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Eyelid Gouty Tophus with Complete Ptosis: The First Case Report

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Abstract: A 79-year-old man presented with a gradually enlarging painless swelling around the right upper eyelid for over 1-year duration. On examination, a hard and non-tender mass noted over right upper eyelid, causing complete mechanical ptosis. CT scan revealed a well-defined enhancing lesion at the right upper eyelid, size measuring 0.9x2.4x1.4cm. Following excision biopsy, the histologic examination revealed fragmented tissue comprising mixture of dense chronic inflammatory infiltrates and scattered amorphous eosinophilic deposits. The deposits are surrounded by histiocytes and multinucleated giant cells. Special stain with Congo Red showed the deposits are non-congophilic. It is consistent with gouty tophus.

Keywords: Gout, Tophus, Eyelid, Complete ptosis

INTRODUCTION

Gout generally progresses through four clinical stages if left untreated: asymptomatic hyperuricemia, acute gout, intercritical or interval gout and chronic tophaceous gout [1]. Although gouty tophi are seen in chronic disease, tophi may be first sign of the disorder [1]. While tophi more commonly occur in the digits, ear pinna, prepatellar bursa, and olecranon, they very rarely occur around the eye [2-4]. We report an unsuspected case of gouty tophus involving the upper eyelid with normal serum uric acid level, prior to any other manifestation of gout. To our knowledge, this is only the seventh case of gouty tophus of the eyelid reported in the literature, and the only one that causes complete ptosis.

CASE REPORT

A 79-year-old man presented with a gradually enlarging swelling around the right upper eyelid. It was painless and associated with a right ptosis. The swelling had developed continuously over a 1-year period. He had primary angle closure glaucoma where both eyes peripheral iridotomy and phacoemulsification were done. In medical history, he had hypertension, ischemic heart disease, benign prostate hypertrophy, polycythemia vera and history of colon carcinoma where surgery was done 6-month prior to the presentation and had completed chemotherapy.

On examination, visual acuity was perception of light (PL) for Right eye and 6/18 for left eye. There was a hard mass over right upper eyelid, resulting in complete mechanical ptosis (Figure 1). It was nontender, not associated with discharge or bleeding, no skin changes and not attached to underlying skin. Slit lamp examination revealed both eyes peripheral iridotomy (PI) were patent, cornea clear, and IOLs insitu. Fundus examination showed right optic disc was pale with CDR 0.9. Left optic disc was pink with CDR 0.6.

CT scan revealed a well-defined enhancing lesion at the right upper eyelid, size measuring 0.9x2.4x1.4cm. No clear plane of demarcation with right globe and no obvious erosion of bony orbits (Figure 2). MR imaging showed a well-defined mildly enhancing lesion with epicentre at the right upper eyelid, measuring 1x2.4x1.5cm. It is hypointense on T1/T2/STIR and hyperintense on T1 FS. Right laterally no clear plane with the anterior aspect of right lacrimal gland. Posteriorly no clear plane with right globe. No intraorbital extension. Superiorly it extends to superior orbital septum (Figure 3 & 4).

Right upper eyelid excision biopsy was done. The specimen was fixed in formalin and submitted for histopathological examination. Gross examination of the formalin-fixed specimen revealed the right upper eyelid mass comprised multiple fragments of brownish tissue measuring 30mm in aggregate diameter. Histologic examination revealed fragmented tissue comprising mixture of dense chronic inflammatory infiltrates and scattered amorphous eosinophilic deposits. The deposits are surrounded by histiocytes and multinucleated giant cells (Figure 5). Special stain with Congo Red showed the deposits are non-congophilic

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(Figure 6). It is consistent with gouty tophus.

Subsequent blood test revealed normal uric acid level.



Fig-1: A hard mass over right upper eyelid, resulting in complete mechanical ptosis



Fig-2: CT scan revealed a well-defined enhancing lesion at the right upper eyelid, size measuring 0.9x2.4x1.4cm.

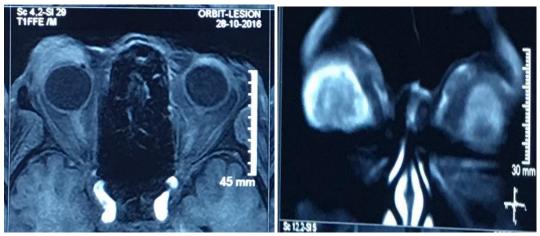


Fig-3,4 MR imaging showed a well-defined mildly enhancing lesion with epicentre at the right upper eyelid, measuring 1x2.4x1.5cm.

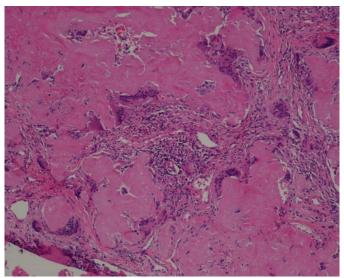


Fig-5: Histologic examination revealed fragmented tissue comprising mixture of dense chronic inflammatory infiltrates and scattered amorphous eosinophilic deposits. The deposits are surrounded by histiocytes and multinucleated giant cells

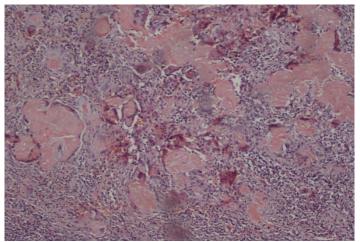


Fig-6: Special stain with Congo red showed the deposits are non-congophilic.

DISCUSSION

Hyperuricemia is the most important risk factor for the development of gout, the risk increasing with a higher urate concentration. But the level of serum uric acid (SU) may be normal in gout, especially in diabetics and alcoholics [5]. In the largest studies of acute gout to date conducted by Schlesinger et al, attacks still occurred despite SU levels being below 6.8 mg/dl, the saturation level for urate. This may be attributed to persistence of tophi and an increased body uric acid pool. In addition, it may be that in some patients SU may not sufficiently represent their uric acid pool [6]. Our patient was non-alcoholic and nondiabetic. His serum uric acid level was within normal limits and was never suspected to have gout before. However, he had history of colon cancer which completed chemotherapy and polycythemia vera. Both of this condition may increase the body uric acid pool due to high turnover of cells.

Tophi generally developed after an average of 11.6 years of gouty arthritis before uric acid lowering therapy became available [7]. They were reported to occur in 12% of patients after 5 years, and 55% after 20 years of untreated disease [8]. However, they can develop without the concomitant arthritis. Iglesias *et al.*, used the term 'gout nodulosis' to describe the subcutaneous deposits of MSU in the absence of initial manifestation of gouty arthritis [9]. Our patient developed gouty tophus involving the upper eyelid with normal serum uric acid level, prior to any other manifestation of gout.

A literature search revealed only six prior cases of tophi in the eyelid or canthi (five at the medial/lateral canthi, one in the superior eyelid) reported between 1986 and 2016 [10]. All cases of eyelid tophi did not presented with ptosis and were painless masses with no discharge, inflammation, or bleeding. These patients had previous histories of gout ranging from 3 to 20 years, with tophi presentation in areas such as the

elbows and first metatarsal [10]. Our case presented as a painless gouty tophus over right upper eyelid with no discharge or skin changes and results in complete mechanical ptosis.

CONCLUSION

Our patient presented as a painless gouty tophus over right upper eyelid with normal serum uric acid level, as the first manifestation of gout in this case. To our knowledge, this is only the seventh case of gouty tophus of the eyelid reported in the literature, and the first case that causes complete ptosis. Ophthalmologists should be aware of this entity because patients may present with a tophus as the initial manifestation of gout.

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