

Adult E-Poster

CASE

We report a case of a 68-year-old woman of Indian ethnicity who first presented to us at 60 years old in 2017 with postmenopausal hirsutism. Treatment with co-cyprindiol (Diane-35) for a year and spironolactone did not alleviate her symptoms but instead worsened them with other virilizing symptoms such as deepening of voice, breast atrophy and androgenic alopecia. Testosterone levels were persistently elevated [43.757 nmol/L (December 2016) – 48 nmol/L (July 2017) - >52 nmol/L (October 2019)]. Computed tomography imaging done in 2020 showed an enlarged right ovary. She was referred to Gynecology and was given one dose of Leuprorelin (Lucrin) on 11/7/2020, with the intention to assess ovarian suppression; however, elevated testosterone levels persisted at >52 nmol/L. The patient eventually underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAHBSO) in May 2021 and histopathology revealed a right ovarian Sertoli-Leydig cell tumor. Post-operation testosterone levels showed rapid reduction to normal at 0.3 nmol/L and remained normal at <0.1 nmol/L in September 2021.

CONCLUSION

This case emphasizes the importance of thorough evaluation in women with postmenopausal virilization, which can be the only sign of rare ovarian tumors. Additionally, this condition can be distressing to patients and affect their quality of life, especially social interactions. The delay in her diagnosis and surgery highlights the need to increase awareness of this condition among clinicians.

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MALIGNANT STRUMA OVARIII IN A PATIENT WITH GRAVES' DISEASE: A CASE REPORT

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INTRODUCTION/BACKGROUND

Malignant struma ovarii (MSO) is a rare ovarian teratoma containing malignant thyroid tissue, accounting for <5% of all struma ovarii cases. Papillary carcinoma is the most common histological subtype, followed by follicular carcinoma. Diagnosis may be challenging, especially when coexisting with thyroid nodules or autoimmune thyroid diseases such as Graves' disease, due to overlapping histologic and functional features.

CASE

A 58-year-old female with longstanding hyperthyroidism due to Graves' disease presented with an abdominal mass measuring 15 × 10 × 9 cm. Laparotomy in September 2023 revealed a right ovarian tumor. Histopathology showed adenomatous struma with focal atypia. Immunohistochemistry revealed BRAF V600E positivity with partial CD56 and CK19 expression, and negative HBME1 and cyclin D1. Although non-classical, this staining pattern can be observed in thyroid carcinoma with oncocytic or clear cell features.

Thyroid ultrasound showed a bilateral multinodular goiter with a TIRADS 4 nodule; FNAB was benign. A thyroid scan revealed diffusely increased uptake (55.3%), and elevated TRAb (4.43 IU/L), consistent with Graves' disease. Total thyroidectomy in June 2024 revealed adenomatous struma with chronic inflammation and no malignancy, likely representing a degenerating nodule in the context of treated Graves' disease.

This case highlights the complexity of diagnosing MSO in the setting of autoimmune thyroid disease. Total thyroidectomy was performed to exclude primary thyroid carcinoma and to support future surveillance or radioactive iodine therapy.

CONCLUSION

MSO should be considered when ovarian tumors contain thyroid tissue with atypia. In patients with Graves' disease, degenerating thyroid nodules may mimic malignancy. A multidisciplinary approach using imaging, histopathology, immunohistochemistry, and autoantibody testing is essential for accurate diagnosis and long-term management.

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ALPHA LIPOIC ACID-INDUCED INSULIN AUTOIMMUNE SYNDROME (IAS): A REPORT OF TWO CASES

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INTRODUCTION/BACKGROUND

Insulin Autoimmune Syndrome (IAS) is a rare cause of hypoglycemia. Alpha-lipoic acid (ALA), found in Bionerv (Vitamin B supplement), induces IAS by modifying insulin structure, leading to insulin autoantibody (IAA) production in genetically susceptible individuals. Most cases are self-limiting. We present two IAS cases, emphasizing diagnostic and management challenges.

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CASE

A 49-year-old man presented with recurrent hypoglycemia. During his first clinical consult, a computed tomography (CT) scan of the pancreas was done and showed normal findings. Prolonged fasting test was consistent with endogenous hyperinsulinemic hypoglycemia. He was later referred to our center. In further history, he revealed that his symptoms started 10 days after taking Bionerv. A mixed meal test showed fasting hypoglycemia with late postprandial hypoglycemia, and markedly elevated serum insulin levels. His IAA was positive, and his sulfonyleurea screen was negative. A diagnosis of ALA-induced IAS was made. Despite stopping ALA and dietary modifications, his symptoms persisted, requiring diazoxide and prednisolone. The patient was monitored with continuous glucose monitoring (CGM), which revealed episodes of alternating hyper and hypoglycemia.

A 69-year-old man with a history of thyrotoxicosis presented with symptoms of hypoglycemia. Prolonged fasting confirmed endogenous hyperinsulinemia. Abdominal CT and endoscopic ultrasound (EUS) were normal, while Gallium-68 DOTOTATE imaging showed mild uptake at a 10 mm pancreatic tail nodule. He was suspected of having insulinoma and was referred to us for further assessment. Further history revealed that the symptoms started after 1 week of taking Bionerv. His IAA was positive. In view of the temporal relationship with ALA, an IAS diagnosis was made. Symptoms improved after discontinuation of ALA, dietary modification and medical therapy. His CGM showed predominant hyperglycemia with late evening hypoglycemia.

CONCLUSION

Although ALA is generally safe, emerging case reports demonstrate its potential to trigger IAS. Detailed drug history and clinical suspicion is crucial to avoid the misdiagnosis of insulinoma and unnecessary interventions.

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OBSTRUCTIVE JAUNDICE FOLLOWING MIBG THERAPY IN MALIGNANT PHEOCHROMOCYTOMA: A CASE REPORT

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INTRODUCTION/BACKGROUND

Malignant pheochromocytoma is a rare neuroendocrine tumor with potential for local invasion and distant meta-

stasis. In inoperable cases, nonsurgical options include I-131 metaiodobenzylguanidine (MIBG) therapy. MIBG-related complications may occur, especially in patients with bulky or anatomically complex tumors. We describe a case of post-MIBG ascending cholangitis due to tumor-related biliary obstruction.

CASE

A 49-year-old Kadazan Muslim female with hypertension, type 2 diabetes, dyslipidaemia, and previous strokes was diagnosed in August 2022 with inoperable malignant right adrenal pheochromocytoma. She presented with right hypochondriac pain, weight loss, palpitations, diaphoresis, and postural hypotension. Imaging revealed a right suprarenal mass (6.3 × 4.9 × 7.5 cm) invading the inferior vena cava and right renal vein. Elevated urine normetanephrine and positive DOTATATE, FDG-PET, and MIBG scans confirmed a functional tumor. Due to high surgical risk, she declined surgery and underwent right adrenal artery embolization with stable disease on follow-up. In July 2024, she received high-dose I-131 MIBG therapy (211 mCi) for palliative intent. Eight days post-therapy, she developed fever and jaundice. Imaging revealed intrahepatic biliary dilatation secondary to tumor compression at the porta hepatis. She was diagnosed with ascending cholangitis complicated with gram-negative sepsis and thrombocytopenia. She was managed with intravenous antibiotics, biliary stenting and supportive care.

CONCLUSION

Obstructive jaundice is a rare but serious complication following MIBG therapy. In this case, tumor compression near the porta hepatis likely exacerbated by post-therapy inflammation or necrosis, led to biliary obstruction. Although preoperative biliary stenting is standard in pancreaticobiliary malignancies, its use in neuroendocrine tumors, including pheochromocytoma, is not well defined. This case supports the potential role of pre-emptive biliary decompression in select high-risk patients undergoing MIBG therapy. Multidisciplinary planning is essential for risk stratification and outcome optimization.