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Case Report

Giant complicated glosso-cervical arterio venous malformation managed with external carotid artery ligation and serial injection sclerotherapy: a case report and review of literature

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ABSTRACT

Giant arterio venous malformation (AVM) of the tongue and floor of mouth are rare. They become life threatening when ignored by the patients. Treatment protocols are not well established. This report describes the treatment of a 35 year old female who presented to us with complains of swelling of tongue, floor of mouth and left sub mandibular region since 15 years causing dysphagia, and oral bleed since one day. Magnetic resonance Angiography revealed a giant AVM at the above site with dilated left external carotid artery (ECA) and left lingual artery. Patient underwent ECA ligation under general anaesthesia and serial injection sclerotherapy at the local site. After 3 months and 6 cycles of sclerotherapy, the patient had a satisfactory outcome. A detailed report with review of literature is presented.

Keywords: External carotid artery ligation, Giant arterio venous malformation, Glosso-cervival arterio venous malformation, Sclerotherapy, Sodium tetradecyl sulphate

INTRODUCTION

Vascular malformations are anomalies of the vascular system that typically occur at birth; however, sometimes they are visible only later in life.[1,2] Normally, they show a size proportional growth without regression. Vascular malformations can be differentiated into high-flow lesions that include arterial and arteriovenous malformations and low flow lesions which include lymphatic, venous and capillary malformations.[3] In the oral cavity, these can present at any site, but most commonly occur on anterior two-third of the tongue, palate, and gingival and buccal mucosa.[4]

Trauma, ischemic event secondary to thrombosis, ectasia, hormonal changes, and puberty can induce proliferation of the arterio venous malformation (AVM) and trigger the growth of the lesion and manifestation of its troublesome symptoms.[5] Typical symptoms of vascular malformations of tongue are bleeding, pain and swelling as well as impairment of speaking, swallowing or even breathing. Although the pathogenetic mechanisms of AVMs are not completely understood, the hemodynamic alterations that lead to the clinical manifestations of AVMs have been described well. An abnormal communication causes shunting of blood from the high-pressure arterial side to the low-pressure venous side. This creates an abnormal low resistance circuit that steals from the high resistance normal capillary bed.

As spontaneous regression of vascular malformations cannot be expected, therapeutic decisions have to be made. Treatment options are surgical excision, laser therapy, embolization and sclerotherapy.

In this case of a giant glosso-cervical AVM the authors performed a left ECA ligation followed by serial injection sclerotherapy at the local site by using sodium tetradecyl sulphate (STS) over 3 months.

There was significant decrease in size of the malformation and the patient was satisfied with treatment outcome both functionally and aesthetically.

CASE REPORT

A 35-year-old female presented to the emergency department with complaints of oral bleed since 1 day, difficulty in breathing since 3 days, dysphagia since 4 years, swelling in tongue, floor of mouth and neck since 13 years. Emergency tracheotomy was done to secure the airway and prevent aspiration of blood.

On examination there was swollen tongue filling the entire oral commissure (Figure 1), firm on touch, non-tender, with overlying surface dried up due to continuous exposure.

Neck examination revealed a 6×5 cm rubbery mass in left submandibular region, which was soft on touch and was non-tender. Bruit was present. Small bleeding points could be seen on undersurface of tongue which were in contact with lower dentition.

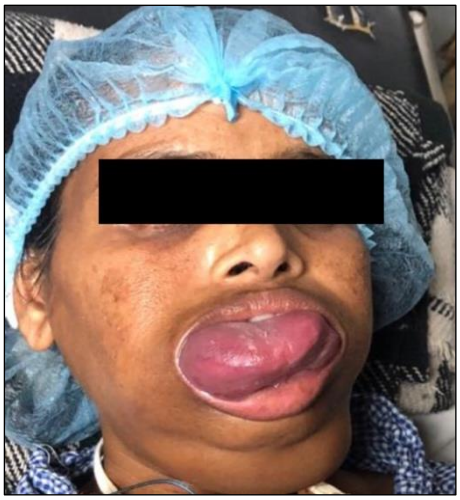

Figure 1: Day 1 of presentation.

Patient could not take even liquid food, because the enlarged tongue occluded almost whole of the mouth.

A magnetic resonance angiography (Figure 2) was done which revealed Av malformation of tongue, floor of mouth and left submandibular region of size 7.8×7.5×4.9 cm with dilated left lingual and left external carotid artery. The lesion had heterogeneous signal intensity with multiple flow voids. An impression of Giant Glosso-cervical AVM was made.

Patient was taken up for for left external carotid artery ligation under general anesthesia and percutaneous injection sclerotherapy with sodium tetradecyl sulphate at the local site.

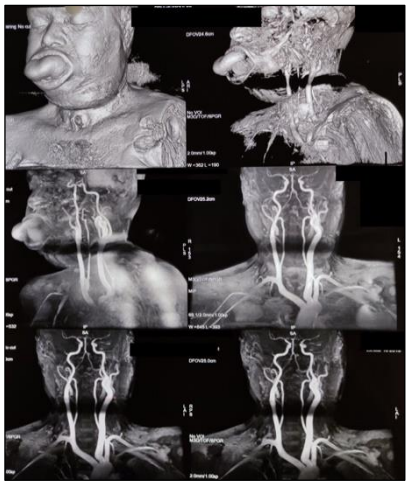

Figure 2: Magnetic resonance angiography image showing the AVM with dilated left ECA and left lingual artery.

A horizontal skin incision was made between the hyoid bone and the superior border of the thyroid cartilage. Subplatysmal skin flaps were raised, and the sternocleidomastoid muscle was retracted posteriorly.

Next, the carotid sheath was opened, and its contents were exposed. The ECA was identified by following the Internal Carotid Artery (ICA) for a few centimeters. After dissecting the ECA beyond its first branch. i.e. superior thyroid artery, it was ligated.

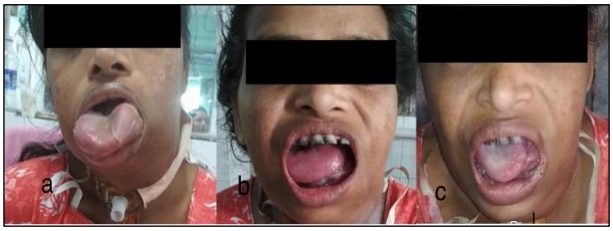

Figure 3: Post ECA ligation and serial injection sclerotherapy a. 2 cycles (1 month) b. 4 cycles (2 month) c. 6 cycles (3 month).

The 3% STS solution was injected into lesion using an insulin syringe and 26-gauge needle. The agent was delivered without radiological guidance. After introducing the needle, the plunger was withdrawn to look for backflow of blood and to confirm appropriate entry of the needle into the centre of the vascular space. The sclerosing agent was then injected until the lesion blanched. A minimum of 1 ml of the agent was injected into each lesion i.e. tongue floor of mouth and submandibular region.

The patients received an oral analgesic and a single postoperative dose of intramuscular (gluteal) injection of dexamethasone (4 mg) to decrease the inflammation and pain post injection.

The patient was followed up 2 weekly (Figure 3) and after 6 cycles of sclerotherapy the swelling reduced significantly, and the size of the tongue was reduced to near normal.

Response to treatment was assessed clinically and by using photographic assessment at each visit. Patient was satisfied with the functional and aesthetic outcome, and also reported a boosted morale and self-confidence.

DISCUSSION

AVM's are composed of a central nidus with anomalous arteriovenous shunts and a network of surrounding collateral vessels.[6] The short circuit or shunting between the high pressure arterial and low pressure venous system accounts for much of the clinical presentation, anatomical changes, and progression of the lesions.[7] In the oral cavity, these can present at any site, but most commonly on anterior two-thirds of tongue, leading to macroglossia and difficulty in mastication ,speech, and deglutition.

AVMs are by far the most difficult vascular anomaly to manage due to the replacement of normal tissue by disease vessels and very high recurrence rates.

Magnetic resonance imaging (MRI) has become the investigation of choice since it depicts the extent and lack of invasion of these lesions. Angiography is useful in poorly defined cases and for embolization before surgery.

Although the discovery of the therapeutic efficacy of propranolol in the management of infantile hemangiomas has revolutionized the care and outcomes of these lesions, there is no standard treatment protocol for the management of AVM.[8,9]

Treatment options of low flow AVM include simple observation, minimally invasive interventions such as sclerotherapy or embolization, laser ablation, cryotherapy, or more aggressive surgical resection.[10-12] Management of these lesions varies according to the patient's age, size of the lesion, location, type of the lesion, as well as surgeon experience.

Giant AVMs present a therapeutic challenge because of their hemodynamic characteristics and their modality of growth. Surgical resection in large AVMs is often associated with extensive blood loss and an incomplete resection frequently leads to regrowth of the tumor to sizes that are often larger than its original size.

The goal of therapy for such large lesions is to eliminate the malformation and conserve as much lingual tissue as possible. Complete surgical excision of these lesions is rarely possible without causing significant functional impairment and disfigurement.

Sclerotherapy is a viable option for the management of AVM's due to its safety, ease of administration, and acceptable aesthetic and functional outcomes. By using sclerotherapy, surgical intervention can often be avoided or at least minimized. Sodium tetradecyl sulphate, 3% STS has been widely uses as sclerosing agent in small varicose veins of legs, as well as venous and lymphatic malformations.[13,14]

The mechanism of action of STS is to produce maximum endothelial damage with minimal thrombus formation leading to fibrosis of the lesion which leads to shrinkage.

There are many other agents used for sclerotherapy: bleomycin, ethanolamine oleate, ethanol, hypertonic saline. The most common side effects are skin necrosis and ulceration, hypersensitivity, and swelling.

Stimpson et al, used 3% STS to treat 12 patients who had venous malformations in the head and neck.[15] They found that a single treatment may be adequate for small lesions, but the injections may be safely repeated until a satisfactory result is obtained in large lesions.

Soft tissue swelling generally increases in the region of the malformation immediately after the injections. Subsequently, necrosis and inflammation induced by the sclerosis subsides with fibrous tissue formation, culminating in progressive reduction in the lesion size.

CONCLUSION

In conclusion, sclerotherapy can be performed as an alternative or in addition to surgery. In many patients, multiple injections have to be performed. However, a significant size reduction helps the patient both functionally and aesthetically and also makes subsequent surgical resection easier if at all required.

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