

CASES

We report four young adults (aged 21–37 years) with heterogeneous manifestations of MAS type 3 involving endocrine and systemic autoimmune diseases. Autoimmune thyroid disease was identified in three patients, all diagnosed with Graves' disease with suppressed thyroid-stimulating hormone, elevated free thyroxine, and positive thyrotropin receptor antibodies. Autoimmune diabetes was present in three patients, including latent autoimmune diabetes in adults (LADA) and type 1 diabetes mellitus, with variable glycemic control (hemoglobin A1c range 6.7–13.9%) and C-peptide levels ranging from preserved to markedly reduced. Myasthenia gravis was observed in three patients. Additional autoimmune conditions included systemic lupus erythematosus, systemic sclerosis, rheumatoid arthritis, and ulcerative colitis. Notably, rare combinations such as Graves' disease with LADA and myasthenia gravis, as well as coexistence with systemic autoimmune diseases, were identified. All patients received individualized multidisciplinary management. Clinical and biochemical improvement was observed in all cases, with stabilization of both endocrine and systemic autoimmune manifestations.

CONCLUSION

This case series highlights the heterogeneous and expanding clinical spectrum of MAS type 3, including rare combinations of autoimmune endocrine and systemic diseases in young adults. Early recognition of autoimmune clustering and comprehensive screening are essential to optimize management and improve outcomes. These findings provide insights into autoimmune disease clustering and support the need for proactive multidisciplinary management strategies.

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A RARE DIAGNOSIS OF INSULIN AUTOIMMUNE SYNDROME CAUSING RECURRENT HYPOGLYCEMIA

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INTRODUCTION

Insulin autoimmune syndrome (IAS), or Hirata syndrome, is characterized by hyperinsulinemic hypoglycemia resulting from the presence of high titers of insulin autoantibodies (IAAs) in the absence of exogenous insulin. We present a rare case of IAS.

CASE

A 75-year-old female with underlying hypertension, dyslipidemia, and osteoporosis presented with symptoms suggestive of spontaneous hypoglycemia of 3 months duration. She was non-diabetic with no history of antidiabetic agents or exogenous insulin use. Notably, she had taken traditional medications 1 month prior to onset of symptoms.

Her fasting blood glucose was 2 mmol/L. Serum insulin was significantly elevated at >1,000 iui/mL and serum C peptide was 2,950 pmol/L, consistent with hyperinsulinemic hypoglycemia. Blood sulfonylurea level was not available. Her complete blood count and renal and liver function were normal. Thyroid function test was normal, and a short corticotropin stimulation test showed adequate cortisol response. Computed tomography (CT) pancreas, endoscopic ultrasound and PET-CT scan with Galium-68 DOTATATE showed no evidence of insulinoma. Serum IAAs were raised at 175 IU/mL, which confirmed a diagnosis of IAS.

The patient was managed with dietary modifications and advised for frequent, small meals with low glycemic index, incorporating oral raw cornstarch. She was also started on oral diazoxide 100 mg twice daily (3 mg/kg/day) due to persistent hypoglycemia. She responded well to diazoxide with no more spontaneous hypoglycemia, however, developed fluid retention which was managed with oral diuretics. Subsequently, we managed to taper and stop the diazoxide after 18 months of treatment. Patient remains well with no further episodes of hypoglycemia.

CONCLUSION

IAS may be triggered by medications or viral infections, occurs more frequently in people with autoimmune conditions, and shows genetic predisposition. However, as in our patient, IAS may be idiopathic, and the cause remains unknown. IAS is frequently self-limiting, and our patient experienced spontaneous remission with no further hypoglycemic episodes after discontinuation of treatment.