

## Idiopathic orbital lipogranuloma

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### Dear Editor,

We are writing this letter to present a very rare case of idiopathic orbital lipogranuloma. Lipogranuloma is a granulomatous inflammatory soft tissue reaction, consisting of lipid deposition and/or an oil-like substance commonly associated with injections, trauma or secondary to systemic diseases. Lipogranuloma has been referred to as oil granuloma, lipid granuloma, oleogranuloma, paraffin granuloma or chalazion<sup>[1]</sup>.

The development of lipogranuloma has been attributed to a foreign body reaction to lipid or oil like substance<sup>[2-4]</sup>. The lesion on the other hand can be idiopathic, occurring due to endogenous degeneration of lipid<sup>[5-7]</sup>.

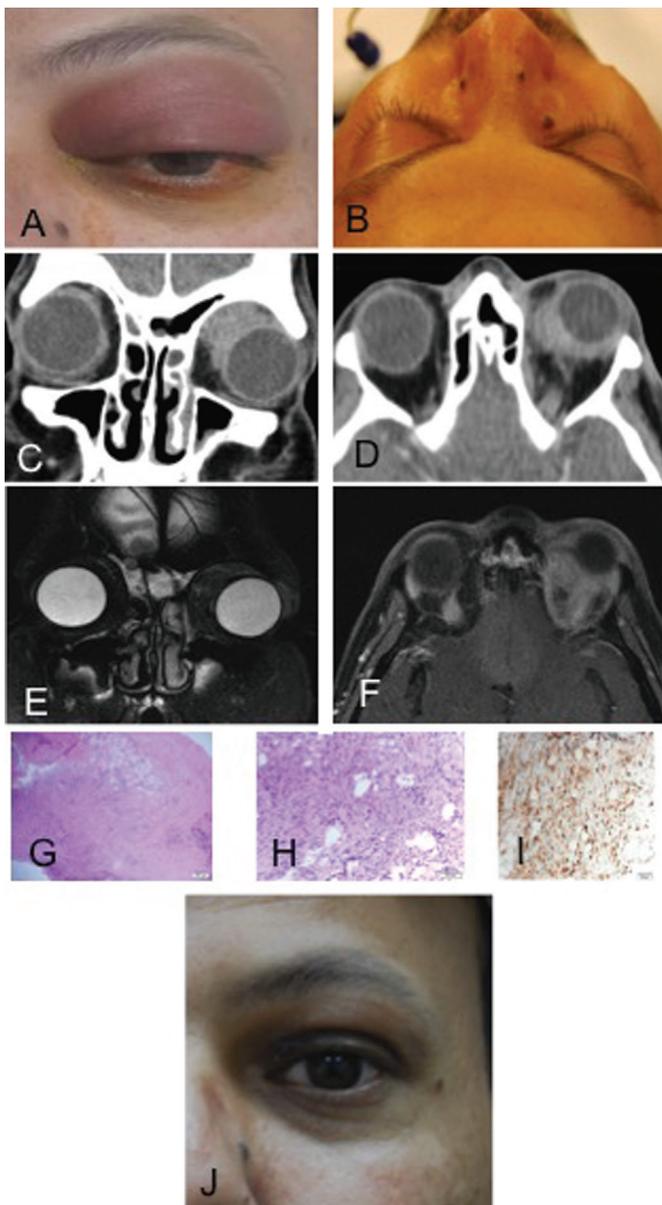
Lipogranuloma of the head and neck including the periorbital and orbital region is very rare. Clinically, periorbital and orbital lipogranuloma commonly manifests as a palpable subcutaneous nodule, eyelid swelling, or even proptosis<sup>[8]</sup>. Currently there are only four published reports of lipogranuloma with orbital involvement<sup>[9-10]</sup>. Previous cases have been reported in males and were associated with systemic disease or trauma. To our

knowledge, we present the first case of idiopathic orbital lipogranuloma in English peer review literature.

A 35-year-old Saudi diabetic male presented with painless gradual swelling and blepharoptosis of the left upper lid associated with diplopia of onemont honset. On ophthalmic examination, he had moderate blepharoptosis, redness and tenderness of the left upper lid, in addition to inferolateral left globe displacement. A palpable mass was noted near the superomedial superior orbital rim that was not easily mobile and poorly demarcated. There was no skin long time discoloration. The left eye was mildly proptosed. Exophthalmometry at base of 110 mm showed the following measurements: 21 mm OD and 23 mm OS. The left eye had limited supraduction and adduction (Figure 1A-1B). The patient denied any history of trauma, any surgery involving the face and lids or any kind of ocular treatment. There was no history of steroid or facial autologous fat injections. Computed tomography (CT scan) of the orbits showed a large well localized soft tissue mass, which is with focally infiltrative margins along the extraconal and intraconal aspects of the orbit. Magnetic resonance imaging (MRI) indicated a soft tissue mass with ill-defined margin along the posterior aspect of the left globe. The mass measurements were larger than what was clinically estimated by palpation, with 50 HU (houns field unit) in density on non-contrast study. The lesion exhibited heterogeneously intermediate signal on both the T1- and T2-weighted images. There was no high signal on T1-weighted images without fat suppression to suggest fat locule. Fat and adjacent extraocular muscles with contrast injection revealed a diffuse enhancing mass with poorly-defined contours (Figure 1C-1F).

An incisional orbital biopsy was performed through the upper eyelid crease. The mass was composed of hard, grey ish material, and was firmly adherent to the surrounding tissue. The histopathologic examination disclosed multiple pieces of soft tissue with evidence of fibrosis, and fat necrosis. Small, round empty spaces were present with (lipoblast-like) vacuoles and larger spaces representing extracellular lipid surrounded by histiocytes and scattered lymphocytes of variable size.

Ibuprofene 600 mg tablets (Sandoz Inc., Princeton, NJ, USA) three times a day were prescribed for 3d. His clinical picture improved in one week follow up then remained asymptomatic for 9mo post surgery (Figure 1J).



**Figure 1 Clinical appearance, radiology images and pathology slides of the patient with idiopathic lipogranuloma** A-B: Clinical pictures, showing left eye proptosis, ptosis, upper lid injection; C-D: Axial and coronal CT-scan with a soft-tissue algorithm showing an irregular-shaped well-defined mass encircling the left globe posteriorly that appeared homogeneous and an isoattenuated adjacent extraocular muscle; E: Coronal T2-weighted image with fat suppression showed the mass infiltrating the extraocular muscles with ill-defined, and heterogeneous isointense soft-tissue mass, extending into the extraconal space; F: The axial MRI contrast-enhanced T1-weighted image with fat saturation shows moderate heterogeneous infiltrative pattern enhancement including extraocular muscles and fat; G-I: Histopathology images G, showing low power appearance of the tissue excised with evidence of fibrosis and an area of fat necrosis (original magnification  $\times 50$  hematoxylin and eosin); H: A higher power histopathology image showing multiple clear areas representing dissolved fatty material upon fixation and processing surrounded by epithelioid cells, lymphocytes and multinucleated foreign body-type giant cells (original magnification  $\times 200$  hematoxylin and eosin); I: The same lesion showing the non-Langerhan's histiocytic infiltrate with positive IHC staining (original magnification  $\times 200$ , CD68); J: Postoperative clinical pictures after 6mo, showing left eye with no proptosis, and no signs of inflammation.

To date, there are very few cases of periorbital or orbital lipogranulomas reported in the literature<sup>[11]</sup>. Our case of an idiopathic lesion affecting the orbitis extremely rare. In most cases, these lesions occur secondary to procedures that may have precipitated the sclerosing lipogranulomatous process such as an intradermal injection of paraffin, and autologous fat injection<sup>[5-6,11]</sup>. Leakage of intraocular silicone oil into the orbit or eyelid tissues high pressure stream from mechanical equipment, foreign substances or accidental injection of oil into the orbit are other causing factors reported<sup>[12-13]</sup>. Periorbital lipogranuloma has also been reported after injection of ophthalmic ointment after laser canaliculoplasty, application of ointment in the lacrimal ductor due to washout of ointment plugs from the sinuses into the surrounding tissues by postoperative hemorrhage after the following procedures: endonasal sinus surgery<sup>[2-3,7]</sup>, subtenon steroid injection<sup>[14]</sup>, periocular ointment treatment<sup>[7]</sup>, vitrectomy surgery<sup>[12]</sup> or associated with chalazion<sup>[15]</sup>. However, the abnormal fat can be endogenous, derived from the panniculus as a result of trauma, ischemia due to vasculitis or, more likely, some other unidentified process that may have been present in our patient. We performed an extremely thorough history taking on known factors related to the lipogranuloma and the patient denied any precipitating events<sup>[10]</sup>.

The most common presenting symptoms of periorbital lipogranuloma is a palpable mass, followed by blepharoptosis and eyelid edema as present in our patient<sup>[8,11]</sup>. Inflammatory symptoms and signs occurring in lipogranuloma can be explained by possible vasculitis in the area of the lesion<sup>[11-12]</sup>. The lesion was located superomedially in the orbital rim leading to ptosis and displacement of the left eye with subsequent diplopia in our patient. Additionally, diplopia in our case could be secondary to infiltration of the oculomotor nerve that was observed in the MRI studies.

The histologic findings of the current case indicated granulomatous processes without any sign of a specific systemic granulomatous disorder. CT scan and MRI image were not typical for lipogranuloma as previously mentioned in literature with classic fat loculi<sup>[4]</sup>. However, our patient had a very larger well localized lesion, typically demonstrating moderate heterogeneous enhancement on contrast-enhanced images following the administration of the intravenous contrast material in both CT scan and MRI.

Characteristically, the lipid dissolves during the fixation process, leaving a ghost or silhouette centrally in the granuloma and gradual enlargement and encapsulation of these lesions contributing to the multi-lobulated appearance on clinical examination mimicking malignancy<sup>[1]</sup>. The polymorphic dropout spaces vary from a non-inflammatory lesion to those with an associated granulomatous inflammatory reaction. In our case, there were small round cells with a (lipoblast-like) vacuole

## Idiopathic periocular lipogranuloma

whereas the larger spaces represented extracellular lipid that were not reported in any other kind of lipogranuloma<sup>[11]</sup>.

The term “idiopathic noninfectious granulomatous inflammation” is reserved for localized granulomatous infiltrates involving orbital soft tissues that lack systemic association to granulomatous inflammatory syndromes including sarcoidosis, Wegener’s granulomatosis, Erdheim-Chester disease, pseudorheumatoid nodules, and necrobiotic xanthogranuloma<sup>[14]</sup>. The histopathologic examination definitively excluded the association because these diseases have other specific findings such as Touton giant cells<sup>[11]</sup>.

Complete excision of the granuloma is the most effective therapy. However, incisional biopsy resulted in resolved symptoms in our case. Treatment can be early surgical removal or extended biopsy, resulting in a cure or immediate severe recurrence. There was no recurrence of the lesion in our patient for a total follow up period of 9mo. Other treatment options include simple triamcinolone injection or debulking<sup>[8,10]</sup>. Spontaneous resolution has been also reported<sup>[8]</sup>.

In conclusion, we have described a very rare case of idiopathic orbital lipogranuloma treated by incisional biopsy with good outcome. Ophthalmologists and ophthalmic pathologists should be aware of this rare occurrence. In similar cases where this diagnosis is suspected, causes of secondary lipogranuloma should be excluded and incisional biopsy will confirm the diagnosis and may resolve the lesion.

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