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CASE REPORT

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Primary squamous cell carcinoma of thyroid: a case report and review of literature

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Abstract

Background: Thyroid gland lacks squamous epithelium (except in some rare situations like embryonic remnants or in inflammatory processes); for that reason the primary squamous cell carcinoma (SCC) of thyroid is extremely rare entity, seen only in less than 1% of all thyroid malignancies and is considered almost fatal. So, far, only few case reports have been published in literature.

Case presentation: Herein we present a 54 years old Saudi female with 3 months history of progressive neck swelling and hoarse voice, who was referred to us by her primary care physician as suspected case of anaplastic carcinoma of thyroid for radical external beam radiation therapy (EBRT). Fine Needle aspiration cytology (FNAC) revealed squamous cell carcinoma. Computed tomography (CT) neck showed 10 × 10 cm mass in left lobe of thyroid invading trachea and skin. Extensive staging work up ruled out the possibility of any primary site of SCC other than thyroid gland. Tumor was found unresectable and was referred to radiation oncology. She received palliative EBRT 30 Gy in 10 fractions. After completion of EBRT, there was progression of disease and patient died 3 months after completion of EBRT by airway compromise.

Conclusion: Primary SCC of thyroid is rare and aggressive entity. FNAC is reliable and effective tool for immediate diagnosis. Surgery is a curative option, but it is not always possible as most of cases present as locally advanced with adjacent organs involvement. EBRT alone was found ineffective. Aggressive combined modality (debulking surgery, radiation and chemotherapy) shall be considered for such cases.

Keywords: Squamous cell carcinoma, Thyroid, Rare, Primary, Fatal

Background

Primary squamous cell carcinoma (SCC) of thyroid is an uncommon malignancy and has poor prognosis [1]. SCC of thyroid constitutes less than 1% of thyroid malignancies and has been found fatal within one year of initial diagnosis [2]. The median age is fifth and sixth decade, but can be seen at any age. Main cause of death in these patients is secondary to respiratory interference by direct invasion or compression of the trachea [3]. When SCC of thyroid is diagnosed, the possibility of the tumor arising from adjacent organs (esophagus, larynx) or representing metastatic disease from primary growth somewhere else (lungs) must be considered before concluding the malignancy as SCC of thyroid.

The etiology of SCC thyroid is uncertain as thyroid gland lacks the squamous epithelium. However three theories have been postulated; first the *embryonic nest theory* suggests that squamous cells are derived from the embryonic remnants such thyroglossal duct, thymic epithelium and ultimobronchial body [4]. Second the *metaplasia theory* suggests that the environmental stimuli (inflammation and Hashimoto's thyroiditis) result in squamous metaplasia [5]. Third the *de-differentiation theory* suggests that existing papillary, follicular, medullary and anaplastic thyroid carcinoma de-differentiate into SCC [6,7].

Herein we present a case of 54 years old Saudi lady with locally advanced primary squamous cell carcinoma of thyroid, diagnosed by fine needle aspiration cytology (FNAC) was treated with radiation therapy.

Case presentation

A 54 year old Saudi female presented in our clinic with neck swelling and hoarse voice. She had noticed this

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swelling for 3 months and it had been rapidly increasing in size over a week causing dyspnoea and dysphagia to solids. Her previous medical history revealed type II diabetes mellitus since last 10 years and hypothyroidism since last 3 years, for that she was taking thyroxin 50 micrograms daily and metformin. She had no history of smoking and her weight was stable.

On physical examination, her vitals were stable. A fixed hard neck mass of size 8 × 8 cm was palpable in the left thyroid lobe with inflammatory surface Figure 1. There was no palpable cervical lymphadenopathy and examination of chest, heart, nervous system and abdomen was normal. Clinical differential diagnosis was anaplastic carcinoma of thyroid.

Ultrasonography showed huge left thyroid lobe partially cystic and solid mass of size 8.5 × 9 cm. Computed tomography (CT) neck showed 10 × 10 cm mass in left lobe of thyroid, partially necrotic invading to adjacent skin and trachea and no lymphadenopathy was found Figure 2. Serum T4, thyroid stimulating hormone (TSH), thyroglobulin and serum calcium were within normal limits. Fine needle aspiration cytology (FNAC) of mass was performed, which revealed squamous cell carcinoma Figure 3. Differential diagnosis was metastatic



Figure 1 A fixed hard neck mass of size 8 × 8 cm was palpable in the left thyroid lobe with inflammatory surface.

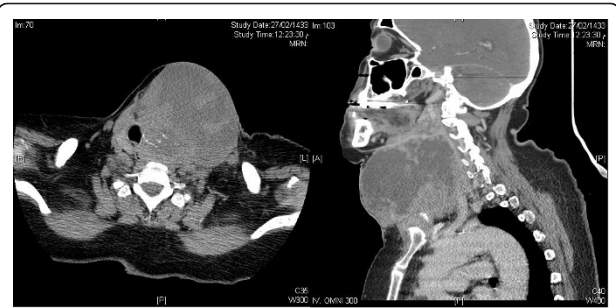


Figure 2 Computed tomography (CT) neck showing 10 × 10 cm mass in left lobe of thyroid, partially necrotic invading to adjacent skin and trachea and no cervical lymphadenopathy.

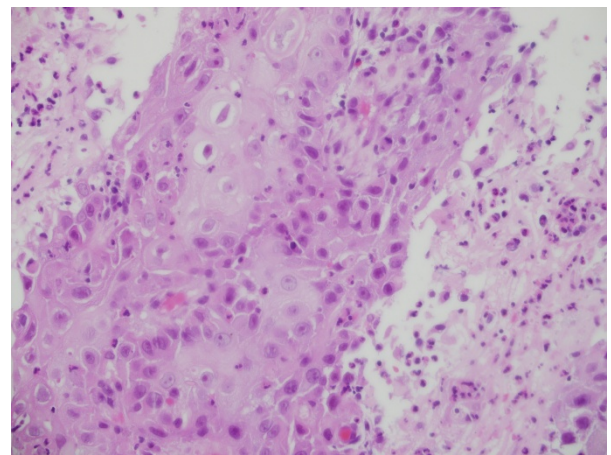


Figure 3 Fine needle aspiration cytology (FNAC) showing nests of pleomorphic cells with abundant eosinophilic cytoplasm and keratin formation along with intercellular bridging.

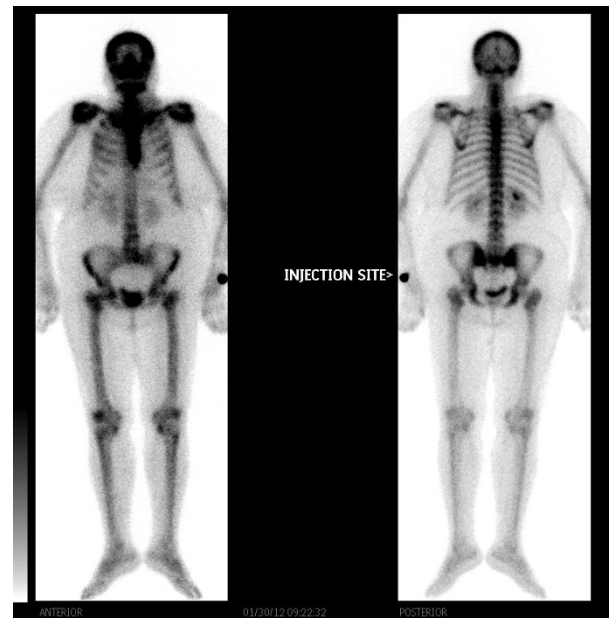


Figure 4 Bone scintigraphy showing no evidence of distant bone metastasis.

Table 1 Previously published case reports (2000-2012) of primary squamous cell carcinoma of thyroid

Author [Ref]	Gender/Age	Presentation	Stage	Associated Problem	Treatment given	Survival	Comments
Zimmer PW [1]	Female/64 years	Asymptomatic neck mass	T2N0M0	-	Total thyroidectomy	7 months	-
Kebapci N [7]	Female/25 years	Right neck mass	T4N1M0	Hashimotos' thyroiditis			
	Papillary carcinoma	Total thyroidectomy and RAI therapy	44 months	-			
Ko YS [8]	Male/87 years	Asymptomatic neck mass	T4N0M0	-	Right lobectomy	NA	CK5/6 + CK19 + EMA,p53 focal + BRAF mutation +
Mercante G [9]	Male/67 years	-	T2N0M0	Follicular carcinoma	Lobe-isthmusectomy + Adjuvant chemoradiation	2 years	-
De Vos FY [10]	-	Neck mass	T4N0M0	-	Induction chemotherapy (Cisplatin + paclitaxel)		
	Total thyroidectomy	20 months	Induction chemotherapy resulted in R0 resection				
Yucel H [11]	Male/88 years	Neck mass	T4N0M0	Hyperthyroidism	Total thyroidectomy Adjuvant radiation therapy	6 months	Patient RAI therapy 20 years back
Eorn TI [12]	Female/43 years	Neck mass	T3N0M0	Papillary carcinoma	Total thyroidectomy Adjuvant radiation therapy 59.4 Gy And RAI	8 months	CK7 + p 63 +
Makay O [13]	Male/53 years Male/71 years	Neck mass, hoarse voice and weight loss	T3N0M0	-	Near total thyroidectomy Chemoradiation 50 Gy + Doxorubicin and cyclophosphamide	2 months 4 months 5 months	-
Fassan M [17]	Female/64 years	Neck mass	T3N0M0	Goiter	Total thyroidectomy	NA	CK 5/6 + CK 7 + CK 19 +
Maamouri F [18]	Female/87 years	Right neck mass	T3N0M0	Papillary carcinoma	Total thyroidectomy And RAI therapy	6 months	-
Chintamani [14]	Female/50 years Male/60 years Male/58 years	Dysphagia, hoarse voice and stridor	T4N0M0	Hyperthyroidism	Total thyroidectomy Adjuvant radiotherapy 50 Gy	12 months	-
Jung TS [15]	Male/56 years	Neck mass, hoarse voice	T3N0M0	Follicular carcinoma	Total thyroidectomy Adjuvant radiotherapy 50 Gy	8 years	-
Sutak J [19]	Female/80 years	Asymptomatic neck mass	T4N1M0	Tall cell variant papillary carcinoma	Total thyroidectomy	-	CK 7 + CK 19 + CK AE1/3 + P53 focal +
Zhou XH [16]	4 patients	NA	T4N0M0	-	Total thyroidectomy Adjuvant radiotherapy 50 Gy + chemotherapy	4 months 6 months 13 months 26 months	Longer survival was seen in combined trimodality treatment
Lam KY [20]	4 females/71 years	Neck mass, stridor	T4N0M0	-	Total thyroidectomy	4 months	CK 7 + CK 19 + CK AE1/3 + P53 focal +

Table 1 Previously published case reports (2000-2012) of primary squamous cell carcinoma of thyroid (Continued)

Kleer CG [21]	7 females/1 male 31-90 years	Neck mass	T4N0M0	Tall cell variant papillary carcinoma	Total thyroidectomy	6 Months-48 months	-
Jones JM [22]	Male/48 years	Hoarse voice, left neck mass	T4N1M0	-	Total thyroidectomy and LND	8 months	-

squamous cell carcinoma from another primary location. CT chest, abdomen, pelvis, magnetic resonance imaging (MRI) of head and neck region, pan-endoscopy, laryngoscopy, esophagoscopy and bone scintigraphy did not reveal any primary lesion or other metastatic disease Figure 4. Radiological stage was made as T4N0M0.

In a multidisciplinary tumor (MDT) meeting it was labeled unresectable and patient was referred for external beam radiation therapy (EBRT) after prophylactic percutaneous endoscopic gastrostomy (PEG) insertion. Due to retrosternal extension of disease, tracheostomy was deferred. Patient received 30 Grays (Gy) in 10 fractions to thyroid. Post radiation therapy, there was progression size of neck mass with progressive dyspnoea. Patient died of airway compromise 3 months of palliative EBRT.

Discussion

Primary SCC of the thyroid gland is an extremely rare and aggressive entity usually presents with classic triad features; (I) rapidly enlarging mass in the older patients behaving like anaplastic carcinoma, (II) it may be associated with other thyroid malignancies and (III) histological features of intercellular bridges and keratin [8-10]. FNAC is reliable and confirmatory tool, but it is mandatory to exclude the metastatic SCC [11].

Treatment with surgery, radiation therapy and chemotherapy alone has been found ineffective in previously published similar case reports, as majority of these patients present as locally advanced cases not amenable for curative resection Table 1. The better survival rates have been achieved with aggressive combination therapy (surgery followed by adjuvant radiation therapy (50-60 Gy) with or without chemotherapy or induction chemotherapy followed by surgery) [11-16].

Conclusion

Primary squamous cell carcinoma of thyroid is a rare and aggressive entity with poor prognosis. FNAC is effective confirmatory tool, but efforts shall be made to rule out metastatic SCC originating from other sites. Surgery, radiotherapy and chemotherapy alone are ineffective. Aggressive treatment with surgery followed by adjuvant radiotherapy with or without chemotherapy is recommended to achieve better outcome.

Consent

Written permission was taken from the patient for publication of the case report.

Abbreviations

SCC: Squamous cell carcinoma; EBRT: External beam radiation therapy; FNAC: Fine needle aspiration cytology; CT: Computed tomography; RAI: Radioactive iodine; TSH: Thyroid stimulating hormone; MDT: Multidisciplinary tumor meeting; PEG: Percutaneous endoscopic gastrostomy.

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Authors' contributions

MAT, MAA Manuscript preparation. RA Data Collection. MF Pathological data. All authors read and approved the final manuscript.

Competing interests

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