Thyroid Plasmacytoma: A Rare Cause of Hoarseness of Voice

Sir,

We read with great pleasure the recent article by Kashyap *et al*. published in July–September issue of your esteemed journal.[1] We hereby take this opportunity to share our experience with a similar case of thyroid plasmacytoma. With the aid of the present case and our previously reported cases of plasmacytomas involving rare sites such as frontal bone, duodenum, pancreatic head, and ovary, we aim to enrich the knowledge of our readers about the unusual presentations and rare locations of extramedullary plasmacytomas (EMPs).[2,3]

A 57-year-old euthyroid male presented for hoarseness of voice of three months duration. There was no history of associated stridor, respiratory distress, and hemoptysis. He denied loss of appetite, loss of weight, past ionizing radiation, or family history of thyroid malignancy. Examination revealed a swelling of approximately 5 cm × 5 cm in dimension, predominantly involving the right anterior half of the neck. Palpation confirmed a single, firm, nontender swelling moving with deglutition. There was no palpable lymphadenopathy or hepatosplenomegaly. The clinical possibilities of benign (e.g., goiter) as well as malignant disorders (e.g., thyroid, parathyroid carcinoma and lymphoma) were kept. Ultrasonography revealed an ill-defined solid hypoechoic mass (4.3 cm × 3.0 cm × 2.3 cm) arising from the superolateral margin of the thyroid gland and extending laterally outside the thyroid capsule. Thyroid and parathyroid hormone levels were within normal limits. Hemogram, erythrocyte sedimentation rate, renal and liver function tests were also normal. Positron emission tomography/computed tomography (PET/CT) scan confirmed a thyroid mass with moderate fluorodeoxyglucose uptake, with SUV_{max} of 3.5 [Figure 1]. Fine-needle aspiration cytology (FNAC) with cell block from the thyroid mass showed infiltration by plasma cells [Figure 2a-c]. Flowcytometric analysis (FCA) of the aspirate showed bright positivity for CD38 and dim for CD138 with cyto-kappa restriction. Immunochemistry done on cell block was strongly positive for CD38 and CD138 [Figure 2d]. Serum protein electrophoresis (SPEP) showed monoclonal gammopathy (M spike of 0.56) in the gamma globulin region. Bone marrow biopsy and serum beta-2 microglobulin were done and a final diagnosis of multiple myeloma (MM, IgG lambda, ISS-II) with thyroid plasmacytoma was confirmed. He was started on weekly cyclophosphamide (300 mg/m^2), oral dexamethasone (40 mg), and injection bortezomib (1.4 mg/m^2 subcutaneous) regimen. After two courses, a repeat evaluation showed 50% reduction in M band

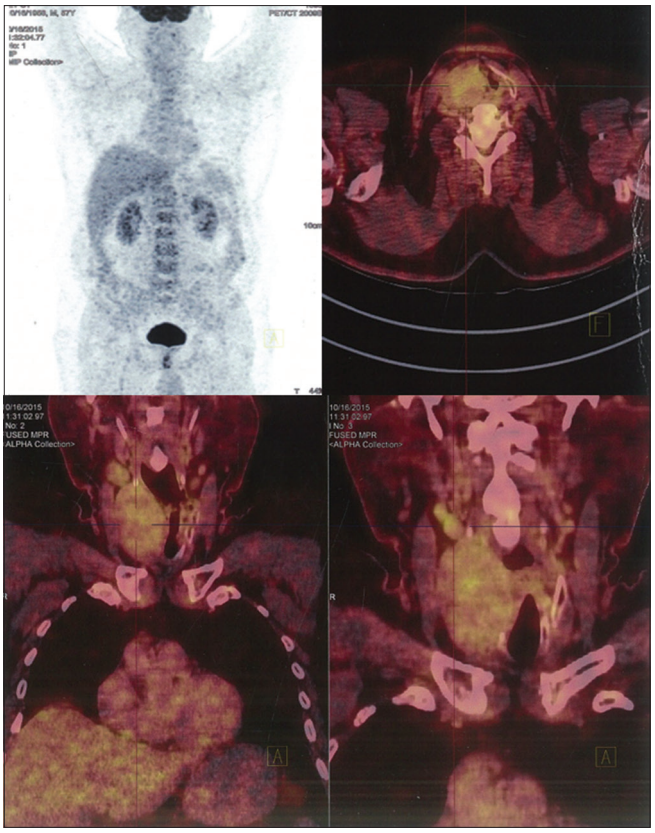

Figure 1: Positron emission tomography/computed tomography scan showing a thyroid mass with fluorodeoxyglucose uptake (SUV_{max}) of 3.5.

levels (from 0.56 to 0.26 g/dl) and neck swelling (both clinically and radiologically). He is planned for autologous hematopoietic stem cell transplantation after achieving disease remission.

The incidence of EM disease in MM patients has significantly increased in recent years owing to more sensitive imaging techniques and prolonged survival of MM patients.[1,4] Extraosseous involvement usually occurs late during the disease course, confers a poor prognosis, and is associated with shorter progression-free and overall survival.[1,4] We have also recently reported EMPs in duodenum, pancreas, and ovary in a single patient.[2] Similar to our case, Vailati *et al.* have also reported plasmacytoma of the thyroid as an initial presentation of MM.[5] Serefhanoglu *et al.* reported an unusual initial presentation of MM involving thyroid gland and pericardium with myelomatous pleural and pericardial effusion.[6]

EMP of the thyroid usually presents as a painless, firm, nontender, mobile, multinodular, or diffuse thyroid mass with no associated cervical lymphadenopathy and patients can be either euthyroid or hypothyroid.[7] In the

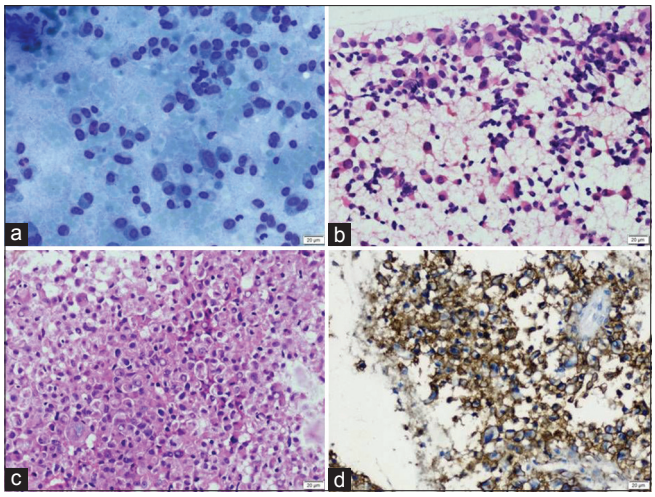

Figure 2: (a) Scattered plasma cells with eccentrically placed nuclei and few showing perinuclear hoff (May–Grünwald, ×100), (b) Scattered plasma cells with few binucleated forms (H and E, ×100), (c) Cell block showing sheets of plasma cells (H and E, ×40), (d): Plasma cells showing CD38 positivity on immunochemistry

present case, the patient was euthyroid and presented with hoarseness of voice. Tandon *et al*. reported a case of plasma cell leukemia in which thyroid involvement was initially masked by Hashimoto's thyroiditis.[8]

The gold standard diagnostic test for thyroid solitary plasmacytoma is histological confirmation. There are very few reports on the diagnosis of these cases by cytology.[9] The cytology smears in the present case had predominant population of plasma cells, most of which were mature along with many binucleate and immature forms [Figure 2b]. These findings are similar to those described by others in the past.[9]

One of the most challenging issues in the diagnosis of solitary thyroid plasmacytoma is to rule out systemic involvement by MM. Normal bone marrow findings, absence of lytic bone lesions, and no or minimal M spike are confirmatory findings seen in solitary thyroid plasmacytoma. Rubin *et al*. reported that 33% of the patients of solitary plasmacytoma of the thyroid can present with monoclonal gammopathy.[10] In the present case, SPEP revealed monoclonal gammopathy, with bone marrow showing ~10% plasma cells, and there were lytic lesions in the frontal bone. Hence, a diagnosis of MM with thyroid plasmacytoma was made. PET/CT in MM also helps to know the disease burden and to know the hidden, asymptomatic EM sites where local radiation therapy could also be beneficial.

Based on the preoperative FNAC findings, a specific diagnosis of thyroid plasmacytomas can be difficult because of its rarity. Moreover, diagnosis can be confusing in many cases because thyroid plasmacytoma may resemble other, more common thyroid lesions, including both benign and malignant neoplasms. Careful cytomorphologic examination and supportive studies are required to ensure the right diagnosis in a suspected case.[10] In the index case, FCA, immunocytochemistry (IHC), and PET/CT validated the diagnosis of MM with thyroid plasmacytoma.

In summary, through this letter, we wish to emphasize that rarely, MM can present with atypical and unusual features, and hence a high degree of suspicion is required to clinch the diagnosis.[11] Utilizing PET/CT studies and other ancillary techniques (FNAC, IHC, SPEP, urine protein electrophoresis, bone marrow biopsy, etc.) results in accurate and rapid diagnosis of thyroid plasmacytomas and other neoplasms.[12,13]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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