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Intralesional corticosteroids as an Alternative Therapy for Orbital Angiolymphoid hyperplasia with eosinophilia (ALHE)

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Abstract: Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare and idiopathic, benign vascular disorder that manifest in adults as smooth-surface red to brown papules or nodules on the head and neck and may involve the ocular adnexa and orbit. The lesion may be persistent and is difficult to eradicate. Treatment has traditionally been surgical excision that may be difficult for orbital lesions. We present a case of orbital ALHE that responded favorably to intralesional corticosteroids.
Keywords: Angiolymphoid hyperplasia, eosinophilia, papules

INTRODUCTION

ALHE is a benign uncommon skin disease occurring slightly more frequently in young females with no racial predominance, but it is reported more frequently in Asians [1]. The aetiology of ALHE is still unknown but proposed pathogenesis includes a benign neoplastic process, a hypersensitivity reaction, inflammatory vascular reaction or a tissue reaction to a previous trauma, arteriovenous shunting, hyperestrogenemia (eg, pregnancy or oral contraceptive use) and infectious agent[2,3]. A peripheral blood eosinophilia is associated in only 20% of cases[2,3]. Histologically, there is vascular proliferation with plump, "cobblestone" endothelial cells surrounded by a cellular infiltrate of lymphocytes, plasma cells, and up to 50% eosinophils [2, 3].

CASE REPORT

A 34-year-old healthy Malay gentleman presented with marked swelling in the left upper eyelid for 7 months with multiple swelling over the scalp and submandibular regions. His left eye vision was 6/9. There was a diffused, firm, non-tender upper lid mass involving the whole lid with marked mechanical ptosis and no overlying inflammation.

Extraocular movement was full and no proptosis. The right eye and posterior segment of both eyes were normal. He underwent surgical excision twice due to disease recurrence and both HPE report were consistent with ALHE.

He initially responded to oral corticosteroid, however, only lasted for 4 months due to poor compliance. Subsequently intralesional corticosteroid was commenced and he responded well.

Fig-1: There was marked diffuse swelling in the left upper eyelid with partial mechanical ptosis.

Fig-2: The left upper eyelid lesion 1 week post first intralesional corticosteroids injection.

Fig-3: The left upper eyelid lesion after completed the sixth intralesional corticosteroids injection.

DISCUSSION

Although ALHE has a benign course and can have a spontaneous remission, treatment is often pursued in large or symptomatic orbital lesions. The most recommended management remains deep surgical excision with a recurrence rate of 33%-50% [1-3]. Complete excision of orbital lesions are often difficult due to poorly delineated margin but other treatment modalities have been reported [1,3]. Therapy includes steroids (intralesional, oral or topical), oral retinoids, pentoxifyllin, intralesional chemotherapy, radiotherapy, cryotherapy and vascular laser therapy has also been used with promising result of low recurrence rate [1-4].

In our case, the patient had significant mechanical ptosis that affects daily activities. Following recurrence after second deep surgical excision and poor compliance to oral corticosteroid, a monthly intralesional corticosteroid (2 ml of triamcinolone acetonide (20 mg/ml)) was given. The lesion showed marked regression after third injection of intralesional corticosteroids and he had completed for a total of six injections.

CONCLUSION

Although there are many treatments modalities reported for ALHE management, we would like to highlight this challenging case because a simple, noninvasive traditional therapeutic approach of using intralesional corticosteroids has shown a positive result in terms of recurrences and aesthetic outcomes.

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