Case Report

Percutaneous Sclerotherapy with Bleomycin for Orbital Lymphangioma: A Case Report

PJO – Official Journal of Ophthalmological Society of Pakistan

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ABSTRACT

Lymphangiomas, or lymphatic malformations, are benign, multi-cystic, hamartomas which are localized vascular malformations believed to arise during embryonic period.[1] Lymphatic malformations most frequently occur in pediatric populations in the region of head and neck and they account for about 25% of benign pediatric vascular tumors and about 4% of all vascular tumors.[1] Orbital lymphangiomas have a prevalence of 1.1 to 5.3 cases per 10,000 live births and are equally common in both males and females.[2] Histopathologically, lymphangiomas consist of endothelium lined, thin-walled dilated lymphatic spaces, filled with eosinophilic proteinaceous material.[1]Management of lymphangiomas is challenging. We report a case of a 9-year-old female who had orbital lymphangioma and was treated with sclerotherapy. Outcomes of treatment and review of literature are presented here.

Keywords: Orbit, lymphangioma, sclerotherapy, bleomycin.

How to Cite this Article: Tanveer T, Ahmad S, Tahir F, Akram F. Percutaneous Sclerotherapy with Bleomycin for Orbital Lymphangioma: A Case Report. 2025;41(4):456-459. **Doi: 10.36351/pjo.v41i4.2128**

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Received: June 16, 2025
Revised: September 1, 2025
Accepted: September 13, 2025

INTRODUCTION

Lymphangiomas are benign, multi-cystic hamartomatous malformations of the lymphatic system seen in children, most commonly in the region of head and neck.[1] Most commonly the lesion becomes apparent following an upper respiratory tract illness or minor trauma when the cystic spaces bleed, resulting in either acute ptosis or proptosis with limitation of extraocular movement. Lymphangiomas can be classified depending on the size and depth of the cystic spaces as microcystic (<2 cm), macrocystic (>2 cm cysts), or mixed.[3]

CASE PRESENTATION

A 9-year-old girl was referred to our hospital with the primary complaint of painless periorbital swelling of the right eye since birth. History revealed that the swelling had gradually increased in size and was associated with recurrent episodes of bleeding and surrounding skin discoloration. Examination of the right eye showed periocular edema with ecchymosis that was pronounced inferiorly. A soft, translucent lobulated hemorrhagic mass was seen filling the entire inferior fornix, 28 × 10mm in size extending onto the nasal bulbar conjunctiva and encroaching 5mm onto the nasal limbus sparing the visual axis (Figure. 1a). The lesion had extensions deep into the inferior fornix and the caruncle, while the posterior limit of the mass was not visible. Subconjunctival hemorrhage was noted at the inferior bulbar and forniceal conjunctiva surrounding the lesion. Hemorrhagic discharge was also noted from the right eye. The mass was painless and immobile on palpation. Examination of the left eye was unremarkable. Visual acuity of the patient at presentation was 6/36 uncorrected OD and 6/6 uncorrected OS. On examination of the oral cavity, a pinkish translucent triangular lesion, with pebbled surface appearance was noted on the palate (Figure 1b). The palatal lesion was 15× 15 × 20 mm in size.

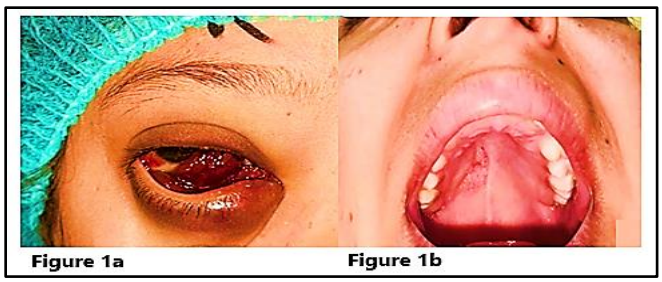

Figure 1a: Periocular edema with ecchymosis. A soft, translucent lobulated hemorrhagic mass is filling the inferior fornix. 1b: Oral cavity shows pinkish translucent triangular lesion, with pebbled surface appearance on the palate.

Orbital MRI showed a diffuse heterogeneous, non-encapsulated, multilocular, poorly defined cystic lesion in the right orbit with multiple air-fluid levels. The lesion extended from eyelids to orbital apex along the medial orbital wall. The lesion infiltrated the right intraconal and extraconal compartments. (Figure 2). Due to the extensive infiltrating nature of the lesion, the margins of the lesion were not clearly demarcated.

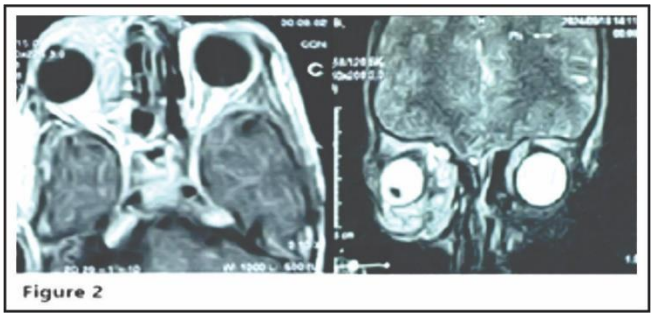

Figure 2: Orbital MRI T1 and T2 weighted showing a diffuse heterogeneous, non-encapsulated, multilocular, poorly defined cystic lesion in the right orbit with multiple air-fluid levels.

Surgical excision could have been challenging and could risk vital orbital structures. Keeping in view good vision of 6/36 in the affected eye, surgical excision was avoided. The patient was booked for sclerotherapy with bleomycin. For the first injection, the patient weighing 20kg, the dose of bleomycin was calculated as 0.5 mg/kg, resulting in a total dose of 10 mg, which is within the maximum recommended dose of 15 mg. Prior to injection, 2 mL of lymphangioma fluid was percutaneously drained with 24G needle on 3ml syringe. A vial containing 15 mg of bleomycin was used for preparation. Initially, 1.5 mL of bleomycin solution was prepared, from which 1 ml was drawn and mixed with 1 mL of 2% lignocaine to create a 1:1 solution of 2 ml. The prepared solution was injected within the lesion under strict aseptic conditions (Figure 3).

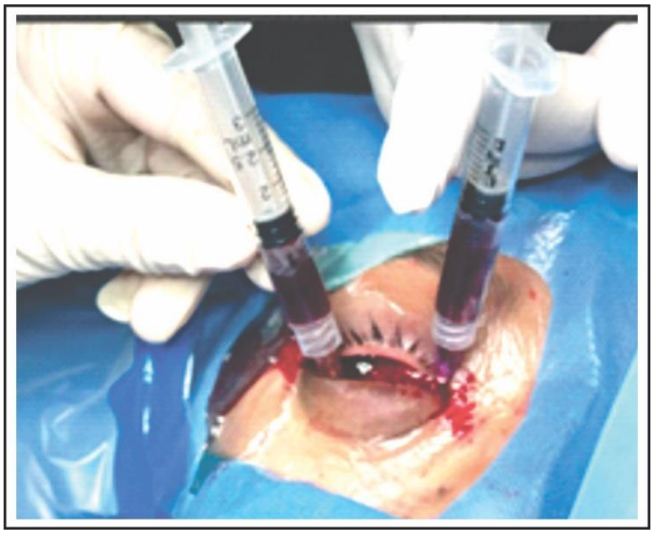

Figure 3: Intralesional injection of bleomycin.

This approach ensured accurate dosing, patient comfort, and optimal therapeutic efficacy. A second injection was given 4 weeks apart from the similar dosing and dispensing method. After the first sclerotherapy, the lesion size did not decrease considerably and no significant improvement in visual acuity was noted. However, after the second sclerotherapy session, there was a significant reduction in the size of the lesion (Figure. 4) with an improvement in the uncorrected visual acuity from 6/36 to 6/18.

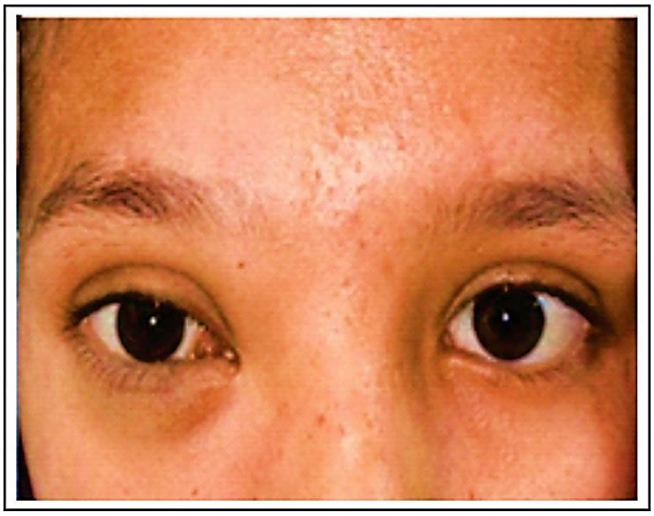

Figure 4: Considerable reduction in size of lymphangioma after second session of sclerotherapy.

DISCUSSION

Lymphangiomas are benign, multi-cystic hamartomatous malformations of the lymphatic system seen in children, most commonly in the region of head and neck.[1] Most commonly the lesion becomes apparent following an upper respiratory tract illness or minor trauma when the cystic spaces bleed, resulting in either acute ptosis or proptosis with limitation of extraocular movement. Lymphangiomas can be classified depending on the size and depth of the cystic spaces as microcystic (<2 cm), macrocystic (>2 cm cysts), or mixed.[3] The clinical appearance of orbital lymphangiomas vary according to the location and extent of the lesion, and they can manifest as painless, gradually enlarging soft tissue masses causing proptosis or ptosis, extraocular movement limitation, physical deformity and compressive optic neuropathy.[2] The management of lymphangioma can be challenging. Recurrence is frequent, complete excision or obliteration is not always possible, and therapies incur the risk of damaging the surrounding structures. There is a considerable chance of scarring and recurrence with surgery. Sclerotherapy is an alternate treatment.[4] A multimodal strategy that incorporates both surgical and nonsurgical (e.g. sclerosants) options is probably most beneficial to the patients.

An acute enlarging lesion can cause compression of the surrounding vital structures, resulting in compressive optic neuropathy.[5] Due to the infiltrative nature of the lesion surgical excision is often difficult and recurrence is common.

Injecting intralesional sclerosing agent is the most common alternative for a lymphangioma that cannot be fully resected or is surgically challenging due to its proximity to vital orbital structures. It has been a preferred treatment for many lymphangioma patients, with positive outcomes and no serious side effects.[6] Sclerotherapy offers several benefits over surgical excision, including ease of use, repeatability, and a decreased chance of nerve damage. Commonly used sclerosants include bleomycin, hypertonic saline, ethanol, acetic acid, OK-432, sodium tetradecyl sulfate and doxycycline.[7]

Bleomycin is a chemotherapeutic agent that possesses antibacterial and antiangiogenic properties, but its primary mechanism of action in treatment of vascular abnormalities is its intralesional sclerosing effect on the abnormal vascular endothelium.[8] Orbital lymphangiomas have been successfully treated with intralesional bleomycin. Its efficacy in treating resistant deep orbital lymphangiomas, with positive clinical results has been documented.[8,9]

For challenging orbital lymphangioma cases where surgical excision is less likely to be successful, sclerotherapy is a viable alternative with positive results.

CONCLUSION

Early identification and treatment of orbital lymphangioma is vital to preserve vision and prevent amblyopia. It can be difficult to manage due to its infiltrating nature. Mostly it cannot be completely removed surgically and there is a significant risk of damage to the important orbital structures. Sclerotherapy, a less invasive, can be a viable, safe and effective first-line treatment for ocular lymphangioma with good results.

Funding: This study was not funded by any organization.

Patient's Consent: Researchers followed the guide lines set forth in the Declaration of Helsinki.

Conflict of Interest: Authors declared no conflict of interest.

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