

A patient with synchronous Head and Neck primaries, a squamous cell carcinoma of the tongue with papillary thyroid cancer - A case report



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ABSTRACT

Background: We report a rare case in Al-Zahra Cancer Center in Dubai of two Head and Neck malignancies in a 53 years old patient; Squamous cell carcinoma of the tongue and papillary thyroid carcinoma occurring as synchronous cancers in the left side of the neck. These two pathologically distinct malignancies of head and neck subsites in one patient is a rare phenomenon to be further evaluated.

Case presentation: Fifty three years old gentleman from Iraq presented with a painless left tongue ulcer with an indurated, irregular margin with elevated edges and necrotic floor, along with left thyroid suspicious nodule, left hemiglossectomy revealed an invasive moderately differentiated squamous cell carcinoma of the tongue pT2 and Left hemithyroidectomy done with well-differentiated papillary thyroid cancer follicular variant.

Conclusion: High index of suspicion and thorough work up is essential in follow up of patients with head and neck primary cancers. The importance of multidisciplinary team work should be stressed on in evaluating patients with Head and Neck cancer and the importance of proper staging before planning a surgery. However, which patients are at increased risk of double primaries, is still unknown.

Keywords:

thyroid, tongue, cancer, synchronous, doubles primaries

I. INTRODUCTION

The incidence of two primary cancers in one patient, whether synchronous (simultaneous) or metachronous (successive) is rare, especially if it comes to malignancies appearing in Head and Neck region. It is always important for the treating oncologists to keep high level of suspicion to the possibility of a synchronous or a metachronous malignancy in a Head and Neck cancer cases.

Patients with Head and Neck primary cancers have increased propensity of having second cancers especially in patients with tongue, pyriform sinus, larynx, oral cavity and tonsillar cancers [1].

Schwartz et al., observed 19% incidence of second cancers in the same region in patients presenting with primary Head and Neck cancers. Among them, 41% were synchronous and 59% were metachronous [2]. As the development of a second malignancy is almost always fatal, there is a need for additional efforts in work up, radiation treatment planning, goals of treatment and follow up schedules in such a group of patients [1,2].

Although there have been reports of second cancers arising within the context of primary Head and Neck cancer, the incidence of three Head and Neck primary cancers is extremely rare, if not extinct. Among Head and Neck tumors, nasopharynx, larynx and hypopharynx have been described as having increased incidence of triplicate malignancies [3].

The incidence of multiple primary malignant neoplasms increases with age and they are encountered more frequently nowadays than before owing to better diagnostic approaches. Although the scattered reports of developing three primaries have been described for other body sites, to our knowledge there is only one study in the literature describing three different malignancies in Head and Neck region in a Saudi patient [14].

Here-in we report a 53 year old gentleman having two primary head and neck malignancies, tongue and papillary thyroid carcinoma.

II. CASE PRESENTATION

Case report : 53 years old gentleman from Iraq lifelong nonsmoker nondrinker with no prior personal or family history of malignancies presented in May 2018 with a painless left tongue ulcer that has been developing over the last 3 months and he was evaluated by his dentist and was reassured it is a minor ulcer that will heal spontaneously, the ulcer continued to grow to more than 2 cm wide with an area of leukoplakia and induration (figure 1).



Figure 1: A malignant ulcer in the left side of the tongue with irregular margin and indurated base and necrotic floor.

On examination there was an induration of the left lateral part of the tongue with irregular margin with elevated edges and necrotic floor, however by palpation it was not extending to the base of the tongue nor the floor of the mouth with non-palpable cervical lymph nodes, a staging CT scan of the chest done showed no evidence of distant metastatic disease.

The patient had a biopsy and the histopathology confirmed a well-differentiated squamous cell carcinoma, however, while doing his workup they found of 1.2 cm nodule in the lower pole of the left thyroid and it is suspicious and considered TIRAD 4, ultrasound of the neck done revealed solid hyperechoic nodule appearing wider than tall and having few internal foci of calcifications noted in the lower pole left thyroid lobe the nodule measures 1.4×0.9 cm with bilateral sub-centimetre thick cervical lymph nodes.

Based on the discussion at the multidisciplinary oncology conference, the decision was to take a biopsy, however, it is considered doing it at the same operative setting and the left hemithyroidectomy with a frozen section to be considered.

MRI of the Head and Neck area showing a hyper intense irregular area at the left side of the tongue 2cm in maximum diameter (T2/T1 N0) (figure 2).

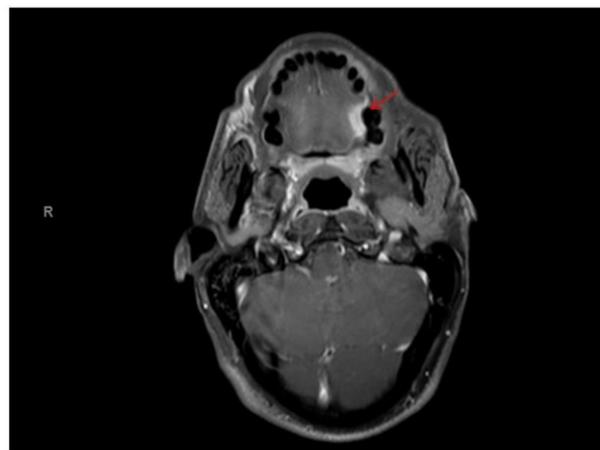


Figure 2: MRI of the Head and Neck area at the level of the tongue, pre contrast study showing a hyperintense irregular area at the left side of the tongue 2cm in maximum diameter (T2/T1 N0).

Pre-and postcontrast MRI of the neck with multiple pulse sequences in different planes done revealed an ill-defined focal abnormal signal intensity in T2 images involving the left side of the tongue opposite the first and second molar tooth associated surface indentation, ulceration and homogenous enhancement, after contrast study no infiltration seen to alveolar margin and it is not crossing the midline with no infiltration of the tongue base, the ulcer measures about 8 mm in thickness /transverse diameter, 18.1×17.8 mm anteroposteriorly and craniocaudad extension respectively with no evidence of buccal space invasion, not invasion nor cortical destruction of the related alveolar margin.

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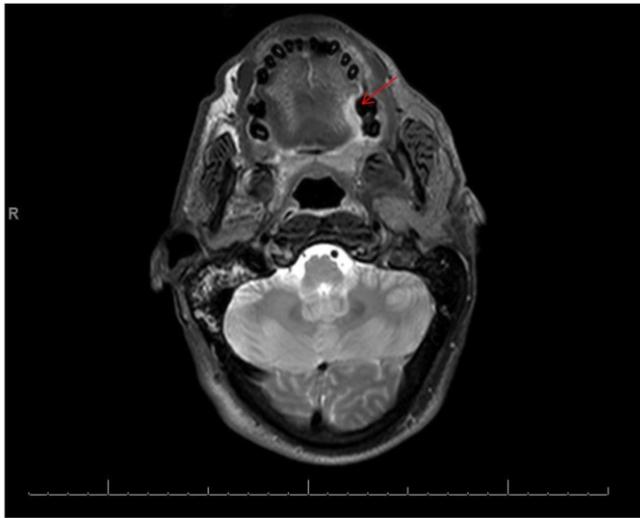


Figure 3: MRI of the Head and Neck area at the level of the tongue, post contrast study showing a hyperintense irregular area at the left side of the tongue 2cm in maximum diameter (T2/T1 N0).

Bilateral deep cervical lymph nodes at the level of I,II,III, IV, the largest on the left side in the left submandibular region measuring 14.3×9 mm.

The histopathological report of the fine needle aspiration cytology done from the ulcer base reveals invasive squamous cell carcinoma well differentiated grade 1, immunohistochemistry studies done:

CK AE1/AE 3: Strongly positive in 100% clone: AE1/AE3
 P 63: Strongly positive in 95% clone: DAK/P 63
 KI 67: Positive and 10%, clone: MIB 1 that suggests being triggered by HPV!

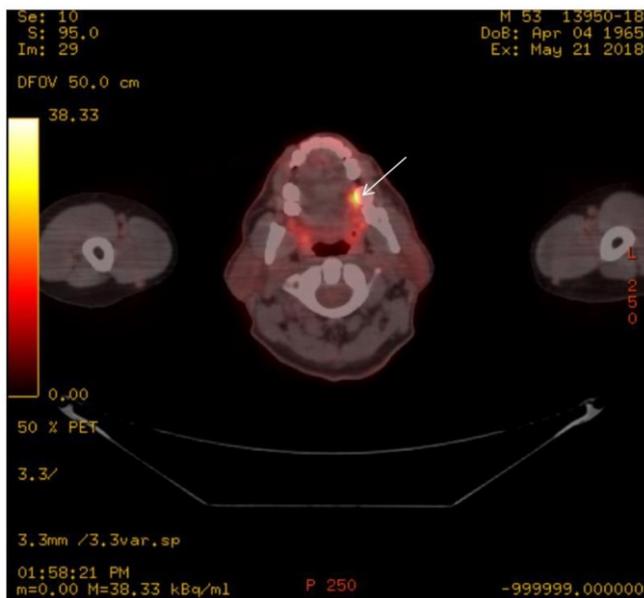


Figure 4: hypermetabolic lesion found is in the left side of the tongue SUV max 8 which is already diagnosed as squamous cell carcinoma

When a suspicious focus in the left thyroid lobe discovered TIRAD 4, a 18F-FDG PET/CT scan for the Head and Neck done to exclude other possible third primary and to evaluate the left thyroid lobe focus, surprisingly, the only hypermetabolic lesion found is in the left side of the tongue SUV max 8 which is already diagnosed as squamous cell carcinoma, and the thyroid gland- which harbors the papillary cancer- was completely normal!

Depending on these criteria the patient was subjected on 22-5-2018 to Left hemiglossectomy together with the left hemithyroidectomy with left supra-omohyoid cervical lymphadenectomy (I, II, III, IV), the frozen section biopsy of the left thyroid lobectomy reveals a follicular lesion pending paraffin section final pathology result which was papillary thyroid cancer follicular variant pT1.

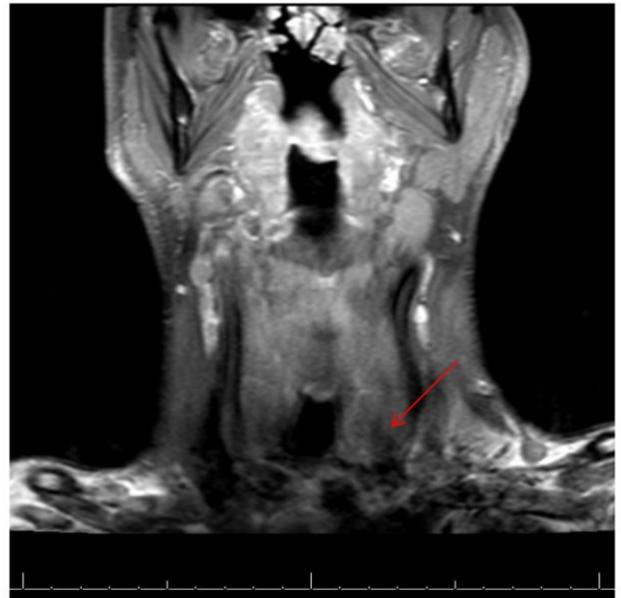


Figure 5: MRI of the Head and Neck, coronal cut, showing hypointense nodule at the left thyroid lobe.



Figure 6: Pre-operative marking of incision site, with nasotracheal intubation with intraoperative nerve stimulator device.

The final pathological results reveals invasive moderately differentiated squamous cell carcinoma of the tongue be T2 and the resection margins are negative, Left hemithyroidectomy with well-differentiated papillary thyroid cancer follicular variant, all resection margins are negative, left modified radical neck dissection with 23 lymph nodes identified all of them are negative for malignancy the largest lymph node is 1 cm.

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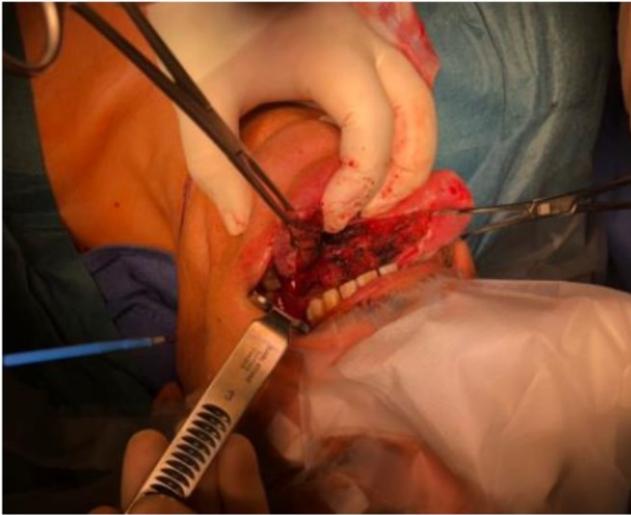


Figure 7: intra operative image with resection of the left tongue ulcer with counter traction of the right side.



Figure 8: Intra operative image with superior and inferior skin flaps elevated for the left selective neck dissection with identification and preservation of spinal accessory nerve, hypoglossal nerve, lingual nerve, ansa cervicalis.

III. DISCUSSION

Multiple primary cancers are a rare entity whose etiology is not well understood although some pathogenic factors like persistent carcinogenic influence, targeted therapy, increasing use of systemic chemotherapy or radiotherapy, hormonal manipulation, immune suppression, genetic alteration, tissue transplantation and improved survival are implicated [3]. The incidence of second primary metachronous tumor is increasing and reported as high as 10% in comparison to synchronous tumour which is much less (4%). A meta analyses shows the frequency of second primary tumor as 3-5%, a third primary tumor as 0.5%, and a fourth primary tumor as 0.3% [5].

The second tumor is called synchronous if it is found simultaneously or within six months of the diagnosis of primary. All malignancies found later than six months after the primary are considered as metachronous [6]. In a study by Panosetti et al. on 855 patients of multiple primary Head and Neck cancer patients, 42% had synchronous while 58% had metachronous tumors. Survival rates varied according to the treatment as evident by the fact that 5 year survival was only 8% among those synchronous tumors in whom treatment was modified whereas it was 28% in patients with unmodified treatment plans [7].

Another study done on 796 patients showed that second primaries were mostly found in esophagus, hypopharynx, buccal cavity, and the lung. Incidence of multiple cancers and synchronous primaries were more prevalent in prospective studies rather than in retrospective series. The five-year survival rate among patients with synchronous cancers in these two studies was 18% as compared to those with metachronous cancers (41- 55%). The prognosis for synchronous cancers deteriorated if the planned treatment had to be modified following the discovery of a second primary [6].

Individuals with a history of multiple cancers should have a complete family history evaluation and follow-up for development of subsequent primaries. Genetic counseling, risk estimation, and cancer screening must be emphasized. Every subsequent occurring tumor must be biopsied [11]. There are different diagnostic modalities to consider in such cases. Systematic pretherapeutic panendoscopy should be used to help detect early asymptomatic second primaries which might be missed during clinical examination [8].

Because of the relatively small proportion of malignant findings and lack of convincing data on its effect on survival rates, the value of panendoscopy has been debated. However, its significance is often crucial for the individual patient, despite the low proportion of positive findings. The study by Hujala K. et al. included 203 consecutive patients with squamous cell cancer of the upper aerodigestive tract who underwent panendoscopy in Turku University Central Hospital as part of the initial diagnostic workup from 1992–1999. Eight patients with synchronous second primaries and 19 patients with metachronous tumors were diagnosed [9].

Haughty et al. recommended routine interval endoscopic intervention within 2 years of treatment for optimum detection of second primaries in Head and Neck cancer patients. Also, a lifetime of clinical surveillance is suggested for aerodigestive tract second neoplasms in oral cavity, oropharynx, and hypopharynx cancer patients and for lung and non-aerodigestive tract neoplasms in larynx cancer patients based on their meta-analysis of second malignant tumors in Head and Neck cancers. Head and Neck second primary tumors were more common in oral cavity, oropharynx and hypopharynx [10].

Screening programs and chemoprevention strategies should be directed toward cancer patients with initial Head and Neck malignancies [9]. The effect of first tumor on the 2nd primary or vice versa is still not fully understood and need to be explored. The second primary tumor is usually more aggressive, more treatment resistant, and metastasizes early, requiring a more aggressive treatment strategy [11].

Our patient had synchronous tumors (tongue cancer and thyroid cancer) in the same time in the left side. It emphasizes the need for vigilant, regular and meticulous follow up of such patients along with appropriate imaging and biopsy techniques. The efficacy of 18F-FDG PET/CT has been discussed in detection of distant metastases in high risk Head and Neck primary cancers, but its role in detecting synchronous and metachronous lesions of the same region is not clear [12].

The role of panendoscopy and subsequent biopsy of suspicious lesion is more promising. Such early suspicious lesions should be biopsied as early as possible in order to offer best curative modality to these patients. The prognosis in such patients is obviously not as good as in patients with primary cancers. The burden of interventional investigations and complication rates are also obviously high in these patients. The prognosis of synchronous tumors is significantly lower when compared to malignancies of a metachronous nature, despite some encouraging individual results. Only the early implementation of aggressive treatment methods for second primaries is successful in terms of survival [13].

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The active surveillance (AS) of low-risk papillary thyroid microcarcinoma (PMC) was adopted as a management modality for PMC in the guidelines issued by the Japan Association of Endocrine Surgeons (JAES)/Japanese Society of Thyroid Surgery (JSTS) in 2011 ^[15] and those of the American Thyroid Association (ATA) in 2015 ^[16].

The AS strategy was first proposed by Dr. Akira Miyauchi at a physicians' meeting of Kuma Hospital as an alternative to the tradition of performing immediate surgery for all PMCs, which in most cases are extremely indolent ^[17].

Even at Kuma Hospital, where AS was proposed and initiated, the implementation of AS required nearly 20 years, and there were considerable differences in the acceptance of AS among physicians. However, as evidence of the safety and superiority of AS over immediate surgery continues to accumulate, it is expected that the acceptance of this management option will occur more quickly and smoothly in other countries, thereby avoiding unnecessary surgery for this mostly indolent disease ^[17].

IV. CONCLUSIONS

Two primaries in head and neck region are rare, proper work up and high level of suspicion of such patient for double malignancies are highly important, Careful follow up with special emphasis on early detection of any subsequent primary tumors is very critical in patients presenting with head and neck cancers. Role of panendoscopy and PET is controversial but can be of substantial help in this regard.

V. CONSENT

Written informed consent was obtained from the patient for the photography and publication of this case report and any accompanying images.

VI. COMPETING INTERESTS

The authors declare that they have no competing interests.

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