		PPV + Silicone oil injection followed by 8 month previous SO removal.	CT scan: hyperdense, lobulated lesions in the upper lid. Ultrasound biomicroscopy: cystic formations with anechoic content underneath the muscular layer of the upper eyelid	A well-delimited mass with pseudocysts and fibrosis within a fat tissue. Mild chronic inflammatory infiltrate. Probably pseudocysts were previously filled with SO.	Excisional biopsy, blepharoplasty, ptosis repair	Not addressed
Iniesta-Sanchez	40	F	Low visual acuity, upper lid ptosis, superior and inferior	Scleral buckle +23-gauge PPV + 18% SF6	ECCE + Congenital glaucoma	Ct scan: gas palpebral, subconjunctival, intraconal,	Not done	Hyperbaric oxygen therapy. Orbital	Leakage of gas through sclerotomies

(continued on next page)

Table 1 (continued)

Author	Age	Sex	Symptoms	Type of retina surgery	Previous eye surgery	Exams	Histological exam	Treatment	Source of tamponade migration
et al., 2016 [6]			lid edema, proptosis, chemosis five days after retina surgery			retrobulbar, compressing optic nerve. Exophthalmus		decompression with gas aspiration	
Dehagani et al., 2017 [2]	36	F	Ptosis, periorbital swelling and lumps attached to the skin five month after retina surgery	23-gauge sutureless PPV + 1300 cs SO injection.	Car accident with blunt ocular trauma	CT scan showed an 11 mm × 8 mm soft tissue density mass in the lateral aspect of the orbit	Not done	Excision of the SO material with improvement of the ptosis	Unstitched Sclerotomy
Powers et al., 2018 [3]	68	M	Ptosis and yellowish mass in the upper lid (fat prolapse?)		Baerveldt implant. PPV + SO for RRD. SO and Cataract removal. Vitreous washout for emulsified SO	Multiple spherical, encapsulated, translucent foreign bodies in the anterior orbital fat, extended diffusely throughout the upper eyelid with extension into the orbicularis and subcutaneous and anterior orbit. SO in the nasopharynx and nasal cavity	Several large foreign body granulomas surrounded by vacuolated macrophages consistent with SO	Debulking and blepharoplasty	Migration of oil through Baerveldt implant
Al Rashed et al., 2020 [13]	56	F	Ptosis and Xanthelasma-like both right lids		Five previous vitrectomies for recurrent RD	Upper and lower right lid, subconjunctival OD	Marked fibrosis with several pools of SO, infiltrating the surrounding connective tissue and fat with no foamy histiocytes, excluding diagnosis of Xanthelasma	Ptosis repair, blepharoplasty, and excisional biopsy of the Xanthelasma-like lesion	Migration of SO through sclerotomies
Algetami et al., 2020 [present case]	53	F	Periocular and maxillary swelling, redness, crepitation and upper lid ptosis one day after retina surgery. Chemosis	23-gauge vitrectomy + Heavy SO injection	Three previous vitrectomies and phacoemulsification with intraocular lens	CT scan: multiple irregular-shaped partially confluent hyperdense areas similar to intraocular SO in the orbit and periocular	Dermis is infiltrated by variable size of empty vacuoles. Multiple large foamy macrophages engulfing empty vacuoles. Chronic granulomatous reaction secondary to SO injection	SO removal from the upper lid and subconjunctival space	Inadvertent opening of sclerotomies from previous surgeries

SO removal, PVR membrane peeling with fluid-air exchange, and SO reinjection.

The fourth PPV was performed under local anesthesia using 8 cc of a peribulbar block (bupivacaine + lidocaine + hyaluronidase) and a 25-gauge needle. The remaining peripheral vitreous was removed via a 23-gauge PPV, and the PVR membranes were removed. Fluid-air exchange was performed, and the subretinal fluid was drained through the original breaks. With an air -fill and during scleral depression, a bulge was seen throughout the equator and macula, and choroidal detachment was suspected. The infusion cannula and trocars were verified to be in the correct positions. Air infusion was elevated to 60 mmHg to increase the intraocular pressure (IOP), but the choroidal protrusion worsened. Immediately, 5.5 mL of heavy SO (Oxane HD; Bausch and Lomb) was injected into the eye, and after the tamponade effect, the retina and choroid were flat. All sclerotomies were secured using 7-0 Vicryl sutures, and subconjunctival dexamethasone (0.5 mL) and cefazoline (0.5 mL) were administered. At the end of the procedure, severe conjunctival chemosis made it difficult to close the lids, and the decision was to patch the eye.

In the first postoperative period, the patient showed severe upper and lower lid edema and severe ptosis with an upper lid margin-pupil reflex distance (MRD1) of -2 mm. The right side of the face presented with emphysema, which was characterized by soft crepitus and evidence of air loculi with no inflammatory signs or proptosis and was in the subcutaneous, probably in the intramuscular location. Conjunctival chemosis was more severe on the temporal side. The cornea and anterior segment were normal, IOP was 14 mmHg, and the retina was flat under

SO, with fresh laser scars and no sign of choroidal detachment or hemorrhage. A superior oil meniscus was visible.

A computerized tomography (CT) scan of the orbits showed multiple irregularly shaped partially confluent hyperdense areas, like the intraocular injected material, located in the extraconal and intraconal orbital spaces as well as in the anterior pre-septal orbit. The material was detected surrounding the ocular bulb and along the superior aspect of the distal right optic nerve (Fig. 1). The diagnosis was SO migration and orbital emphysema. Conservative management with artificial tears and anti-inflammatory drops along with cold compresses was initiated.

After one month, the retina remained flat, and the emphysema had been absorbed with no more crepitus. The lid swelling improved, with mild mechanical ptosis (MRD1OD = +2 mm; OS = +4; normal levator function). Swelling and conjunctival chemosis of the OD improved, but several small translucent round or oval cysts persisted mainly in the temporal subconjunctival space.

We decided to debulk the migrated SO from the upper lid and the periorbital temporal area. An incision in the upper lid crease was used, and multiple spherical, encapsulated, translucent SO-containing cysts approximately 1 mm in diameter were removed. Although similar SO cysts were also observed in the levator aponeurosis, it was normally attached to the tarsal plate. The subconjunctival loculated cysts from the temporal area were removed through a direct conjunctival incision. The material was sent for histopathological examination, and it revealed subcutaneous tissue including adipose tissue and muscle fibers infiltrated by vacuoles of variable size with multiple large foamy macrophages engulfing the empty vacuoles (Images 1,2). The same reaction

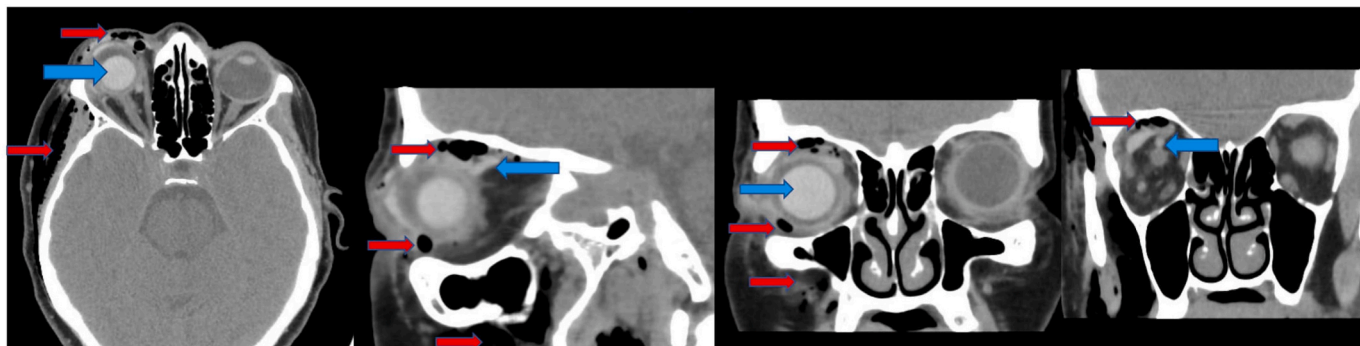


Fig. 1. Vitrectomy followed by intraocular silicone oil tamponade for treatment of retinal detachment: **a** - Axial computed tomography (CT) image shows an area of homogeneous hyperdense silicon oil (blue arrow) in the right globe. **b, c, d** - Sagittal and coronal non-contrast CT scans showing pre-septal, intraorbital, and extraconal silicone oil extravasation (blue arrows) close to the optic nerve superior surface (**d**) with episcleral thickening and distortion of the globe (**c**). Orbital emphysema, with the air appeared as black (red arrows) extending circumferentially preseptal, extraconal, extending along the facial planes into the right cheek. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

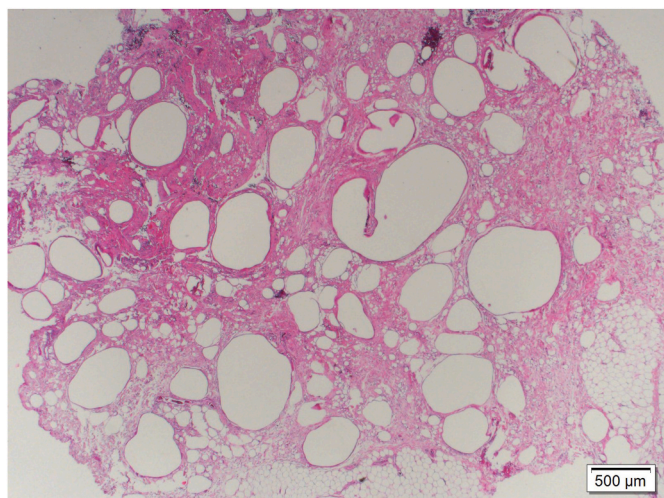


Image 1. Histologic section of subcutaneous tissue and conjunctiva filled by a variable size of empty vacuoles (silicone oil) infiltrating around the pilosebaceous units, muscle fibers, and sweat glands.

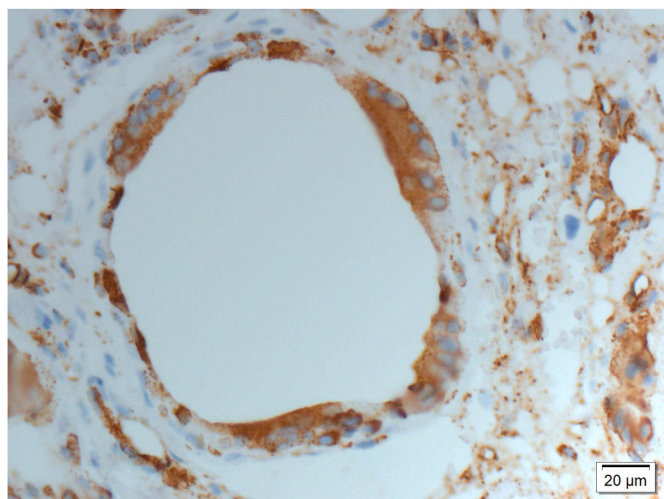


Image 2. Multiple large, foamy macrophages are noted engulfing the empty vacuoles representing chronic granulomatous inflammation secondary to silicone oil.

was observed in the subconjunctival cysts, leading to the diagnosis of chronic granulomatous reaction secondary to SO injection.

Further examination after three months did not reveal any more air or swelling in the periorbital area, but the lid remained thicker, and the mild upper lid ptosis persisted. Another procedure for SO removal from the lid and subconjunctival space was performed. The eye was stabilized over time, and the retina was attached to a final visual acuity of hand motion for OD.

3. Discussion

We present a rare case of SO migration and emphysema affecting the right upper and lower lids, periorbita, orbit, and the OD subconjunctival space after a fourth vitrectomy to treat a recurrent RD. The emphysema regressed in one month, but even with lid debulking, the persistence of the migrated SO induced mild mechanical ptosis.

Our patient showed simultaneous SO migration and emphysema, which was shown to affect all tissues near the eye on the first post-operative day. A non-inflammatory, painless swelling, and yellowish mass in the upper lid that may or may not be associated with chemosis secondary to SO migration has been reported to persist from 6 months⁸ to 19 years after retinal⁷ or glaucoma surgery.⁶ The diffuse swelling and skin changes can show the appearance of a xanthelasma or pseudo-xanthelasma to the upper lid,^{6,9,13} and the infiltration of the lid tissues induces mechanical ptosis.

The crepitus observed in almost the entire hemiface of our patient was a sign of air in the tissues, but the severe chemosis prevented us from observing air in the subconjunctival space. Only two previous cases of orbital emphysema following vitreoretinal surgery have been reported, and both involved more overflow than in our case, which affected the orbit, mediastinum, and face³ or periorbita bilaterally and caused cervicofacial subcutaneous emphysema.⁴

The SO and air migration in our case could have even occurred because of inadvertent eye perforation during the peribulbar anesthesia injection.^{4,6} However, we used a 25-gauge needle for anesthesia injection, which would have substantially limited the migration of heavy SO through the minimal opening. The large emphysema in our case also excludes the possibility of the migrated air originating from the peribulbar block or subconjunctival medication administered at the end of the procedure, since these syringes were carefully purged of air before administration.

A 23-gauge trocar system was used to perform the PPV. SO migration has been reported after PPV using 20-^{7,8,11} and 23-gauge needles,^{2,3,5} but most cases involved more than one previous intraocular surgical procedure, similar to the present case. In addition, SO leakage can result from inadequate or improper closure of sclerotomy,¹ as observed in one

case of suture-less vitrectomy and migration of SO from the vitreous space to the orbit.² However, in the present case, all the sclerotomies were secured because the patient was undergoing her fourth surgery, with at least nine previous sclerotomies and the sclera may have been thinner as a result.

The intraoperative choroidal elevation observed in the posterior pole during the air-fluid exchange and the scleral depressed revision of the peripheral retina almost at the end of our surgery subsided once SO was injected inside the eye. Although fluid-air exchange is an essential step in PPV, the infusion of positive air pressure of 30–35 mmHg during flushing of gas into the vitreous cavity and the subsequent injection of SO can allow air or SO to directly escape from the vitreous cavity to the sub-tenon space through the opening of previous sclerotomies,^{3,5} or from a scleral rupture in an occult scleral weak area,¹ leading to emphysema and SO migration. The gradually expandable SF6 tamponade or a mixture of 18% octa-fluor-propane (C3F8) gas can also result in the eventual leakage of microbubbles through sclerotomies toward the periorbital and intraconal space.⁵

The migrated substances in our patient were detected by CT scans and were shown to have reached the subconjunctival space, orbit, periorbita, and lids. CT scans are also important to determine the possibility of suprachoroidal/retrobulbar hemorrhage or endophthalmitis, which can be other complications after ocular surgery.

Treatment was not necessary for emphysema, which spontaneously resolved in the first postoperative month in the present case. The air was reabsorbed slowly, and the hemifacial and lid edema progressively disappeared. Although periorbital subcutaneous emphysema is a self-resolving condition and may not require any active intervention, orbital emphysema can be potentially vision-threatening and shows the potential to spread into deeper tissue planes such as the retropharyngeal space.⁴ Compressive optic neuropathy, central retinal artery compression, pneumomediastinum and airway obstruction are other ocular and life-threatening complications of emphysema.⁴

On the other hand, the migrated SO in this case required removal. After one month, the SO persisted, leading to a thickened lid, and we decided to debulk the lid and periorbital tissues. During the procedure, we observed several small inert round translucent cysts infiltrating all layers of the lids and nearby tissues, as confirmed by the histological examination. SO cysts in the periorbital tissues have already been reported, and they are known to result in granulomatous reactions characterized by macrophages with clear vacuoles containing SO and marked fibrosis.^{1,6–13} SO is known to cause only minor inflammatory reactions. However, this chronic reaction can occur because SO leakage from the vitreous cavity can possibly trigger tissue granulation, with the possibility of evolving into an inflammatory granulomatous process and persistent lymphoedema.^{7,13} The remaining SO in the orbit can continue to migrate to the lids with the possibility of chronic granulomatous reaction with edema, lymphedema, and persistent mechanical ptosis, which can have an indolent and chronic evolution.

4. Conclusion

We report a rare case of SO migration and emphysema infiltrating the hemiface, mainly the periorbita, orbit, lid, and subconjunctival space, in a patient after the fourth vitreoretinal surgery. Air and SO probably escaped through an old sclerotomy or a potentially occult scleral defect as a result of high infusion pressure and scleral depression. The risk of SO and air migration increased with multiple sclerotomies, thin sclera, or inadvertent globe perforation/rupture, and surgeons must review these risk factors prior to surgery. Self-limited periorbital emphysema can resolve spontaneously unless intervention is indicated in severe cases. Options for a migrated SO include removal of the collected material since its retention can lead to chronic edema in the region.

Research ethics

Approval was obtained from the Institutional Review Board of King Khaled Eye Specialist Hospital. This report does not contain any personal information that can lead to patient identification.

Patient consent

Written consent to publish this case was not obtained. This report did not contain any personal information.

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Authorship

All authors attest that they meet the current International Committee of Medical Journal Editors (ICMJE) criteria for authorship. Contributions of authors are as follows: Algethami A – data acquisition and interpretation; Elkhamary SM – evaluation and revision of the image examinations; Schellini SA – assisted the patient performing debulking and revised the paper critically; Talea MA, – assistant physician; Semidey VA, information analysis, drafting, and approval of the final manuscript.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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