CASE REPORT

An interesting case of enlarged head size in a patient with squamous cell carcinoma of tongue

Bhumika Vaishnav[1], Pailla Ruchitha[1], Aniruddh N Wadivkar[1], Saish Mondkar[1]*

[1]Department of General Medicine, Dr. D. Y. Patil Medical College, Hospital and Research Center, Dr. D. Y. Patil Vidyapeeth (Deemed to be University), Pimpri, Pune -411018 (Maharashtra) India

Abstract

Head and Neck Lymphedema (HNL) is one of the complications of Head and Neck Carcinoma (HNC). It can be a consequence of the disease progression or can occur as a complication of HNC management. HNL patients not only have facial disfigurement but also have psychological effects and poor quality of life. Severe lymph edema can cause impairment in speaking, chewing, and hearing. The gold standard treatment for HNL is Complete Decongestive Therapy (CDT). We present a case of a 59-year-old male with metastatic squamous cell carcinoma of the tongue who came with complaints of painless and gradual swelling of the face, lips, eyelids, and neck leading to doubling of the head size. He had severe lymphedema of the head and neck which was refractory to treatment.

Keywords: Head and Neck Neoplasm, Lymphedema, Carcinoma, Squamous Cell, Tongue Neoplasm

Introduction

Lymphedema-swelling caused by impaired tissue drainage as a result of lymphatic dysfunction, has long been recognized as a potentially serious disease sequel or a complication of treatment for patients with various malignancies. It is more commonly associated with genitourinary and breast malignancies where patients have lymphedema of genitalia and limbs respectively [1]. Globally, the prevalence of Head and Neck Cancer (HNC) is 3-5% [2]. Treatment options for HNC include surgery, radiation therapy, and chemotherapy. Several painful, function-limiting, and systemic side effects occur due to HNC and its treatment. Head and Neck Lymphedema (HNL) is an under-studied and undertreated complication of HNC [3]. We treated a patient of metastatic tongue adeno-carcinoma who developed severe HNL, resistant to all treatment modalities, leading to serious physical and psychological impairment. This case highlights the importance of recognizing this condition early on in the course of therapy so that such devastating physical and mental morbidity can be prevented.

Case Report

A 59-year-old male was admitted, with complaints of face and eyelids swelling for 21 days. The patient developed facial swelling involving lips and eyelids which later on progressed and involved the cheeks, forehead, ears, and neck. The swelling was non-itchy, painless, and associated with an increase in the size of the head. The patient could not see, hear or speak due to the swelling since seven days. He had no history of fever, cough, breathlessness, trauma or insect bite. The patient was a case of squamous cell carcinoma of the tongue since one year. He had undergone right hemi-glossectomy and received 25 cycles of radiotherapy.

Six months before presentation, the patient had a local recurrence of the tumor for which he

underwent modified radical neck dissection. Post-operatively he was on mechanical ventilation for a prolonged period and had a tracheostomy *in-situ*. The patient was later started on chemotherapy with Cis-Platinum and 5-Fluorouracil (5 cycles completed before admission).

On local examination, the head size was increased; the facial swelling involved the eyelids, lips, pinna, and cheeks (Figures 1, 2). The oral cavity and the eyes could not be examined due to the swelling. Skin over the swelling was stretched, and shiny without rashes or discharging sinuses, or any local rise in temperature. General and systemic examination was normal.

On admission, laboratory parameters- complete hemogram, renal, liver and thyroid function tests, serum electrolytes, ECG, chest X-ray, ultrasound of abdomen and pelvis were normal. Absolute eosinophil count and serum IgE levels were normal.

Figure 1: Facial swelling on admission

Figure 2: Relative size of the head compared to the rest of the body

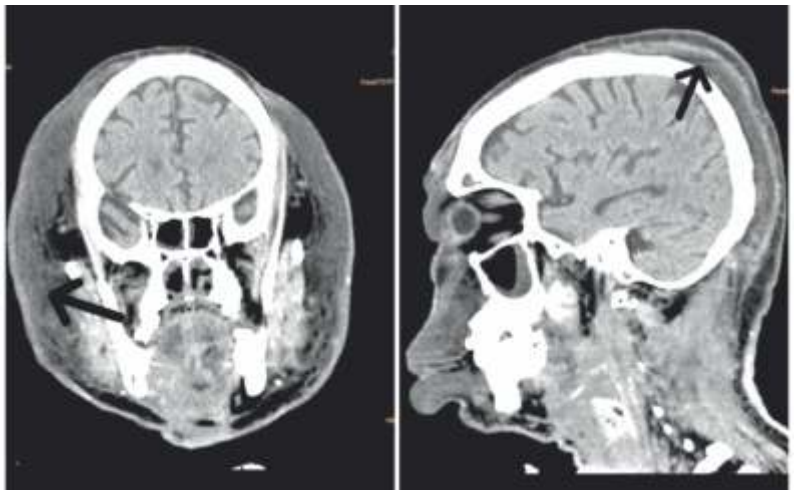

Figure 3: CT images of thickened skin with subcutaneous edema of the scalp and face (black arrow)

Computed Tomography (CT) with contrast of the face, neck showed post-modified radical neck dissection status with markedly thickened skin with diffuse subcutaneous edema in the neck, entire scalp, upper-lip, pre septal compartment of both orbits, bilateral eyelids, and face with glossomegaly suggestive of lymphedema (Figures 3 and 4). There was no evidence of neck vessel thrombosis. CT chest showed multiple small soft tissue density lesions of size 3-4 mm scattered in both lungs suggestive of lung metastasis.

Over the course of hospitalization, treatment for facial lymphedema was initiated with the assistance of lymphedema therapist which included manual lymph drainage, a pneumatic compression device for face, and dermatological consultation to prevent the skin changes associated with lymphedema. But the patient did not respond these treatment modalities and there was no improvement in the lymphedema.

On day 10 of admission, patient developed fever spikes and hypoxia with right lung lower zone consolidation due to Hospital Acquired Pneumonia (HAP). He was started on antibiotics and supportive measures. On day 19, patient's general condition worsened and he succumbed to sepsis (pneumonia with respiratory failure) and septic shock. The final diagnosis was metastatic squamous cell carcinoma of the tongue complicated with head and neck lymphedema (Foldi's stage III/M D Anderson Cancer Centre Lymphedema Stage III) with HAP and septic shock.

Discussion

Lymphedema occurs due to the accumulation of protein-rich lymphatic fluid in the interstitium and fibro-adipose tissues of the body causing swelling of the involved area, restriction of movements, discomfort/pain, and skin changes. It can be primary or secondary to injury or obstruction to the lymphatic system. Lymphedema secondary to malignancies is common and can be due to lymphatic obstruction by the tumor mass, lymph-node metastasis, post-operatively after lymph-node excision, local radiotherapy or systemic chemotherapy. Lymphedema is most commonly seen in breast malignancies [4]. Pathophysiology of lymphedema includes stasis of lymph, remodeling of lymph vessels with inflammation and finally adipose tissue deposition, and fibrosis [5].

HNC are more commonly seen in patients with tobacco, betel nut and alcohol addictions and are often associated with locoregional complications like HNL, which can further deteriorate the quality of life in these patients [6]. HNL post-HNC is usually limited to the neck region (90%) and involvement of the face and eyelids is relatively uncommon [7]. HNL has physical as well as psychological effects on the patients including cosmetic impairment and poor quality of life. Severe HNL can cause impairment of speech, hearing and vision as was the case in our patient [8]. Intraoral and pharyngeal edema may cause dysphagia and poor intraoral hygiene [9]. Differential diagnoses for HNL includes superior vena cava syndrome, allergic reactions like medication/chemotherapy-induced angioedema, and cellulitis due to local infection.

HNL is one of the most under-recognized and under-treated complication of HNC. Due to multiple implications of this condition, it is imperative to find an adequate therapeutic strategy for the patients. Management of HNL includes Complete Decongestive Therapy (CDT) or Decongestive Lymphedema Therapy (DLT), Manual Lymph Drainage (MLD), Compression bandages, skin-

care, physiotherapy and microsurgical techniques like vascularised lymph node transfer, lymphatico-venous anastomoses and suction assisted protein lipectomy [10]. CDT is considered the gold standard in management of HNL and aims at decongesting the edematous surface and improving lymph drainage**.** It is administered by a certified lymphedema therapist in two phases; an intensive phase of outpatient treatment is provided 3–5 days weekly over a period of 2–4 weeks followed by the maintenance phase at home [7].

Successful treatment of HNL depends on several factors like staging of the malignant disease, patient co-operation, compliance to the OPD and home-based CDT regimes and initial staging of the lymphedema. Our patient had severe stage III lymphedema with stage IV malignant disease and thus did not respond at all to various treatment options. HNL post-HNC is difficult to cure. Early diagnosis and immediate treatment measures, as mentioned above, can limit the physical and psychological side effects of this condition. A team approach which includes a clinician, oncologist, lymphedema therapist, psychiatrist, and a good nursing care support is needed for this often neglected complication of HNC.

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Author for Correspondence:

Dr. Aniruddh N Wadivkar, Department of General Medicine, Dr. D. Y. Patil Medical College, Hospital & Research Centre, Sant Tukaram Nagar, Pimpri Colony, Pune- 411018 Email: anwrealfc@gmail.com
Cell: 9619375064

How to cite this article:

Vaishnav B, Pailla R, Wadivkar AN, Mondkar S. An interesting case of enlarged head size in a patient with squamous cell carcinoma of tongue. *J Krishna Inst Med Sci Univ* 2023; 12(4):160-163

Submitted: *27-July-2023* ***Accepted:*** *27-Aug-2023* ***Published:*** *01-Oct-2023*