

## Ophthalmology | Case Report

# Retina and optic nerve complications and hearing injury caused by Sclerotherapy of Facial Vascular Malformation

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## Abstract

This report describes a complication of symptoms consistent with transiency visual acuity damage and hearing injury following sclerotherapy with a polidocanol injection to a facial vascular malformation. After the direct injection of 5cc polidocanol-room air emulsion into the subcutaneous malformation, a 3-year-old boy developed bilateral eyelid swelling the next day, and on 7 days later at referral, visual acuity in the left eye was light perception. Fundoscopy revealed retinal hemorrhage; fluorescein angiography (FA) disclosed hyperreflective leakage on the optic disk 15 days later, and OCT showed nerve fiber edema between the disk and macular. The orbital magnetic resonance imaging (MRI) demonstrated the normal extraocular muscles. In a month later, bilateral eyelid swelling resolved but the visual acuity remained 20/500, and 20/20 in two months later, but the hearing injury remained. It needs much more special attention in sclerotherapy for facial malformation because the sclerosants may cause severe toxicity or inflammation in retina and optic nerve, resulting in transiency visual acuity damage and acoustic nerve injury. The related examination including ophthalmic investigation after the sclerotherapy is necessary.

**Keywords:** Polidocanol; Sclerotherapy; Facial Vascular Malformation

## Introduction

Sclerotherapy is an intervention to inject chemical agents as sclerosing solutions (sclerosants) into veins or vascular abnormalities to induce localized thrombosis and eventual fibrosis and obliteration of the vessels. Polidocanol sclerosing therapy is used to be recommended as first-line treatment for orbital vascular malformations such as lymphangioma and hemangioma [1,2] and is widely used by radiologists to treat facial vascular malformations including hemangiomas in the skin and oral cavities. However, it has been reported as a rare and severe complication of foam sclerotherapy including eventual bulbar phthisis [3], anaphylactic reactions, severe

neurologic events including cerebrovascular accident (CVA) and transient ischemic attack (TIA), superficial venous thrombosis, tissue necrosis, edema, amaurosis, central retinal and posterior ciliary artery occlusion and nerve damage have already been reported [4-6].

In this study, we describe a patient with facial subcutaneous vascular malformation who developed the retinal toxicity inducing transient visual acuity damage and the hearing injury after intralesional injection of a sclerosant, polidocanol.

## Case Report

A 3-year-old boy had a subcutaneous facial vascular malformation of about 1.6\*0.8 cm in irregular area located at the inferolateral of the left cheek region. The boy developed a blue lesion on the skin gradually in past one year and was finally diagnosed as vascular malformation when he was 3 years old. Angiographic examination may not be required in cases of Hemangioma, but a Color Doppler Ultrasonography could have been performed before proceeding for sclerosant injection. A Color Doppler Ultrasonography scan imaging at that time showed a low-echo mass in the left cheek (Fig. 2). But angiographic examination was not performed. In October, 2019, the boy was referred to radiologists for sclerotherapy.

After signing the informed consent, the patient underwent 2 percutaneous injections of 3% polidocanol to the subcutaneous vascular malformation in the left cheek region under ultrasound visualization of a 23-gauge needle tip by radiologists on monthly basis. 5 cc polidocanol was prepared by mixing 2 cc polidocanol with 8cc room air filtered through a 0.22-um membrane filter. No abnormalities were found after the first two injections under the sedation. However, at the third regular injection, the process was successful except the peculiar phenomenon that the little boy began to cry loudly, whose left cheek showed redness. At that time no clinical abnormalities were seen and no administration was given. On day 2, the boy developed lid swelling on both sides and hemifacial swelling on the left side (Fig. 3). The extraocular muscles (ECM) movements were checked immediately after the boy developed eyelid swelling and the results were normal. A funduscopy was not performed immediately because of the swelling in eyelid and the refusal of the parents, as the ophthalmic complication due ischemic damage are common following retrograde flow of sclerosant into the ECA and OA branches.

On day 3, he experienced much more severe painful swelling, and was administered with Cefalexin and dexamethasone (1mg/1kg/day) combined with hot compress daily for 4 days (day 3 to 6). On day 7, the eyelid swelling was resolved with normal eye movements, and the visual acuity of the left eye declined to 20/500 with 20/20 on the right eye, and the pupillary reflex of the left eye at the first examination was positive, and the hearing injury of left ear was found.

On day 15, funduscopy revealed 1 papilla disc (PD) retinal hemorrhage located on the inferotemporal region of the optic disc, and the temporal veins were slightly expansive and tortuous (Fig. 4). The OCT showed nerve fiber edema between the disc and macula fovea (Fig. 5). The orbital magnetic resonance imaging (MRI) demonstrated the normal extraocular muscles (Fig. 6).

On day 17, Fluorescein angiography (FA) and Indocyanine green angiography (ICGA) were performed with intravenous injection of half-dose fluorescein sodium and indocyanine green under sedation. FA and ICGA showed the same blocked fluorescence corresponding to the hemorrhagic lesion with high-reflective fluorescein leakage from optic nerve head at middle phase on FA indicating the normal retinal perfusion and swollen optic nerve fiber (Fig. 7). The choroidal circulation in indocyanine green angiography showed normal. On day 23, the central vision of the left eye was 20/500, and 20/20 after two months, but the hearing injury remained the same. The retinal hemorrhage was resorted, and the RAPD was normal.

## Discussion

The causes of visual acuity damage in this patient could be two-fold: (1) sclerosants could flow into these arteries from the vascular malformation, and (2) an allergic reaction or inflammation to sclerosants could cause eyelid swelling, retina and optic nerve inflammation or toxicity. Clinically, on the next day he developed eyelid swelling on both sides and hemifacial swelling on the left side. Magnetic resonance imaging disclosed normal extraocular muscles (ECM) in the bilateral orbit after 7 days when the patient was found to have visual acuity damage in the left eye. These clinical features indicated general edema involving the eyelid and the whole orbital tissue, and extending to contralateral eyelid and hemifacial edema, suggestive of allergic reaction to the sclerosant as described previously. The inflammation of middle ear also caused hearing injury. The lesions of the vascular malformation would have vascular connections with one another, and intralesional injected sclerosant might travel from the subcutaneous vascular malformation to the facial and orbital lesions and, finally, to the central retinal artery and posterior ciliary arteries which induced the inflammation or toxicity of the retina and optic nerve. Furthermore, the sclerosant might disseminate through collaterals of the external carotid artery system to the ipsilateral face and the contralateral eyelids.

Funduscopy revealed retinal hemorrhage locating the inferotemporal edge of the left optic disk and fluorescein angiography (FA) disclosed hyperreflective leakage on the optic disk 15 days later. OCT showed nerve fiber edema between the disk and macular, indicating the lesion of retinal ganglion cells (RGC) and retinal nerve fiber layer (RNFL).

Studies comparing room air and physiologic gas foam sclerotherapy preparations found no difference in treatment efficacy [7-9]. Given the high estimated prevalence of complication, and the question of whether endothelin or air embolism (or potentially both) causes severe neurologic events in patients treated with foam

sclerotherapy, current guidelines recommending physiologic gas and immediate administration seem reasonable but potentially impossible to implement on a wide scale basis. The role of compounded versus branded preparations of polidocanol and sotradecol and their potential role in central nervous system effects has not been elucidated. Continued investigation into the potential role of product, gas, volume, and technique to identify optimal approaches and products may further refine the consistency and safety of foam sclerotherapy.

The injection of sclerosants may induce anaphylactic reactions, severe neurologic events during or after the administration of sclerosants. Furthermore, the related examination including ophthalmic check should be performed before and after the sclerotherapy in case of mitigate related risks. There are multiple management options including Drugs (Oral Corticosteroid, Propranolol, Topical Imiquimod), Intralesional injections (Corticosteroid, IFN alpha, Sclerosant), Laser and Cryotherapy, Surgical Resection etc. It needs much more special attention in the sclerotherapy in child patients.

Mario Albert [8] observed a healthy man who experienced an unexpected hemiplegia with abrupt interruption of the middle cerebral arterial circulation immediately after injection of foam polidocanol. Then the patient was required to accept injection in a slight Trendelenburg position that distributed most dose of sclerosant flowing into left thigh and consequently avoided thrombosis obstructing the blood vessels in the right thigh. A combined therapy of keeping Trendelenburg positioning and supporting supplemental oxygen via nasal cannula contributed to the alleviation of the symptoms.

In our case, the ophthalmic investigations were not performed immediately on 2nd day, and the treatment began on 3rd day. The Ophthalmic investigations were supposed to be performed immediately, which could have given the opportunity to treat the complication timely and aggressively (including systemic corticosteroids, systemic anticoagulants, Hyperbaric O<sub>2</sub> therapy etc.). The management of dexamethasone impact therapy is not given to the children. The complaints of visual acuity damage which had been found on 7 days later due to the swelling lids and the diagnosis and prompt salvaging treatments were delayed even though the visual acuity recovered after two months.

## Conclusion

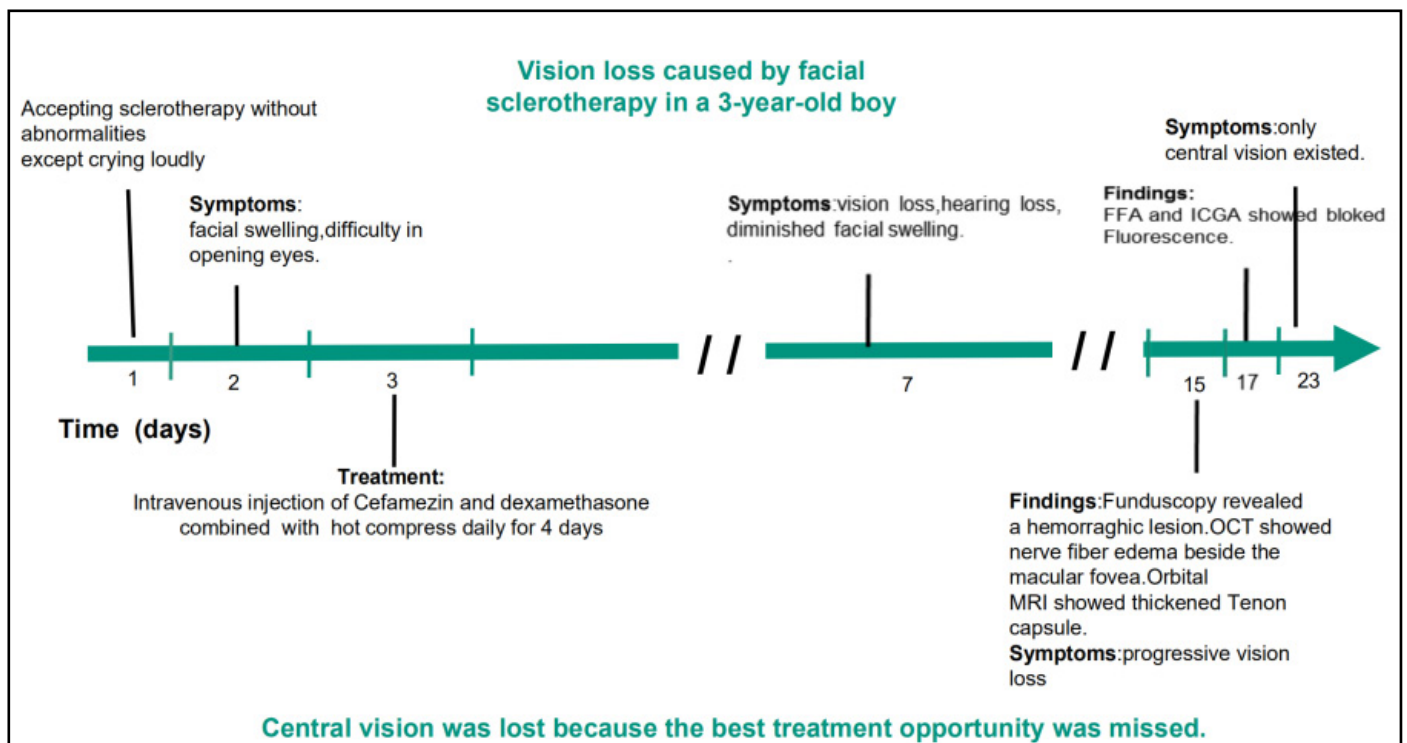
In conclusion, sclerotherapy for facial subcutaneous vascular malformations near to the ocular orbit might cause some rare but severe complications such as retina and optic nerve toxicity or inflammation inducing retinal hemorrhage, optic disc swollen (hyperreflective leakage in FA), and causing the visual acuity damage and hearing

injury. The related examination including ophthalmic investigations after the sclerotherapy is necessary and immediate treatment is important to the prognosis.

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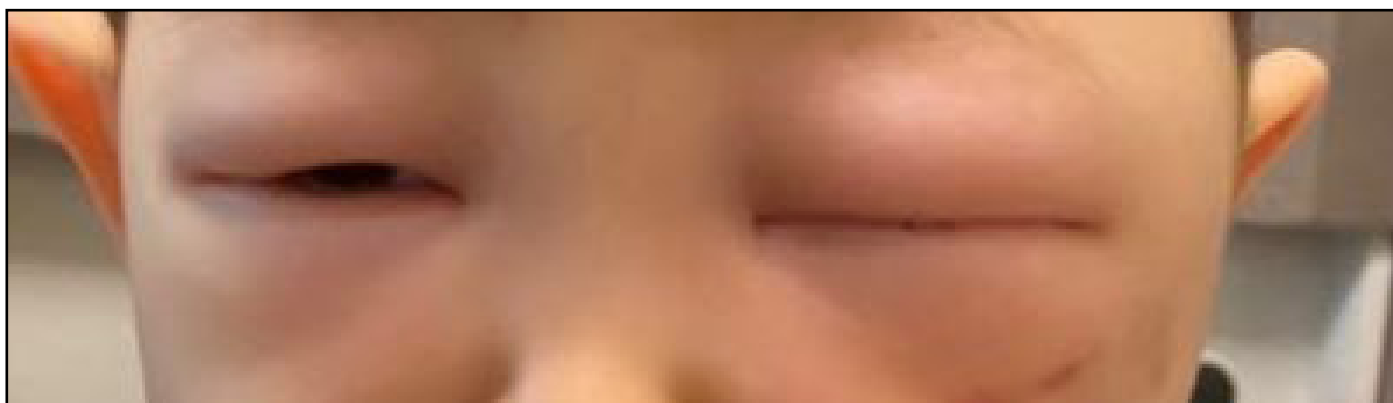
Figures



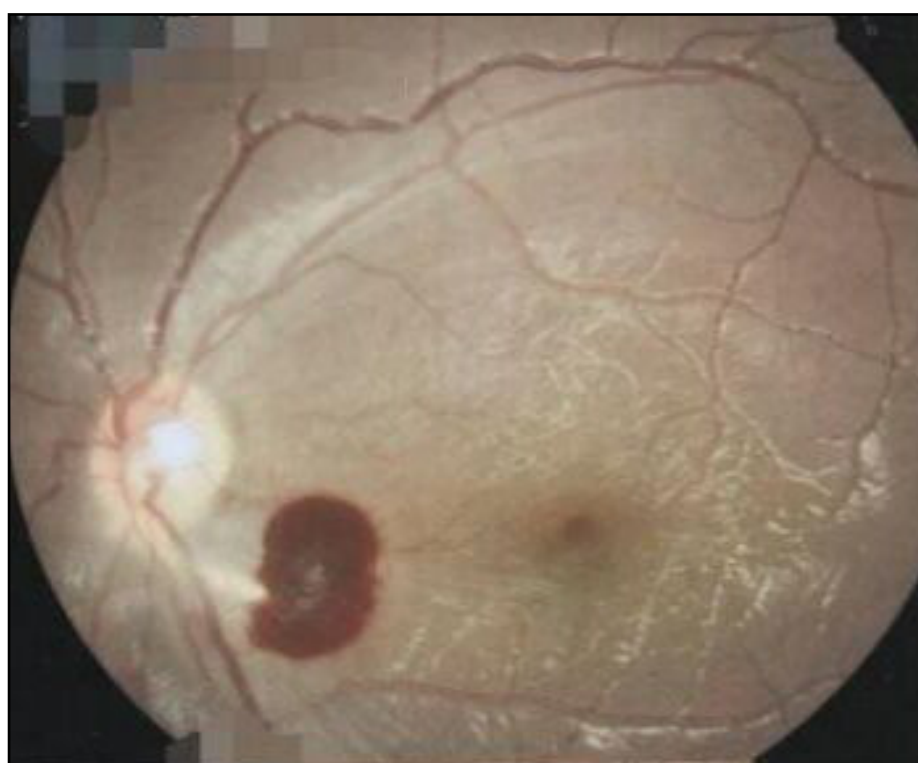
**Figure 1:** Clinical Timeline: Vision loss in a 3-year-old boy caused by facial sclerotherapy. FFA: Fundus Fluorescence Angiography; ICGA: Indocyanine Green Angiography; OCT: Optical Coherence Tomography; MRI: Magnetic Resonance Imaging



**Figure 2:** A color doppler ultrasonography subcutaneous scan: A low-echo mass in the left cheek with the size of 1.6\*0.8 cm



**Figure 3:** Lid swelling on both sides and hemifacial swelling on the left side



**Figure 4:** Subcutaneous B-scan: A low-echo mass in the left cheek with the size of 1.6\*0.8 cm

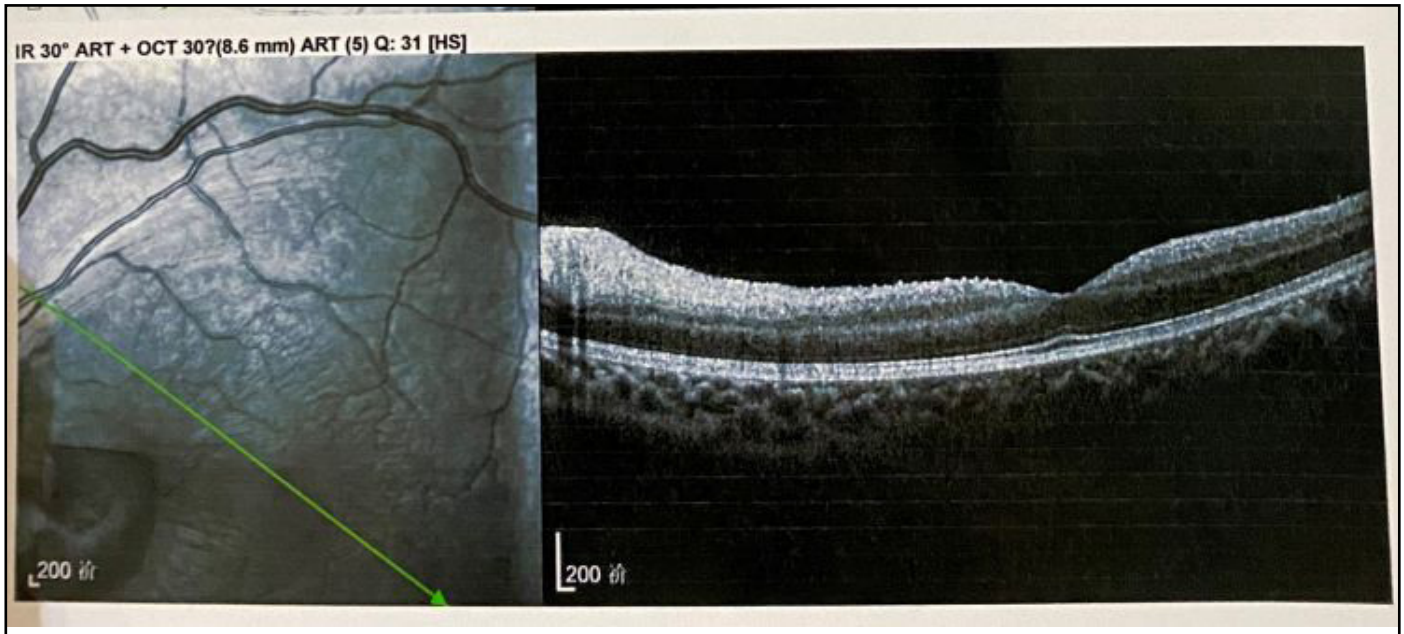


Figure 5: The OCT showed nerve fiber edema between the disk and macular.



Figure 6: The orbital MRI showed normal extraocular muscles (EOM) in the bilateral orbit after 7 days



**Figure 7:** FFA and ICGA of the left eye. Hyperreflective leakage on the optic disk could be seen.