malignancy was strengthened when the symptoms of Gradenigo syndrome was associated with symptoms like dysphagia, nasal regurgitation of fluid and intermittent aspiration and cough indicating involvement of the left V, VI, VIII and X cranial nerves.

Nasopharyngeal carcinomas are epithelial neoplasms that are seen only sporadically in the Western hemisphere. In southern China, however, they are endemic; they represent the third most common malignancy in men there. This carcinoma is also common in Indian subcontinent. It has a bimodal presentation but the main problem is late diagnosis.^{3,5} They may present in various ways as such as cervical lymphadenopathy, multiple cranial nerve palsies nasal discharge, epistaxis etc. However, the presentation primarily with Gradenigo's syndrome is indeed a rare one.

CONCLUSIONS

Diagnosis of nasopharyngeal carcinoma requires a high index of suspicion in view of its initial, sparse, non-specific symptoms. Although ideally the disease should be detected in its early stages, we believe that it is useful to recommend that in cases of Gradenigo's syndrome a full systematic otorhinolaryngological exploration be made to effectively rule out this disorder.

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Short Communications

MACROGLOSSIA DUE TO VENOUS MALFORMATION

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Abstract : Massive vascular malformation involving tongue can cause significant functional impairment. In this report we describe a rare case of extensive venous malformation involving tongue leading to obstructive sleep apnoea, inability to speak, eat, severe discomfort due to exposure induced dryness and ulceration. Multimodality treatment approach comprising of sclerotherapy, electrocautery and radiation therapy was used in this case.

Keywords: Macroglossia, venous malformation, Sclerotherapy, Electrocautery, Radiotherapy.

INTRODUCTION

Vascular malformations are extremely common lesions, accounting for approximately 7% of all benign tumors. Although the head and neck constitute less than 14% of the total surface area of the body, approximately 50% of all vascular malformations occur in this region⁴. Venous malformation is a soft, compressible, non-pulsatile mass with rapid refilling. Expansion occurs on compression of the jugular vein or valsalva maneuver. In addition to the occasional devastating cosmetic effects of large vascular formation, some may cause significant functional impairment by encroaching on the eye, tongue or throat. They pose a challenging therapeutic and reconstructive problem. Treatment options are variable and include sclerotherapy, photocoagulation and surgical resection.

CASE REPORT

A 14-year old male presented to us with complaints of an enlarged tongue with difficulty in speaking. There was history of recurrent ulceration of tongue with odynophagia. Patient also complained of recurrent episodes of breathing difficulty during sleep. On examination anterior two-third of tongue was enlarged with bluish discoloration, non-tender, soft, and spongy in consistency, compressible with rapid refilling. There was no palpable thrill or bruit on auscultation. Base of tongue was uninvolved. Indirect laryngoscopy was normal. Swelling had areas of phlebothrombosis. Swelling used to expand during sleep, valsalva maneuver and on bilateral jugular vein compression. CT Angiography showed generalized hyper intense contrast uptake with normal arterial phase, macroglossia involving anterior two-

Fig 1: Pre intervention

Fig 2: Post intervention

third tongue and sparing base of tongue. No single feeding vessel was identified. Delayed emptying phase with contrast retention was suggestive of venous malformation.

The patient was treated with sclerotherapy with sodium tetradecyl sulfate twelve injections over 6 months were administered. There was moderate improvement in size however dysarthria persisted. Patient was given an option of reduction glossectomy as the vascularity had decreased and bulk reduction could have helped but he refused surgery. This was followed by Submucosal cauterization using unipolar electrode. Ten applications were given over a period of 6 months. Significant improvement was seen initially however size started increasing slowly after 6 months. Patient was then referred for radiotherapy. He received 35Gy over 10 weeks. Patient initially complained of radiation induced dryness and ulceration which subsided on conservative management. There was significant decrease in size of tongue and dysarthria, sleep apnoea improved. Patient is comfortable and is in follow up 1 year after radiotherapy.

DISCUSSION

Venous malformations are present at birth, grow proportionately to the child, and often enlarge during puberty. Venous malformations present in a spectrum, ranging from an isolated skin varicosity or localized spongy mass to complex lesions infiltrating various tissue planes. Venous malformations have been treated by a variety of techniques over the years, including irradiation, electrocoagulation, cryotherapy, intravascular magnesium or copper needles, surgical excision, lasers, compression, and sclerotherapy^[3,4,5]. All these techniques have their particular indications and limitations. Treatment of Venous malformations is based on the location, appearance, and complications such as pain, bleeding, and associated functional problems. Surgical excision is useful only for localised and limited lesions. Aggressive excision can lead to significant loss of motor function, cosmetic problems, nerve damage, or massive bleeding in patients with extensive involvement because of the complicated anatomy of the head and neck region. Motor dysfunction due to hypoglossal nerve injury, anesthesia of the

tongue due to lingual nerve injury, decreased tongue mobility due to scarring or residual speech and masticatory problems are risks of reduction glossectomy. Larger resections involve greater risk of complications and more postoperative morbidity. Some patients as in this case might not give consent for glossectomy as the thought itself about the procedure and the possible morbidity postoperatively can be quite distressing.

Sclerotherapy has the advantage of no external scarring, and few complications in comparison with surgical treatment. There are various choices of agents for sclerotherapy i.e. 5% sodium morrhuate, sodium tetradecyl sulphate, ethanolamine oleate, OK432, bleomycin, ethanol, and hypertonic saline, alone or in various combinations, have all been used^{1,3}. There is no ideal vaso-occlusive substance applicable to all venous malformations. Ethanol sclerotherapy requires general anesthesia because the procedure is very painful. Potential complications of sclerotherapy include local skin necrosis, transient nerve palsy, haemoglobinuria, blood loss, and anaphylaxis^[1,3,5]. There has been a debate about the use of radiotherapy in benign diseases especially in children and adolescents for the fear of potential malignant transformation later in life. However it has been justified in selected resistant cases which do not respond to other modalities and are not amenable to surgical resection. Acutely, radiation damage manifests as cutaneous erythema, tissue edema, bleeding, and delayed tissue healing. Often multiple episodes of erythema occur during the first month of treatment. This is attributable to both local extra capillary cell injury and direct capillary damage. These effects are most often temporary and respond well to conservative management.

In this case we used a multimodality therapeutic approach comprising of sclerotherapy, electrocoagulation followed by radiotherapy. This approach was chosen as the lesion was large and no single modality successfully managed the problem. However author is of the opinion that instead of using different approaches sequentially, it would have been more effective to use them simultaneously. This may however cause more severe side effects. Initial improvement seen after sclerotherapy did not

relieve some of the symptoms of patient like dysarthria in spite of moderate reduction in the size of tongue. This was then followed by electrocoagulation which improved the condition but soon after size again started increasing. It was at this juncture that the radiotherapy was resorted to as a last option since patient had refused surgical resection. There was a significant improvement in the symptoms and patient is able to articulate well. There are no episodes of sleep apnea and patient is able to eat without any discomfort. Besides mild ulceration following radiotherapy, which subsided, on conservative management no complications were encountered in this case with any of the given therapies till now.

CONCLUSION

Large venous malformation of tongue is challenging lesions to treat, due to high vascularity and serious risk of airway compromise. Management involves different approaches including sclerotherapy, electrocoagulation, laser ablation, surgery and possibly radiotherapy. Treatment should preferably involve multimodality approach as no single modality gives complete solution to the problem.

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Short Communications

LYMPHANGIOMA TONGUE

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Abstract: We present a rare case of Lymphangioma tongue which presented to us as macroglossia and wide excision of the lesion was done. We also reviewed the literature and an attempt to draw a meaningful conclusion regarding treatment has been done.

Keywords : Lymphangioma, Macroglossia, Lymphangioma Tongue, Sleep - Apnea, V-Glossoplasty

INTRODUCTION

Lymphangiomas are congenital in origin and are hamartomas rather than true neoplasm's. They either originate as a result of sequestration of lymphatic tissue derived from portions of primitive sacs with no connections to lymphatic system. According to another school of thought, they may arise from endothelial fibrillar membranes which sprout from the wall of the cyst, penetrate the surrounding tissue, canalize and produce more cysts.

Lymphangiomas usually present at birth. Thirty five percent of Lymphangiomas of all types occur in cheek, tongue, floor of mouth; twenty five percent in neck; fifteen percent in axilla; rest in other places like pectoral region, mediastinum, groin, pelvis and retroperitoneum.

The typical Lymphangioma of oral cavity is characterized by irregular nodularity of mucosa with gray and pink projections. Most of these tumors manifest themselves at birth and / or shortly afterwards. The lesion may present with symptoms and signs viz. stridor, mediastinal widening, branchial plexus compression. Other presentations may be spontaneous hemorrhage, infection, or as in present case macroglossia leading to compromise in all the functions of tongue and oral cavity e.g. swallowing, mandible-dental deformities⁽¹⁾, speech disturbances, respiratory disturbances, and last but not the least, cosmetic defects.

CASE REPORT

An eight year female child presented in our OPD with enlarged tongue, to the extent that it protruded out of the mouth. The