ADULT-ONSET NESIDIOBLASTOSIS CAUSING HYPERINSULINEMIC HYPOGLYCEMIA: DIAGNOSIS AND TREATMENT CHALLENGE. A CASE REPORT

Author: D.T. Cosma[1], A. Silaghi [2,3], C. Georgescu[2,3], I.A. Veresiu[1,3]
Coordinator: I.A. Veresiu
[1]Diabetes, Nutrition and Metabolic diseases Clinical Center, Cluj-Napoca
[2]Endocrinology Clinic, Cluj-Napoca
[3]"Iuliu Hatieganu" University of Medicine and Pharmacy, Cluj-Napoca

Nesidioblastosis is defined as a diffuse proliferation of primitive pancreatic islet cells budding from ductal epithelium.[1]

A 41-year-old male operated for a duodenal ulcer (1999) was referred to our center via emergency room with suspicion of an insulinoma after an episode of loss of consciousness with a glycemia (G) of 24mg/dl. Current symptoms started 4 days prior to admission with dizziness, blurred vision, sweating, tremor in the upper limbs which resolved after ingestion of foods with high glycemic index.

Further questioning revealed 2 similar episodes in September 2012 investigated in a cardiology and respectively neurological service and one more in March 2012 during surgery for intestinal adhesion with G=18mg/dl, no further investigated.

At admission: altered general status with retrograde amnesia, dry skin and mucous membrane and deep tenderness in the epigastrum, BP=125/75mmHg, Pulse=65b/min.

Laboratory findings	**72-hour fast**	**72-hour fast (under 300mg HCH)**
sO_2= 92.2% Ca^{2+} = 1.03 mmol/l Cl = 110 mmol/l Lac = 24 mg/dl G = 102 mg/dl PTH = 81mg/dl	Started 09:30 Stopped: 12:00 G = 32 mg/dl Plasma insulin= 86.3 µU/ml G/I = 0.37	Started 08:00 Stopped 12:00 08:00: G = 150 mg/dl 10:00: G = 118 mg/dl 12:00: G = 121 mg/dl

Abdominal US	**Contrast CT and MRI**	**Endoscopic US**
- no visualization of the pancreas; - bilateral renal microlithiasis.	- Excluded renal microlithiasis; - normal pancreas.	- pseudolobular aspect in the pancreas tail and body.

The patient was guided to a surgical clinic with dietetic recommendation and Phenytoin 200mg/day for insulinoma localization and surgical treatment. He was admitted into the surgical clinic after 3 weeks from the previous hospitalization with only 2 moderate hypoglycemic episodes under Phenytoin treatment.

Short Octreotide test

Debut:
08:00 G=60mg/dl;
100 ug s.c. Octreotide
09:00 G=94mg/dl
10:00 G=34mg/dl

Serotonin serum = 75µg/l
Chromogranin A = 53 µg/l
5-hydroxy-indoleacetic acid = 3mg/24h

Due to repeated hypoglycemic episodes after Phenytoin treatment was stopped, exploratory laparotomy was performed with intraoperative ultrasonography which exhibited two possible tumors in the pancreas tail and body. A subtotal splenopancreatectomy was performed. Microscopic view was consistent with nesidioblastosis and no tumor was found. The patient remains euglycemic ten months post-operatively suggesting that the source of insulin excess had been removed.

Conclusions

Nesidioblastosis:
- is a rare cause of adult hypoglycemia;
- pre-operative differentiation from insulinoma is difficult;
- suspected when imaging studies are negative.

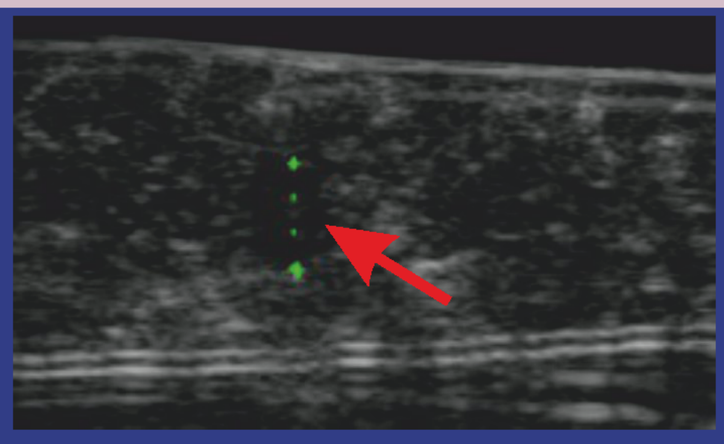

Fig. 1: Intraoperative US: hypoechogenic nodule (6.3mm) in the pancreatic tail

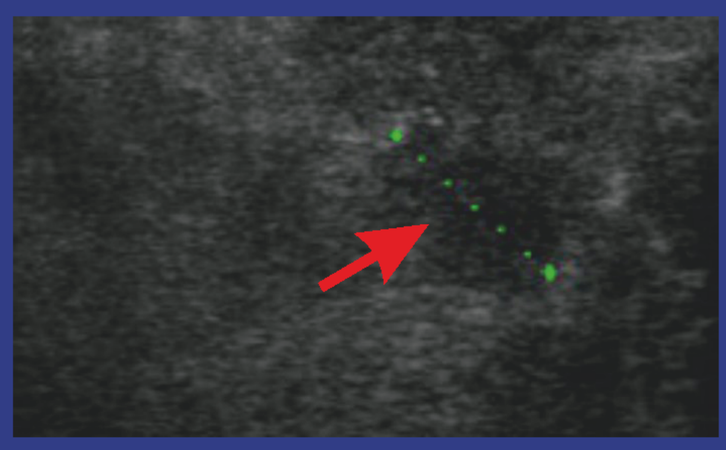

Fig. 2: Intraoperative US: hypoechogenic nodule (11.8mm) in the pancreatic body

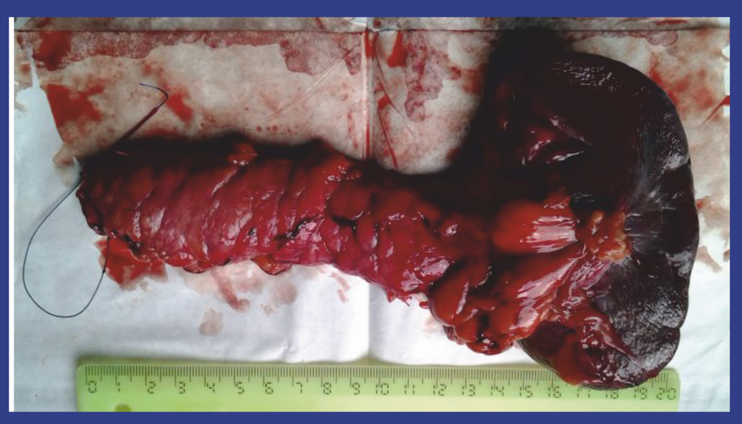

Fig. 3: Post resection view: accentuated lobulation in the pancreatic tail and body

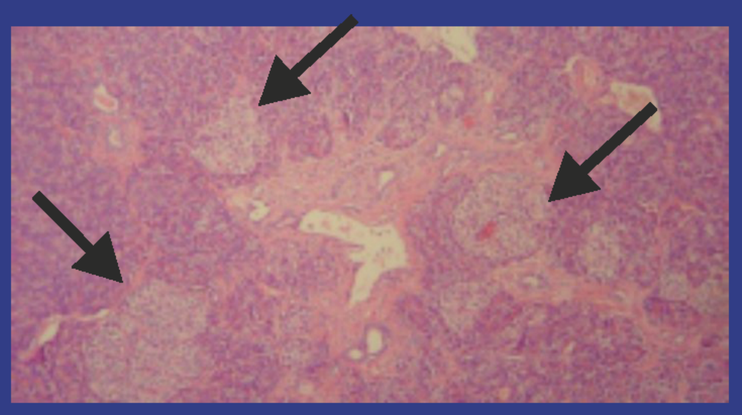

Fig. 4: Several islet cells surrounding pancreatic duct (H&E stain, OM x 10)

Fig. 5: Small islet arising from pancreatic ductule (H&E stain, OM x 40)

References:

1. Fong TL, Warner NE, Kumar D (1989) Pancreatic nesidioblastosis in adults. Diabetes Care 12:108-114

17 ECE
1104--EP
Endocrine tumours
Daniel-Tudor Cosma
DOI: 10.3252/pso.eu.17ece.2015
Poster presented at: ECE2015 16–20 May 2015 DUBLIN, IRELAND 17th EUROPEAN CONGRESS OF ENDOCRINOLOGY