

Recurrence of Papillary Thyroid Carcinoma Associated with Mucoepidermoid Carcinoma: An Uncommon Case Report

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is most usually found in the salivary glands, but it has also been reported in the lung, esophagus, breast, pancreas, and thyroid. MECs are considered to be low-grade carcinomas, and their occurrence in the thyroid is extremely rare. A case report of a patient with the dual pathology of a composite mucoepidermoid carcinoma of the thyroid and a papillary thyroid carcinoma is presented in this study. A 38-year-old female patient was referred to NINMAS for a ¹³¹I whole body iodine scan (DxWBS) with the complaint of big swelling on the right side of the neck and a discharging sinus. She was diagnosed with papillary thyroid carcinoma (PTC) in 2001 after a total thyroidectomy. Comprehensive neck dissection on the right side with left-sided selective neck dissection was done in 2020. Histopathology revealed metastatic papillary thyroid carcinoma on the left side and mucoepidermoid carcinoma on the right side. The patient did not take any RAIT. After a short period of time, the patient started developing a huge neck mass with a discharging sinus. An ultrasonogram of the mass showed a large, heterogeneous mass with microcalcification on the right side of the neck. Biochemical reports revealed a serum thyroglobulin (Tg) level of >300 ng/mL. DxWBS showed a big area of iodine concentration on the right side of the neck. Excision of mass with a skin flap was done in 2022. She received 20cGy fractions of radiotherapy at the right neck. The patient received ¹³¹I radioactive iodine therapy at 150 mCi in 2023. The patient is now very ill, with weakness of both lower limbs, blurring of vision, and a diagnosed brain metastasis as well.

Keywords: Papillary thyroid carcinoma, mucoepidermoid carcinoma, whole body scan.

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INTRODUCTION

Mucoepidermoid carcinomas (MEC) are malignant glandular epithelial neoplasms recognized by mucous, intermediate, and epidermoid cells, along with columnar, clear cell, or

oncocytoid features. The median age of MEC in adults is 49 years, and in children it is 13 years. Females have a slight predominance of MEC (1). It accounts for approximately 35% of all malignancies of the major and minor salivary glands, especially in the parotid gland (2). But it has also been reported in the lung, esophagus, breast, pancreas, and thyroid. MECs are considered to be low-grade carcinomas, and their occurrence in the thyroid is extremely rare.

MEC in the thyroid gland is postulated to originate from the remnants of the ultimobranchial body, intrathyroidal remnants of the salivary glands, the follicular epithelium, C cells, parathyroid, and thyroglossal duct. MEC is known to present with other variants of thyroid cancer, such as papillary thyroid carcinoma (3). This case reported a dual pathology of a composite mucoepidermoid carcinoma of the thyroid and a papillary thyroid carcinoma with lymph node metastasis.

CASE REPORT

A 38-year-old female patient was referred to NINMAS for a whole-body iodine scan (DxWBS) with the complaint of a big swelling on the right side of the neck and a discharging sinus. She was diagnosed with papillary thyroid carcinoma after a total thyroidectomy in 2001. Many years later, she again complained of right-sided neck swelling. In 2020, she underwent right-sided comprehensive neck dissection with left-sided selective neck dissection (II to IV). Histopathology revealed mucoepidermoid carcinoma on levels III and IV and metastatic papillary thyroid carcinoma on levels III and IV. The patient did not take any radioactive

iodine therapy (RAIT). After a short period of time, the patient developed a huge neck mass on the right side with a discharging sinus. Ultrasonography of the neck showed a large, heterogeneous mass with microcalcification on the right side of the neck, measuring about 4.3 x 3.2 cm. Multiple enlarged lymph nodes on both sides of the neck at levels II to IV with altered echotexture were also found, the largest one measuring about 6.7 x 5.4 cm on the right side and 8.3 x 5.5 cm on the left side. A CT scan revealed a large lobulated soft tissue mass at the right side of the neck, extending from the submandibular region to the right supraclavicular region, with involvement of the right sternocleidomastoid muscle and partial encasement of the right carotid vessel. Bilaterally marked cervical, supraclavicular, and suprasternal lymphadenopathy (right>left) was also noted. The upper chest showed multiple pleural-based and intra-parenchymal pulmonary nodules in both lungs. Biochemical reports revealed a serum thyroglobulin level of >300 ng/mL. DxWBS showed a big

area of iodine concentration on the right side of the neck. The patient underwent excision of the huge neck mass with reconstruction by a platysma mucocutaneous flap in October 2022. HPR revealed invasive squamous cell carcinoma, grade II. An immunohistochemistry report revealed mucoepidermoid carcinoma. She received 20cGy fractions of radiotherapy at the right neck. Two months later, the patient got radioactive iodine therapy at 150 mCi in 2023. At that time, her S. thyroglobulin level was 5.09 ng/mL. One month later, the patient developed multiple symptoms like weakness of both lower limbs, blurring of vision, and headaches. An MRI of the brain revealed multiple rim-enhancing areas with edema and a mass effect, suggesting brain metastases. Now the patient is receiving radiotherapy for the brain. An ultrasonogram of the neck again revealed an enlarged lymph node on the right side of the neck at level III, measuring about 2.5 X 1.5 cm, with a loss of architecture. The patient's condition is deteriorating day by day.



Figure 1: a) Huge neck mass with discharging sinuses before surgery and b) After excision of neck mass and reconstruction by platysma myocutaneous flap



Figure 2: High resolution ultrasound image of neck showing a big heterogenous solid mass with microcalcifications on the right side of the neck

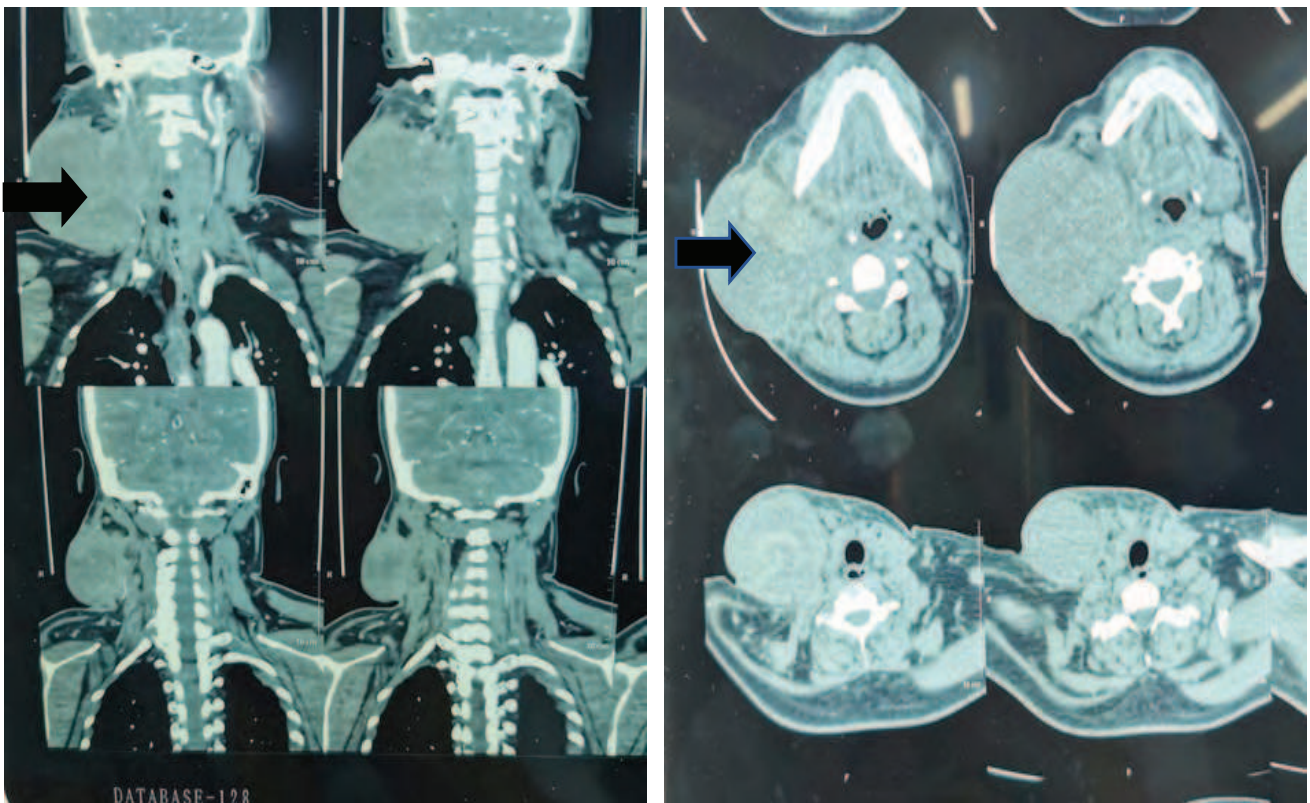


Figure 3: CT scan image showing a large lobulated soft tissue mass at the right side of neck extending from the submandibular region to the right supraclavicular region

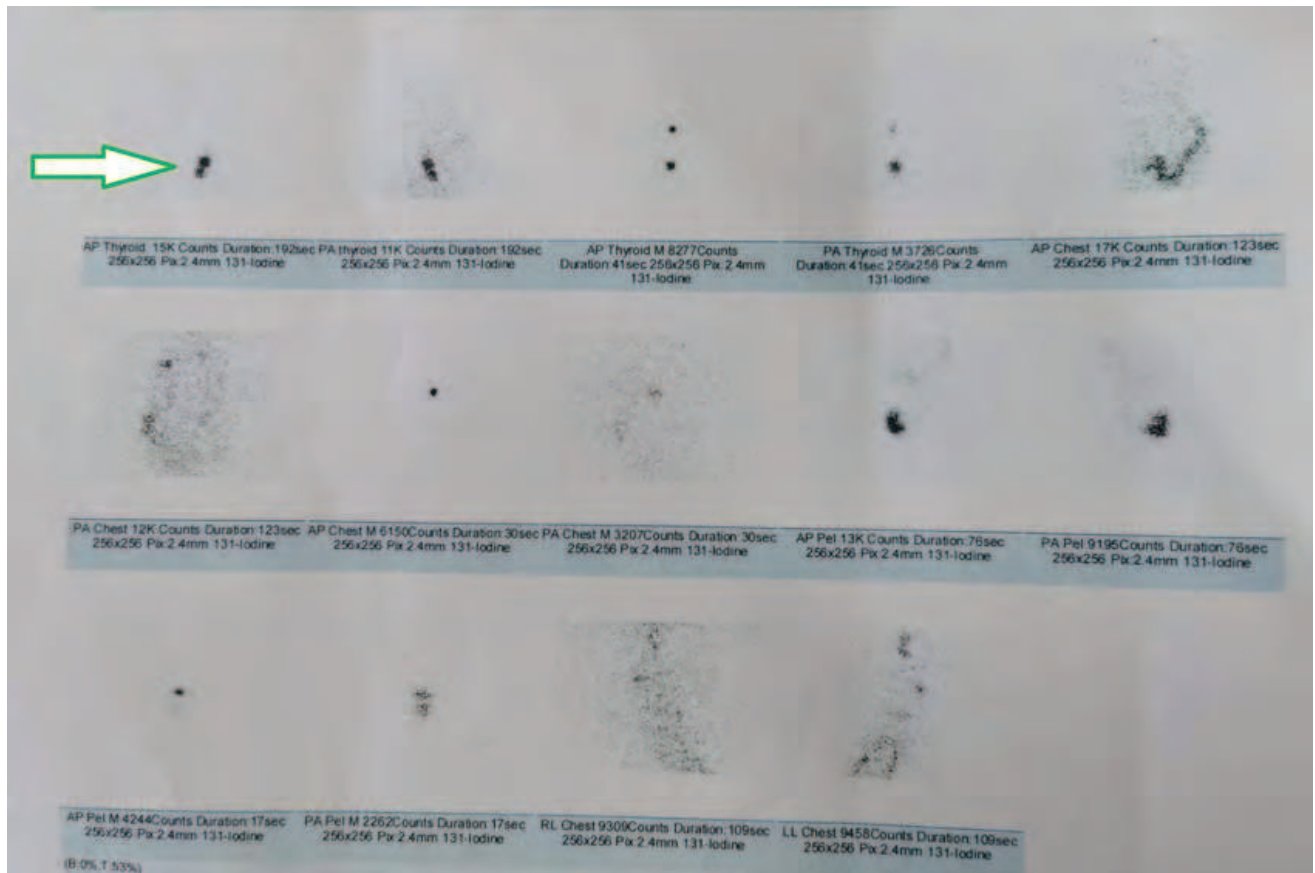


Figure 4: Image of large dose ¹³¹I scan showing a big area of radio-iodine concentration on the neck.

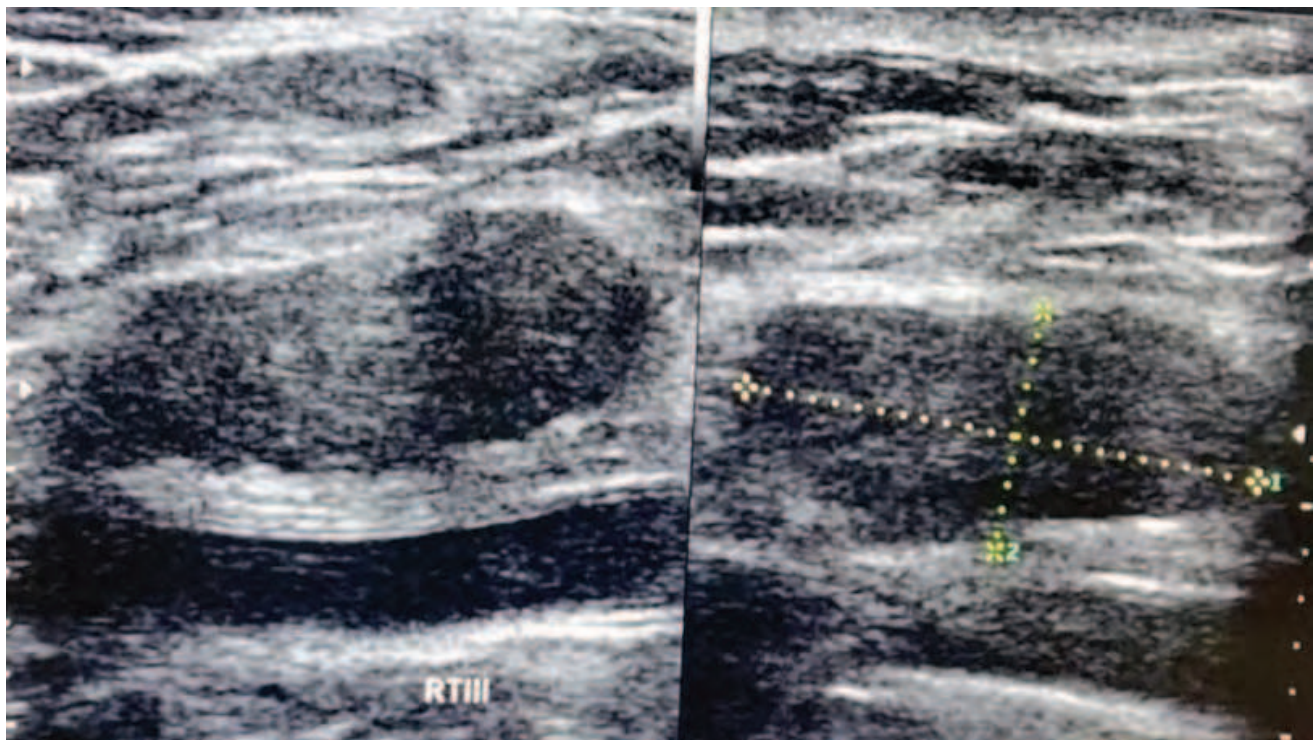


Figure 5: High resolution ultrasound image of neck revealed enlarged level III lymph node on right with loss of architecture after short interval of RAIT

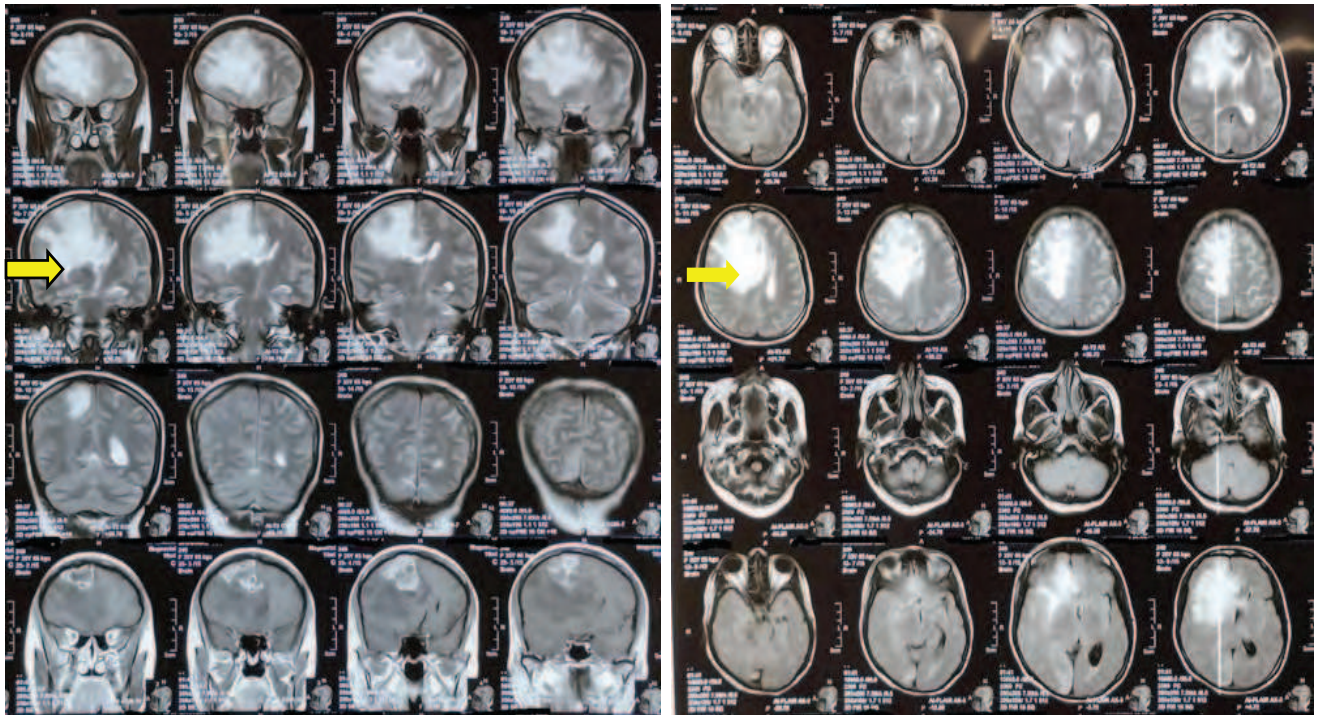


Figure 6: MRI of Brain showing multiple rim enhancing areas with edema and mass effect- suggestive of metastasis after short interval of RAIT.

DISCUSSION

Papillary thyroid carcinoma (PTC) is the commonest form of well-differentiated thyroid cancer and the commonest form of thyroid cancer to result from exposure to radiation (4). In the thyroid, MECs are quite rare malignant neoplasms, accounting for 0.5% of thyroid malignancies. This patient was first diagnosed with primary papillary thyroid carcinoma. She had no family history of thyroid carcinoma or exposure to radiation. Some publications suggest that MECs can arise from metaplastic dedifferentiation of papillary thyroid carcinoma, follicular thyroid carcinoma, or oncocytic carcinoma (5, 6). Primary thyroid MEC is frequently found in PCT (7). In this case, it may be associated with the dedifferentiation of papillary thyroid carcinoma.

Primary mucoepidermoid tumors are described as having a slow prognosis. Most doctors acknowledge MEC of the thyroid as a low-grade malignant tumor. In the past, primary mucoepidermoid tumors of the thyroid have been described as having indolent behavior, although those tumors that have been locally invasive or those that have presented with concurrent papillary and mucoepidermoid components have

been noted to be more aggressive. (8). The incidence rate of extrathyroidal extension is 14.8%, and approximately 42.2% of patients have nodal metastases. Patients with thyroid MEC often present with a painless and usually unilateral mass in the thyroid with reduced uptake in thyroid scans. In this case, the patient had a rapidly growing, painless neck mass. On DxWBS, a large area of radiotracer concentration was seen in the right thyroid bed before the operation. In this case, both extrathyroidal extension and nodal metastases were seen. Iodine avidity was not found in the pulmonary area.

The prognosis for thyroid MEC is good, with several cases of disease-free survival of more than 10 years reported (9). However, the majority of cases in the published literature describe patients who were diagnosed with cervical lymph node metastases or local invasion by locally advanced tumors. Treatment involves surgical excision with a complete thyroidectomy. The prophylactic central neck node dissection for patients with MEC is controversial. External beam radiotherapy and chemotherapy have been used to treat MEC; however, there is still debate about adjuvant therapies (10).

There have only been 46 documented cases of mucoepidermoid carcinoma, an uncommon thyroid cancer. Although MEC of the thyroid is usually considered a low-risk malignant tumor, several cases with adverse outcomes have been reported. The tumor had poor prognostic features at diagnosis and was treated with palliative thinking.

In MEC with only locoregional spread, treatment with total thyroidectomy, neck dissection, and radiation has shown good clinical outcomes. MEC could develop from the de-differentiation of pre-existing WDTC (papillary and follicular thyroid cancer), with a high likelihood of nodal metastasis (11). But direct invasion into deeper structures is a poor prognostic factor, and in such cases, treatment with surgery and radiotherapy has been unsuccessful in halting the advancement of the disease. In this case, the patient developed brain metastases within a short period of time. The patient is now hospitalized due to brain metastases and is receiving radiotherapy for palliative purposes.

CONCLUSION

In the reported case, a very rare dual pathology of thyroid coexisting initially patient with papillary thyroid carcinoma after 20 years patient develop huge neck mass diagnosed as mucoepidermoid carcinoma on IHC. The multidisciplinary strategy was effective in treating the patient's; nevertheless, because of the disease's aggressiveness, the patient quickly developed brain metastases and lymph node recurrence.

REFERENCES

- Asiry S, Rao RA. Mucoepidermoid carcinoma. PathologyOutlines.com website. <https://www.pathologyoutlines.com/topic/salivaryglandsMEC.html>.
- Mesolella M, Iengo M, Testa D, DI Lullo AM, Salzano G, Salzano FA. Mucoepidermoid carcinoma of the base of tongue. *Acta Otorhinolaryngol Ital.* 2015 Feb;35(1):58-61. PMID: 26015654; PMCID: PMC4443580.
- Nimeshi S Jayakody, MoradFaoury, Lisa R Fraser, Sanjay Jogai, Nimesh N Patel, Composite poorly differentiated mucoepidermoid carcinoma of the thyroid and follicular variant of papillary thyroid carcinoma. Report of a case and review of the literature, *Journal of Surgical Case Reports*, Volume 2019, Issue 4, April 2019, rjz092, <https://doi.org/10.1093/jscr/rjz092>
- Ponnandai S Somasundar, MD, MPH, FACS; Chief Editor: Neerav Goyal, MD, MPH, FACS. <https://emedicine.medscape.com/article/282276>
- Yeungnam Univ J Med 2021; 38(2):169-174. Published online: 5 March 2021 DOI: <https://doi.org/10.12701/yujm.2021.00941>
- Ruth S. Prichard, James C. Lee, Anthony J. Gill, Mark S. Sywak, Larry Fingleton, Bruce G. Robinson, Stanley B. Sidhu, and Leigh W. Delbridge. Mucoepidermoid Carcinoma of the Thyroid: A Report of Three Cases and Postulated Histogenesis. *Thyroid*. Feb 2012.205-209. <http://doi.org/10.1089/thy.2011.0276>
- Abir Al Ghuzlan, Service de pathologiemorphologique, Gustave Roussy, 114 Rue Edouard Vaillant, 94805 Villejuif, France. Email: abir.alghuzlan@gustaveroussy.fr DOI: 10.1177/1066896915626281
- Jung YH, Kang MS. Composite follicular variant of papillary carcinoma and mucoepidermoid carcinoma of thyroid gland: a case report. *J Korean Med Sci* 2010;25:1683-7
- Wenig BM, Adair CF, Heffess CS. Primary mucoepidermoid carcinoma of the thyroid gland: a report of six cases and a review of the literature of a follicular epithelial-derived tumor. *Hum Pathol.* 1995;26:1099-1108.
- Le QV, Ngo DQ, Ngo QX. Primary Mucoepidermoid Carcinoma of the Thyroid: A Report of a Rare Case with Bone Metastasis and Review of the Literature. *Case Rep Oncol.* 2019 Mar 19; 12(1):248-259. doi: 10.1159/000498917. PMID: 31011324; PMCID: PMC6465688.
- Prichard RS, Lee JC, Gill AJ, Sywak MS, Fingleton L, Robinson BG, et al. Mucoepidermoid carcinoma of the thyroid: a report of three cases and postulated histogenesis. *Thyroid.* 2012 Feb;22(2):205-9