

Dual malignancy in adolescence: A rare case report of metachronous papillary carcinoma of thyroid following dysgerminoma of ovary

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Dual malignancy is rare in adolescents. Dual malignancy with the second malignancy of thyroid is rare. No association has been reported between dysgerminoma of ovary and carcinoma thyroid in medical literature. Despite a thorough PubMed search (key words — Papillary carcinoma of thyroid, metachronous, dysgerminoma ovary), we were unable to find a previous reported case of metachronous papillary carcinoma of thyroid (PTC) following dysgerminoma of the ovary. After surgery, the patient is being regularly followed up for recurrence/development of new primary. We report this unusual and rare case in a 17-year-old female patient.

Key words: Dual malignancy, dysgerminoma, papillary carcinoma of thyroid (PTC)

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INTRODUCTION

Dual malignancy in young adults is a rare entity. Of all well-differentiated thyroid carcinoma patients, 5% are familial. Majority familial thyroid cancer are nonmedullary familial thyroid cancer, and have been shown to be present in familial cancer syndromes such as familial adenomatous polyposis, Cowden syndrome, Carney complex, Pendred syndrome, and Werner syndrome.^[1] Herein, we report a rare case of a 17-year-old female patient diagnosed with papillary carcinoma of thyroid (PTC) with an antecedent history of dysgerminoma of the right ovary. To our knowledge, no association has been reported between dysgerminoma of the ovary with carcinoma thyroid in literature.^[2]

CASE REPORT

A 17-year-old female, following right lobectomy of the thyroid gland, with a histopathological report of PTC was referred to the Department of Surgical Oncology

at Sri Aurobindo Medical College and Post Graduate Institute in Indore, Madhya Pradesh in India for further management in 2014. Examination of the neck revealed a scar from a previous surgery, with no palpable neck nodes. Indirect laryngoscopy (IDL) revealed bilateral mobile vocal cords. Blood investigations revealed serum T3 1.19 ng/mL (0.8-2 ng/mL), serum T4 of 5.88 ug/dL (5.1-14.1 ug/dL), serum TSH of 6.04 Uiu/mL (0.27-4.20 Uiu/mL), serum level of lactate dehydrogenase (LDH) 157 Iu/L (140-280 Iu/L). Previous history revealed hysterectomy with right salpingo-oophorectomy performed 3 years back during evaluation of primary amenorrhea with right ovarian mass [Figure 1]. Diagnostic laparoscopy revealed rudimentary double uterus with a huge right ovarian mass. Gross examination revealed 1300g right ovarian mass with fallopian tube with didelphys of rudimentary uterus. Microscopy confirmed the diagnosis of dysgerminoma of the right ovary Stage I a [Figure 2a]. The patient had taken chemotherapy bleomycin, etoposide, and cisplatin (BEP) regime, i.e., bleomycin IV per week X9; dose at 20 U/m², etoposide 100 mg/m² days 1-5 q3wk X3, cisplatin 20 mg/m² days 1-5 q3wk X3 at a 4

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week interval in the adjuvant setting. Magnetic resonance imaging (MRI) of the neck and the pelvis was performed before completion; thyroidectomy revealed postoperative changes with irregular nodular soft tissue in the region of the right lobe of thyroid with subcentimeter lymph node in bilateral deep upper cervical region [Figure 3a] and left ovary was normal in appearance, measuring 2.9 cm × 4.7 cm [Figure 3b]. Completion thyroidectomy with bilateral central neck node dissection was performed that was negative for residual malignant disease. Radioactive iodine scan (I^{131}) performed 1 year after completion thyroidectomy did not reveal any residual disease.

DISCUSSION

Billroth, in 1889, was the first to report dual malignancy.^[3] Second malignancy can be synchronous or metachronous. Two or more histologically different malignancies identified within the first 6 months of the first malignancy is defined as synchronous and if the second tumor is identified beyond 6 months, it is defined as metachronous. Both the tumors should be malignant and neither should be metastasis of each other and both the tumors should be morphologically and microscopically distinct from each other.^[4] In our case, there were two distinct pathologies of dysgerminoma of the ovary and PTC, both anatomically and histologically different entities with a gap of 3 years between both [Figure 2a and b]. The incidence of second primary in a cancer patient is about 10%.^[5] Studies have reported relative risks of second primary cancers ranging from 1.08-1.3.^[6] Dysgerminomas account for 1-5% of all ovarian malignancies in the first two decades of life.^[7] Of the cases, 80% are reported in females in the second decade of life with complaints of delayed menarche and primary amenorrhea, which were the presenting complaints of our patient as well, 3 years back. Dysgerminoma is the only germ cell malignancy with a significant rate of bilaterality. The treatment of early dysgerminoma is resection of the primary lesion. Chemotherapy is advised in the adjuvant setting to prevent metastasis. In patients whose contralateral ovary has been preserved, some diseases can develop in 5-10% of the retained gonads over the next 2 years.^[8] The patient was advised BEP regime after hysterectomy, following diagnosis of dysgerminoma. The possibility of developing dysgerminoma in the contralateral ovary was ruled out with MRI of the pelvis that reported a normal left ovary and Sr LDH 157 Iu/L. PTC is the most common well-differentiated cancer of the thyroid. It represents approximately 80-85% of well-differentiated thyroid cancers. Histopathology report following right lobectomy of the thyroid revealed PTC, following which, completion thyroidectomy with central neck dissection (CND) has been performed [Figure 5]. Radiation exposure is the most well-known risk factor for PTC.^[9] No such history was present



Figure 1: CECT abdomen showing mass replacing the right ovary with nonvisualization of the uterus

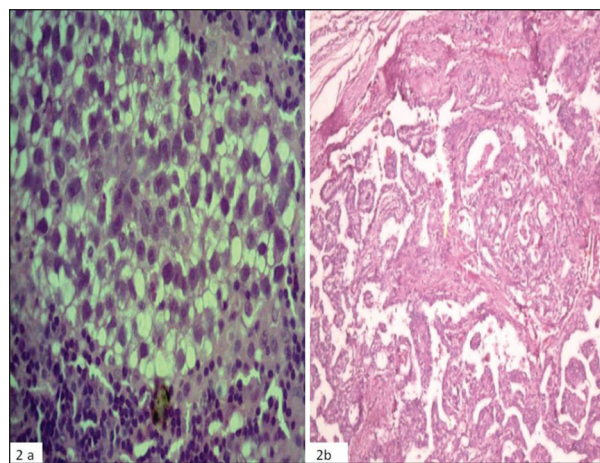


Figure 2: (a) Microphotograph showing islands of large polygonal tumor cells surrounded by lymphoid cells. The tumor cells possess clear cytoplasm and centrally placed nuclei with vesicular chromatin and prominent nucleoli. (H&E 400×) and (b) Microphotograph showing tumor cells arranged in papillary fronds having vesicular coffee bean nuclei. (H&E 100×)

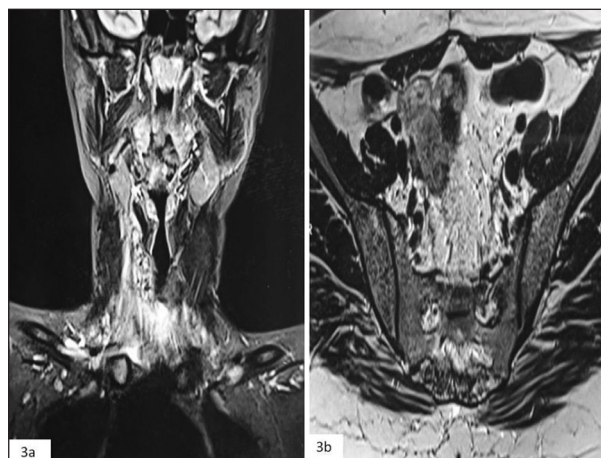


Figure 3: (a) MRI of the neck showing postoperative changes in the neck with irregular nodular soft tissue in the region of the right lobe of thyroid with subcentimeter lymph node in bilateral deep upper cervical region and (b) MRI of the pelvis showing normal-appearing left ovary with absent right ovary and uterus

in our case. A large retrospective study evaluating second primary malignancy in thyroid cancer patients found second primary malignancy in 0.45% of cases, out of which only 0.067% cases had thyroid cancer as the second primary malignancy and all had a history of exposure to radiation.^[10] This finding was not observed in our case. The association between carcinoma thyroid, cancer of breast, kidney, salivary glands, brain, central nervous system, scrotum, and leukemia has been reported.^[2] DTC accounts for 95% of thyroid malignancies, 5% being familial. But association between dysgerminoma and carcinoma thyroid has not been reported in literature to the best of our knowledge. Despite a thorough PubMed search, we were unable to find a previous reported case of metachronous papillary carcinoma thyroid following dysgerminoma of the ovary.

CONCLUSION

Metachronous malignancy with the index tumor of ovarian cancer has been reported, but the case of index tumor of dysgerminoma of the ovary with a second primary tumor diagnosed as PTC is unusual. The case of dual primary malignancy has not been documented in literature; we present this case due to the rarity in presentation.

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Conflicts of interest

There are no conflicts of interest.

AUTHOR'S CONTRIBUTION

SC contributed in the conception of the work, conducting the study, writing and approval of the final version of the manuscript, and agreed for all aspects of the work.

SMD contributed in the conception of the work and approval of the final version of the manuscript, and agreed for all aspects of the work.

DYM contributed in revising the draft and approval of the final version of the manuscript, and agreed for all aspects of the work.

SS contributed in the conception of the work, revising the draft, and agreed for all aspects of the work.

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