Unusual association of primary hyperparathyroidism, papillary thyroid carcinoma, and follicular adenoma in a young female

Sirs,

Nonmedullary thyroid neoplasms are rarely described along with primary hyperparathyroidism (PHPT). Preoperative diagnosis of associated thyroid disease is very important for appropriate surgical management.

A 31-year-old female presented with recurrent renal colic and easy fatigability for 4 years duration. Two years back she underwent cholecystectomy for symptomatic gall stone disease. No past history of fracture, gravelluria, or pancreatitis. On examination, firm, asymmetrical grade 2 goiter (Rt > Lt, 7 × 5 cm). The albumin adjusted serum calcium was 11.7 mg/dL (N, 8.5-10.2 mg/dL) and inorganic phosphate was 2.3 mg/dL (N, 3.5-5.0 mg/dL), alkaline phosphatase was 26 KAU (N, 3-13 KAU), intact PTH of 238 pg/ml (N, 15-65 pg/ml) and 25 hydroxy vitamin D was 5 ng/ml (N, 9-37 ng/ml). Complete blood count, renal function test, and thyroid profiles are within normal limits.

The skeletal survey was noncontributory. Ultrasound neck revealed a hypoechoic mass in the right upper pole of the thyroid gland with enlarged right superior parathyroid gland. Ultrasound of abdomen showed bilateral nephrocalcinosis and nephrolithiasis. 99mTc sestamibi scintigraphy was suggestive of right superior and left inferior parathyroid adenoma. Fine needle aspiration cytology (FNAC) of the thyroid lesion showed lymphocytic thyroiditis. Considering the young age, double parathyroid adenoma and coexisting thyroid enlargement MEN-I work-up was done and all were within normal limits.

Based on the clinical, biochemical, and imaging findings, a diagnosis of PHPT with diffuse goiter was made and the patient was subjected to bilateral neck exploration. She underwent right hemithyroidectomy along with right superior and left inferior parathyroid gland resection. Histopathological findings revealed right superior and left inferior parathyroid adenoma [Figure 1a]. Microscopically the thyroid gland showed papillary carcinoma along with lymphocytic thyroiditis [Figures 1b and c] and follicular adenoma in another focus [Figure 1d]. There was no evidence of lymph nodal metastasis. On the second postoperative day, patient developed features of hypoparathyroidism and managed with calcium infusion (1 mg/kg/h) and oral calcitriol (0.5 µg/day). Subsequently patient underwent completion thyroidectomy and radio iodine ablation (85 mci).

The most common associated thyroid tumor in PHPT includes MTC, follicular adenoma, and rarely papillary thyroid carcinoma. The proposed mechanism of coexisting thyroid abnormalities is radiation therapy of head and neck followed by calcium act as a goitrogen. Sato, et al reported a case with four tumors in the neck. To the best of our knowledge our patient is the second single case who had concurrent four tumors in the neck with no functional symptoms related to the thyroid. FNAC may not pick up all the lesions, particularly in multi-focal involvement, as it happened in our case. Thus surgical treatment is mandatory in such cases with histopathology as the gold standard for diagnosis. This case underscores the need of preoperative FNAC of the thyroid nodule for appropriate surgical intervention and to avoid unexpected surprises postoperatively.

To conclude, preoperative evaluation for hyperparathyroidism should be carefully made in cases with multiple neck swellings and possibilities of concomitant thyroid lesions needs to be considered in rare instances.

Figure 1: (a) Microphotograph of parathyroid gland showing thin capsule with follicular arrangement of cells (H and E, ×180). (b) Microphotograph of thyroid gland showing papillary thyroid carcinoma with follicular arrangement along with lymphocytic thyroiditis (H and E, ×280). (c) High power of the thyroid gland showing nuclear stratification and optically clear nuclei of papillary carcinoma (H and E, ×540). (d) High power of the thyroid gland showing moderately thin capsule with follicular arrangement of cells in follicular adenoma (H and E, ×240)
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REFERENCES

1. Ogburn PL, Black BM. Primary hyperparathyroidism and papillary
adenocarcinoma of the thyroid: Report of four cases. Proc Staff Meet
Mayo Clin 1956;31:295-8.
2. Stoffer SS, Szpunar WE, Block M. Hyperparathyroidism and thyroid
disease. Postgrad Med 1982;71:91-4.
3. Lever EG, Refetoff S, Straus FH 2nd, Nguyen M, Kaplan EL.
Coexisting thyroid and parathyroid disease – Are they related?
Surgery 1983;94:893-900.
4. Sato Y, Sakurai A, Miyamoto T, Hiramatsu K, Hashizume K,
Katai M, et al. Hyperfunctioning thyroid adenoma concomitant with
papillary thyroid carcinoma, follicular thyroid adenoma and primary
hyperparathyroidism. Endocr J 1998;45:61-7.

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Amenorrhea with
myxedema: A hidden
clue

Sir,

Menstrual disturbances are common in thyroid dysfunction
with menorrhagia in hypothyroidism and oligomenorrhea
in hyperthyroidism. Primary hypothyroidism (PH) indicates
thyroid gland dysfunction where as hypothalamic pituitary
abnormality results in central hypothyroidism (CH).

Myxedematous features coupled with elevated thyrotropin
are seen in PH differentiating it from CH. In an obvious case
of  hypothyroidism, amenorrhea rather than menorrhagia
and normal thyrotropin levels indicates CH.

We report
a young woman with secondary amenorrhea, myxedema,
normal thyrotropin and abnormal neuroimaging.

A 30-year-old woman presented with 3-year history of
irregular menses progressing to secondary amenorrhea for
the past 1 year along with easy fatigability and weight gain.
She delivered two children and there was no history of
peripartum complications or lactational failure. She denied
seizures, headache, vomiting, visual defects, polyuria and
loss of  axillary or pubic hair. Screening for hypothyroidism
with thyrotropin was normal on two occasions.

Examination revealed normotension, obesity, bradycardia,
hypothermia, pallor, dry skin, pretibial edema, dull
expression-less face with puffy lips and no goiter [Figure 1a].
Rest of  the systemic examination was normal except for
delayed relaxation of  deep tendon jerks. Biochemistry
showed dyslipidemia and elevated creatinine phosphokinase.

Hormonal profile revealed free triiodothyronine
−2.9 pmol/L (normal 3.1‑6.8), free thyroxine −0.34 ng/dL
(normal 0.7‑1.15), thyrotropin −3.2 mIU/L (normal
0.3‑4.5), prolactin −14 ng/ml (normal 0‑15), LH 0.6 IU/L
(normal 0‑7), FSH 2.5 IU/L (normal 2‑10), estradiol
26 pg/mL  (normal 25‑120). Peak cortisol and GH
after hypoglycemia were 22.4 µg/dL and 7.16 ng/mL,
respectively. MRI showed flat pituitary gland with empty
sella [Figure 1b]. She was diagnosed as central hypothyroidism
and hypogonadism with primary empty sella and treated
with levothyroxine, estrogen + progesterone, calcium and
vitamin D supplements.

Our case posed a diagnostic dilemma with florid
manifestations of  hypothyroidism but normal thyrotropin
repeatedly. Myxedema is classically described in cases of
PH and rarely with central hypothyroidism. Screening
with isolated thyrotropin lead to delayed diagnosis, as
patients of  CH have normal or low thyrotropin. Delayed
presentation of  the primary empty sella with CH and
hypogonadism is another unusual finding in our case.

She had no history to suggest Sheehan’s syndrome or
secondary empty sella. Primary empty sella is seen mostly
in females and endocrinal disturbances are seen in about
10‑35% of  patients.

Dynamic testing of  pituitary
hormones resulted in unmasking of  hitherto unreported
abnormalities in these patients.

Figure 1: (a) Clinical photograph showing myxedematous facies
(b) Pituitary
MRI showing empty sella in coronal section