CENTRO HOSPITALAR UNIVERSITÁRIO LISBOA NORTE, EPE | HOSPITAL DE SANTA MARIA | Hospital Pulido Valente

MEN-1 phenotype or a series of unfortunate events?

Maria Inês Alexandre[1], José Vicente Rocha[1], Ana Gomes[1]

40 year-old-woman

Diagnosed with **Graves' disease**

Started on thiamazole 15 mg/day

Some days later…

Episodes of lightheadness, hunger and visual disturbances associated with **low glucose** that reverted with food ingestion

Hypoglycemias

New blood tests were requested three weeks after

TSH ↓↓ = <0.01 uU/mL
FT4 ↑ = 2.07 ng/dL (N:0.85-1.7)
TRAbs 7.84 U/L

Glucose ↓ = 39 mg/dL

PTH ↑ = 125 pg/mL (N:10-55)

Graves' hyperthyroidism

Hypoglycemia

Elevated PTH

Cranial CT-scan : (…) small area of hypocaptation in the pituitary that suggested a possible microadenoma.

On one of these episodes a cranial CT-scan was also requested

Hypoglycemias (insulinoma???)
+ Hyperparathyroidism
+ Pituitary adenoma
???

MEN-1?

Endocrinology appointment

Hypoglycemia investigation
Glucose 41 mg/dL
Elevated insulin levels (50.4 uU/mL; N:2.6-24.9)
High titers of insulin autoantibodies (30 U/mL; N<20)
Normal abdominal CT-scan

Insulin autoimmune syndrome (IAS) triggered by thiamazole?

Patient started prednisolone and thiamazole was reduced

Hypoglycemia episodes resolved

Hyperparathyroidism investigation

Normal calcium and phosphate (9.3 mg/dL; 2.5 mg/dL, respectively)
Low vitamin-D (25-OH VitaminD=9 ng/mL)

Successfully treated with cholecalciferol

Pituitary adenoma investigation

Cranial MRI revealed normal pituitary
Blood tests: Normal pituitary function

- **Insulin autoimmune syndrome** is a rare condition characterized by hypoglycemia and autoantibodies directed towards insulin. The thiamazole-related autoimmunity has been reported previously, mostly in Japan.
- The **association between this rare condition** and other features (hyperparathyroidism and pituitary adenoma) could mislead into the diagnosis of MEN-1.
- This highlights the importance of a detailed anamnesis and to **carefully access the need for some complementary exams**, as some incidental findings can lead to unnecessary costly exams and worries.

1- Endocrinology, Diabetes and Metabolism Department, Hospital de Santa Maria, CHULN, EPE, Lisbon, Portugal