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Dinesh Kumar GR**

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Case Report: Herein, we report such a unique case where in three simultaneous neoplasms were diagnosed in single patient, with histology of different tissue origin and managed successfully by surgery.

Conclusion: The risk of occurrence of new tumors in patients with multiple primary tumors should always be considered. Triple simultaneous primary tumors diagnosis reduces the incidence of secondary metastasis. It is essential to have regular checkups for better outcome of these patients.

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CASE REPORT

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Introduction: Synchronous multiple tumors in the head and neck, has been reported to be around 0.5%. The development of simultaneous triple primary malignant neoplasms of different histology, confined to the head and neck region reported till now is very rare. **Case Report:** Herein, we report such a unique case where in three simultaneous neoplasms were diagnosed in single patient, with histology of different tissue origin and managed successfully by surgery. **Conclusion:** The risk of occurrence of new tumors in patients with multiple primary tumors should always be considered. Triple simultaneous primary tumors diagnosis reduces the incidence of secondary metastasis. It is essential to have regular checkups for better outcome of these patients.

Keywords: Triple primary tumors, Different histology, Parapharyngeal mass, Thyroid carcinoma, Basal cell carcinoma

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INTRODUCTION

Multiple malignant neoplasms are described based on their chronology of presentation as simultaneous, synchronous or metachronous. Tumors presenting at the same time in the initial examination are classified as simultaneous tumors, a terminology which has recently been introduced [1]. Tumors presenting within six months of diagnosis of index tumor is termed synchronous tumors. Metachronous tumors are those presenting after six months of diagnosis of index tumor. It is rare for a patient to suffer from three primary simultaneous malignancies of different histology. It is even rarer for these tumors to present at the same time in a confined anatomical region such as head and neck area. The prognosis of patients suffering from multiple primary cancer in head and neck is poor and early detection is essential. The development of second primary tumor was of equivalent prognosis to recurrence of primary tumor. The diagnosis of three simultaneous tumors during single visit, reduces the morbidity associated with the management of loco regionally advanced stage of the disease, and prolongs the survival thereby providing favorable outcome.

Hemavathi Umeshappa¹, Chandrashekhar M², Ashok M Shenoy³, Dinesh Kumar GR⁴

Affiliations: ¹MDS, FHNS, Maxillofacial Surgery, Fellow in Head and Neck Surgical Oncology, Registrar in Surgical Oncology, Apollo Hospital, Bannerghatta Road, Bangalore, Karnataka, India; ²MS, FICS, Surgical Oncologist, Senior Consultant, Department of Surgical Oncology, Apollo Hospital, Bannerghatta Road, Bangalore, Karnataka, India; ³MS, ENT and Surgical Oncologist, Senior Consultant, Department of Surgical Oncology, Apollo Hospital, Bannerghatta Road, Bangalore, Karnataka, India; ⁴MS, Junior Consultant, Department of Surgical Oncology, Apollo Hospital, Bannerghatta Road, Bangalore, Karnataka, India.
Corresponding Author: Dr. Hemavathi Umeshappa, MDS, FHNS, Maxillofacial Surgery, Fellow in Head and Neck Surgical Oncology, Registrar in Surgical Oncology, Apollo Hospital, Bannerghatta Road, Bangalore, Karnataka, India; Mobile: 09663906416; Email: drhemau85@gmail.com

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CASE REPORT

A 63-year-old female visited us with a chief complaint of swelling over the left side of neck since 16 years. Patient noticed mass in neck which was initially small and gradually increased to present size. She did not have difficulty in breathing or hoarseness of voice. Patient developed symptoms of dysarthria and difficulty in swallowing since six months. Patient does not have the history of exposure to ionizing radiation or tobacco habits or familial predisposition.

Clinical examination revealed an extra oral mass of size 5x6 cm involving the parapharyngeal space. There was no rise in ear lobule. No signs of facial nerve involvement was present. On palpation of neck lymph nodes were not enlarged. A second lesion of size 1x1 cm involving on the right side of the upper lip. A solitary lobulated, midline swelling of size 3.5x4.0 cm involving left lobe of the thyroid gland. Swelling moves with deglutition, non tender on palpation but the inferior border of thyroid gland was indistinct suggesting substernal extension. Mouth opening was adequate, intraorally there was bulging of pharyngeal mucosa of oropharynx with shift of uvula to right side (Figure 1).

Computed tomography (CT) scan showed 9.1x7.8 cm well circumscribed heterogeneously enhancing lobulated mass in the left upper neck predominantly involving the left parapharyngeal space probably arising from ectopic salivary tissue. No extension to involve the vagus nerve, IJV or carotid vessels. Another lesion of size 6.5x4.3 cm nodule in left lobe of thyroid with retrosternal extension into superior mediastinum and tracheal compression (Figure 2). Fine-needle aspiration cytology of left parapharyngeal mass proved to be Adenoid cystic carcinoma, and left thyroid nodule suspicious of follicular neoplasm.

After preoperative workup, the patient underwent three operative procedures (Figure 3).

Left Parapharyngeal Mass: Excision of tumor through transparotid-transcervical approach of left parapharyngeal space with supra omohyoid neck dissection on left side. Intraoperatively, tumor was well encapsulated, measuring 9x8 cm involving left parapharyngeal space. The main trunk of facial nerve identified and its branches preserved. Deep lobe of parotid gland was separated and the tumor excised.

Left Thyroid Nodule: Left thyroid lobectomy was done. Frozen section reported as follicular carcinoma. Completion thyroidectomy with central compartment neck dissection done. Intraoperatively, tumor was encapsulated measuring 5.5x5 cm involving left thyroid gland, without extrathyroidal extension, Recurrent laryngeal nerve was free of tumor and preserved. Parathyroid glands identified and preserved.

Upper lip lesion: Wide local excision of lesion involving the right side of the upper lip, followed by reconstruction with local advancement flap was done.

Postoperatively, patient was on endotracheal airway for two days and recovery was uneventful. Facial nerve function was intact. There was no change in voice.

Histopathology of specimens proved to be

- (i) Adenoid cystic carcinoma of left parapharyngeal mass. Tumor size of 8x5 cm, no involvement of regional lymph nodes. Staging AJCC – pT3 pNo.
- (ii) Follicular carcinoma of thyroid gland. Tumor size of 6.0x5.5 cm, presence of capsular invasion, with no lymphatic invasion. Staging AJCC – pT3 pNo.
- (iii) Basal cell carcinoma of right upper lip. Tumor size of 1.5x1 cm.

Staging AJCC – pT1 pNx.

Based on HPR findings patient underwent radioactive iodine (I-131) therapy for thyroid and adjuvant radiotherapy for left parapharyngeal mass. Patient visited us after eight months for first follow-up after adjuvant therapy. Patient is free of disease and advised to come for regular follow-up biannually.

Figure 1: Preoperative image with lip lesion.

Figure 2: Computed tomography scan showing left parapharyngeal mass and retrosternal thyroid nodule.

Figure 3: Intraoperative image after excision of parapharyngeal mass and total thyroidectomy.

DISCUSSION

Multiple synchronous primary neoplasms are increasing in literature and reported as high as 10% [2].

Patients of head and neck malignancies have increased risk of developing synchronous primary tumors. Tumors presenting within six months of diagnosis of index tumor is termed synchronous tumors. Synchronous primary tumors are commonly seen in upper aerodigestive tract such as oesophagus, with oropharynx in head and neck as index tumor. Initial workup with imaging and endoscopy is useful to diagnose the incidental finding of synchronous tumors. Survival of synchronous tumors is lower than the metachronous tumors. Synchronous/simultaneous neoplasms arising in the upper aero digestive tract are very common based on the concept of field cancerization [3]. The dysplastic changes of the mucosa produces tumors of monoclonal origin which shows the obvious relationship with the etiological factors such as smoking or tobacco chewing habits or previous ionizing radiation [4].

Cases of metachronous multiple neoplasms are very commonly reported in literature. Forrest et al. described a metachronous nine primary neoplasms over a 16-year period which were all treated by surgery. Taylor and Torrence reported a woman who suffered from metachronous six malignancies, accompanied by failure due to metastatic disease [3]. Multiple primary simultaneous neoplasms occurring in single patient are unusual. Cases of simultaneous triple primary neoplasms diagnosed and successful surgical management is very rare.

In this case, we have diagnosed all three neoplasms clinically and histologically proved to be of different tissue origin located in three different sites of the head and neck during first visit of the patient. The patient does not have the history of exposure to ionizing radiation or tobacco habits. All three neoplasms were managed surgically in her first visit successfully. Simultaneous multiple primary tumors in head and neck region, surgery seems to be superior to radiotherapy or chemotherapy in that surgery offers an effective simple, quick, low morbidity approach, especially in early stage disease. Radiotherapy and chemotherapy are the alternative modalities available and can be reserved for the management of secondary tumors. Radiotherapy and chemotherapy impairs the immunity resulting in increased incidence of new tumor developing. The diagnosis of the multiple tumors of head and neck during single visit at early stage, the morbidity associated with the management of loco-regionally advanced malignancies with multi-modality approach and also reduced the number of surgeries under anesthesia if tumors were diagnosed in a subsequent follow-up. The prognosis of the patient suffering from multiple primary tumors in head and neck is poor and early detection of these cases is mandatory to extend the survival of patients with a good prognosis by providing effective therapy at the initial stage of the cancer [5]. As per the prognosis considered Adenoid cystic carcinoma has increased incidence of local recurrence 42% and distant metastasis if managed with radiotherapy. Surgical management has reduced incidence of local recurrence of

these tumors thereby improving the survival rate of these patients. Basal cell carcinoma has excellent prognosis as it rarely metastasize. Follicular thyroid carcinoma also shows favorable outcome with radioactive iodine therapy.

CONCLUSION

The presentation of triple primary simultaneous tumors in head and neck of different histology is very rare. It is even more unusual for these tumors to present at the same time in a confined anatomical region such as head and neck area. The occurrence of multiple primary tumors have high risk of developing new tumors, should always be considered in any patient with malignant disease, if treatment is to be effective as possible. Triple simultaneous primary neoplasms diagnosis is essential to reduce the incidence of secondary metastasis and improve the survival rates. Besides interventions, it is ultimately important to have regular checkups in each visit for better outcome of the these patients.

Author Contributions

Hemavathi Umeshappa – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Chandrashekhar M – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Ashok M Shenoy – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Dinesh Kumar GR – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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