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A Case Report On Effect of Intralesional Injection of Bleomycin In Case Of Orbital Lymphangioma

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I. INTRODUCTION

- Lymphangiomas are benign hamartomatous lymphatic tumours, characterized by multiple communicating lymphatic spaces and cystic spaces.
- Orbital lymphangioma presents usually in childhood.The most frequent presenting feature of orbital lymphangiomas are proptosis and ptosis of the affected eye. These symptoms are often not clinically present until an inciting factor causes an increase in size of the lesion. This is often due to mild trauma causing hemorrhage into the lesion or acute upper respiratory infection leading to swelling of the lesion
- The major management falls in 4 categories
- 1)Observation
- 2)Sclerotherapy
- 3)Systemic medication
- 4)Surgery

Aim

To study the effect of intralesional bleomycin in case of orbital lymphangioma in a 7 year old female.

II. METHODOLOGY

A 7 year old female came to our institution with complaints of swelling , restriction of ocular movements in upward and downward direction ,eyeache and sudden diminision of vision within 3 days. She was having visual acuity of Right eye 6/6 and Left eye counting fingers 6meters with eccentric proptosis and having left eye upper lid and lower lid edema with ecchymosis. Patient was investigated with MRI orbit with report of retro orbital mass with proptosis suggestive of lymphangioma.

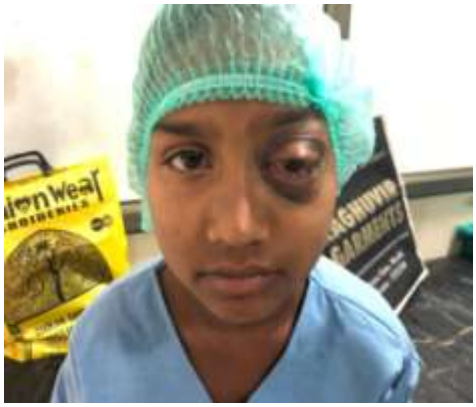

Figure 1Appearance of patient before administration of bleomycin injection

Figure 2Appearance of patient before administration of bleomycin injection

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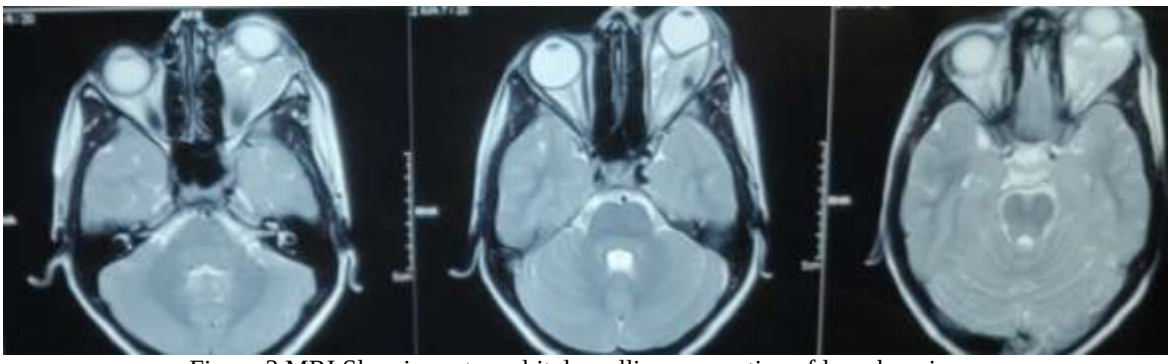

Figure 3 MRI Showing retro orbital swelling suggestive of lymphangioma

III. RESULT

Visual acuity of patient improved to Right eye 6/6 and Left eye 6/18 from visual acuity of left eye counting fingers 6 meters on post operative day 1 with resolution of papilloedema and decrease in amount of proptosis.

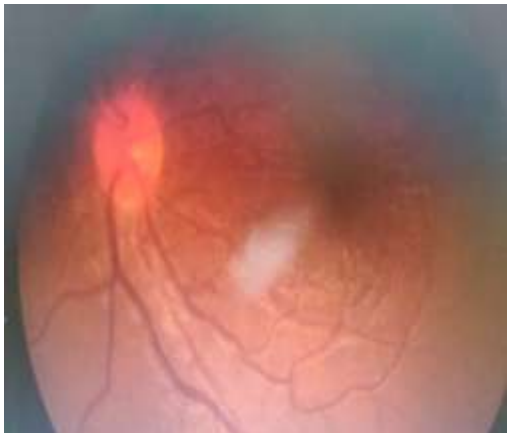

Figure 4Fundus Showing resolution of papilloedema

Figure 5Picture Showing resolution of proptosis after sclerotherapy

IV. CONCLUSION

- A significant improvement is seen after the administration of Intralesional injection of bleomycin with improvement in visual acuity, orbital movement , proptosis and resolution of papilloedema.
- Lymphangioma are thought to arise from a combination of the following: a failure of lymphatics to connect to the venous system, abnormal budding of lymphatic tissue, and sequestered lymphatic rests that retain their embryonic growth potential. These lymphatic rests can penetrate adjacent structures or dissect along fascial planes and eventually become canalized. These spaces retain their secretions and develop cystic components because of the lack of a venous outflow tract. The nature of the surrounding tissue determines whether the lymphangioma is capillary, cavernous, or cystic.
- At present, complete excision remains the treatment of choice for lymphangioma. For complex lymphangioma, complete removal may require multiple operations and may not be possible without damaging adjacent vital structures.
- The complications from the surgical excision of a lymphangioma are related to the location and structures adjacent to the mass; these include damage to a neurovascular structure (including cranial nerves), chylous fistula, chylothorax, hemorrhage, and recurrence. Most recurrences occur within the first year but have been reported to occur as long as 10 years after excision.
- Unlike in hemangiomas, spontaneous resolution of lymphangioma is uncommon. Recurrence is rare when all gross disease is removed. If residual tissue is left behind, the expected recurrence rate is approximately 15%. Postoperative complications, including recurrence, wound seromas, infection, and nerve damage, occur in 30% or more of cases. Recurrence rates vary depending on the complexity of the lesion and the completeness of excision.

- Complete surgical resection may be difficult due to the presence of multiple loculations and extensive disease.
- Sclerotherapy provides a treatment option in such patients, offering a viable alternative to surgery in patients with macrocystic lymphatic malformations (LM). Although the popularity of sclerotherapy in the treatment of LM is growing, there is no consensus regarding the type of sclerosant. This is largely due, in part, to a lack of understanding on sclerosant mechanism of action.
- Umezawa first developed bleomycin as an antitumor agent in 1966 and its mechanism of action was by inhibition of DNA synthesis. This drug was also known to produce a sclerosing effect due to its direct action on the endothelial cells producing non-specific inflammatory reaction. Desired effect of sclerosis is achieved by local action of bleomycin, which depends on availability of drug per unit surface area of lesion.
- Side effects of Bleomycin are fever, transient increase in size of swelling, hemorrhage, leukocytosis, infection, and pulmonary fibrosis. Pulmonary fibrosis has been associated with intravenous bleomycin administration exceeding the total cumulative dose of 400 mg. Bleomycin doses used in sclerotherapy are small in comparison, typically 1% to 5% of the lowest dose associated with possible pulmonary fibrosis. We used 0.5 mg/kg of dose. Others have used it in doses ranging from 0.3 to 3 mg/kg.
- Although surgical excision has been considered to be treatment of choice by most of the surgeons but it is associated with tedious dissection along with lot of morbidity in the form of disfigurement and damage to vital structures and ugly scar. Therefore, sclerotherapy of lymphangioma has gained popularity during recent years.In short term follow up no recurrence was encountered.
- To conclude, use of bleomycin as an intralesional agent for lymphagioma appears to be safe and rewarding. This therapy may be used as primary modality instead of surgery in selected group of patients.

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