CASE REPORT

Unusual Presentation of Thyroid Storm in a Patient with Metastatic Follicular Carcinoma of Thyroid

Ashwinee Rahalkar¹⁰, Kul Ranjan Singh², Pooja Ramakant³, Anand Mishra⁴

Received on: 16 November 2022; Accepted on: 16 November 2022; Published on: 30 December 2022

ABSTRACT

Thyroid storm (TS) is a rare, acute life-threatening complication with a high mortality rate. We present a case report of the unusual presentation of TS in a patient with metastatic follicular thyroid carcinoma with poorly controlled diabetes mellitus.

Keywords: Diabetes mellitus, Metastatic follicular thyroid carcinoma, Thyroidectomy, Thyroid storm.

Indian Journal of Endocrine Surgery and Research (2022): 10.5005/jp-journals-10088-11201

HISTORY AND EXAMINATION

A 53-year-old lady hailing from an area endemic to goiter, presented with non-traumatic sudden onset paraplegia of 3 months duration. She had a gradually progressive anterior aspect neck swelling for the last 30 years which had no compressive features or features suggestive of hypo- or hyperthyroidism. She had poorly controlled type II diabetes mellitus for the last 10 years and bladder and bowel incontinence for the last 10 days.

Examination revealed a $6 \text{ cm} \times 5 \text{ cm}$ hard thyroid swelling with restricted mobility and an impalpable lower border.

Evaluation revealed subclinical hyperthyroidism with thyroid stimulating hormone (TSH) <0.01 ulU/mL and normal fT4 and total T3 levels. Her deranged blood sugars were optimized with insulin and hypercalcemia (total calcium level of 11.8 mg/dL and ionic calcium of 6.9 mg/day) controlled with saline diuresis and intravenous zoledronic acid infusion.

Preoperative two-dimensional (2D) echocardiogram was normal with a left ventricular ejection fraction of 62%.

High-resolution ultrasound of the neck showed a partially calcified nodule of 61 mm × 35 mm involving isthmus and left lobe – Thyroid imaging reporting and data system (TI-RADS) V lesion.

Contrast-enhanced computed tomography of the neck and thorax revealed multiple heterogeneously enhancing thyroid nodules with retrosternal extension (Fig. 1) and displacement of the trachea and its compression. The interface of the thyroid and strap muscles was ill-defined (Fig. 2). Thyroid gland was abutting more than 90% circumference of the left brachiocephalic vein and focally abutting the left common carotid artery. Few subcentimetric level IB, II lymph nodes were present. Multiple lung lesions suggestive of metastasis were obvious in the lung field (Fig. 3). Osteolytic lesions were seen in the bilateral proximal humerus, D6, D7, and D8 vertebra. Fine needle aspiration cytology (FNAC) was consistent with Bethesda V.

After optimization of blood sugar levels and correction of hypercalcemia, the patient was planned for total thyroidectomy followed by radioiodine ablation.

INTRAOPERATIVE FINDINGS

The patient underwent an uneventful total thyroidectomy. Inner strap muscles were adherent to the tumor and excised *en bloc* with the tumor.

¹⁻⁴Department of Endocrine Surgery, King George's Medical University, Lucknow, Uttar Pradesh, India

Corresponding Author: Pooja Ramakant, Department of Endocrine Surgery, King George's Medical University, Lucknow, Uttar Pradesh, India, Phone: +91 9791507780, e-mail: poojaramakant@gmail.com

How to cite this article: Rahalkar A, Singh KR, Ramakant P, et al. Unusual Presentation of Thyroid Storm in a Patient with Metastatic Follicular Carcinoma of Thyroid. Indian J Endoc Surg Res 2022;17(2):69–71.

Source of support: Nil

Conflict of interest: Dr. Pooja Ramakant is associated as Editorin-Chief of this journal and this manuscript was subjected to this journal's standard review procedures, with this peer review handled independently of the Editor-in-Chief and his research group.

There was a tumor thrombus extending in both internal jugular veins (Fig. 4). Thrombectomy was done for the removal of tumor thrombus. Tumor thrombus to the left brachiocephalic vein was also present (Fig. 5), which was extending retrosternally. In view of the poor performance status of the patient, a sternotomy was not done and a thrombectomy of brachiocephalic was deferred. Bilateral recurrent laryngeal nerves were identified and preserved.



Fig. 1: CT scan showing retrosternal extension

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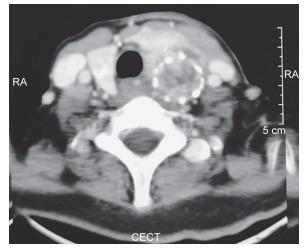


Fig. 2: CT scan showing ill-defined planes with strap muscles

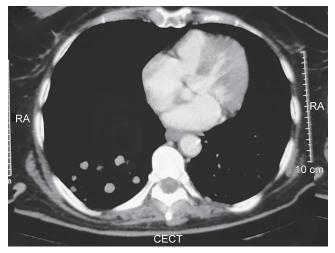


Fig. 3: CT scan showing multiple lung metastasis



Fig 4: Intraoperative image showing extension of tumor thrombus in internal jugular vein

POSTOPERATIVE PERIOD

70

The patient was stable in the immediate postoperative period with no signs and symptoms of hypocalcemia. She was ambulant and



Fig. 5: Intraoperative image showing tumor thrombus in left brachiocephalic vein

accepting oral feeds. Two units of packed red blood cells (PRBC) were transfused.

On postoperative day 3, the patient developed palpitations with a pulse rate of 150 beats per minute, regular in rhythm. Saturation was 90% on room air which improved to 97% on 4 L of oxygen through nasal prongs. She was irritable with a Glasgow coma scale of 13/15. The electrocardiogram showed no ST-T wave changes. Raised lactate and hypocalcemia were evident in arterial blood gas analysis. After a loading dose of injection hydrocortisone 100 mg IV, tablet aspirin, tablet clopidogrel, and tablet atrovastatin she was started on esmolol infusion and calcium gluconate infusion. Although heart rate (HR) reduced she has persistent tachycardia in the range of 125.

The ProBNP level was 11,362 (normal range: 0–125) and Trop T was 0.092 (normal range: <0.014) and D-dimer levels were in the normal range. She remained afebrile and a repeat thyroid profile suggested S.TSH <0.01, with increased free T4 level, 22.75 (normal range: 4.87–11.72 μ g/dL), and T3 level: 3.56 (normal range: 0.58–1.59 ng/dL).

She was immediately started on tablet neomercazole 30 mg, tablet propranolol LA 40 mg, and injection dexamethasone 8 mg and continued on maintenance dose. Injection enoxaparin was given subcutaneously for suspected pulmonary embolism.

A repeat electrocardiogram and echocardiogram had a loss of T wave, left ventricular ejection fraction of 45–50%, and left anterior descending artery territory hypokinesia.

Despite all resuscitative efforts and best care, the patient succumbed on postoperative day 5.

DISCUSSION

Thyroid storm, also known as thyrotoxic crisis, is today a rare, acute life-threatening complication of hyperthyroidism with multi-system involvement. Usually occurs after some precipitating factor like infection, thyroid surgery, non-thyroid surgery, administration of iodinated contrast medium, or abrupt withdrawal of antithyroid medications. Also, TS has an extremely high mortality rate 8–25% despite the best resuscitative and support.¹

The common presenting features of TS are fever, cardiovascular manifestations (tachycardia, arrhythmia, and heart failure), central nervous system (agitation, delirium, anxiety, psychosis, and coma),



gastrointestinal (nausea, vomiting, diarrhea, abdominal pain, intestinal obstruction, and acute hepatic failure).

Normal thyroid function is essential for maintaining equilibrium in glucose metabolism and hyperthyroidism makes glycemic control challenging in diabetics.² Thyroid hormone excess increases intestinal glucose absorption, hepatic production of glucose from glycogen, and decreases insulin secretion from the pancreas along with increased insulin resistance and renal clearance. Untreated thyrotoxicosis can also predispose a patient with type 2 diabetes mellitus to DKA by affecting adipokines and altering carbohydrate metabolism.^{3,4} The patient was diabetic with poor glycemic control and had an unusual presentation of TS.

High-grade fever with diaphoresis is the key presenting feature in most of the patients. However, elderly persons like ours may remain afebrile. A Japanese study found that central nervous system involvement was a poor prognostic factor and was associated with increased mortality.⁵ In our case also, fever was absent and the patient had central nervous system involvement in the form of agitation.

There are very few case reports where a thyroid crisis has occurred secondary to thyroid cancer with metastases.^{6–10} In our case we are not sure about the trigger factor for thyroid crisis, could be secondary to metastatic disease, surgical stress, or iodinated contrast medium causing this TS.

CONCLUSION

Early diagnosis and timely management of TS are extremely important. A high index of suspicion is required for diagnosing TS, especially in diabetic patients with thyroid cancer and functional distant metastases.

ORCID

Ashwinee Rahalkar // https://orcid.org/0000-0002-1737-0118

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