INSULINOMA PRESENTING ITSELF AS A NIGHT PAROXYSMAL DISORDER WITH SPONTANEOUS RECOVERY

BOLESNIK SA INZULINOMOM KOJI SE PREZENTIRAO KAO PAROKSIZMALNI NOĆNI POREMEĆAJ SA SPONTANIM OPORAVKOM

LEJA DOLENC GROŠELJ, DUŠAN BUTINAR*

Descriptors: insulinoma; polysomnography; night paroxysmal movement disorder

Summary. A 64-year-old woman with night paroxysmal episodes is described. Her symptoms began 9 months ago with attacks of bizarre movement, which were always present in the second part of the night. She had no attacks during the daytime. Her husband reported confusion and disorientation followed by long periods of unresponsiveness. The patient underwent a night polysomnography recording. Around 4 o'clock in the morning bizarre movements with stereotypic behaviour appeared. She was rolling her head from side to side, moaning, and stretching her limbs. These periods first lasted for minutes, and were constantly repeated during the night. EEG findings suggested metabolic encephalopathy. At that time finger prick test revealed a profound hypoglycaemia (1.2 mmol/l), high insulin (200 pmol/l), and C-peptide (6.63 nmol/l). Ultrasonography and MRI confirmed the insulinoma in the head of the pancreas. To our knowledge our case is the first patient with insulinoma attacks only during sleep time.

Deskriptori: inzulinom, polisomnografija, poremećaj paroksizmalnih pokreta noću

Sažetak. U radu je opisana 64-godišnja žena s epizodama paroksizama tijekom noći. Simptomi su započeli prije 9 mjeseci s atakama bizarnih pokreta, koje su se uvijek javljale u drugom dijelu noći. tijekom dana nije imala atake. Njezin suprug navodi da je bila konfuzna i dezorijentirana, nakon čega je slijedilo duže razdoblje nereagiranja. Pacijentici je učinjena noćna polisomnografija. Oko 4 sata ujutro pojavili su se bizarni pokreti uz stereotipno ponašanje. Vrtjela je glavom s jedne na drugu stranu, stenjala i istezala udove. Ovo je isprva trajalo nekoliko minuta, a tijekom noći stalno se ponavljalo. EEG nalaz je upućivao na metaboličku encefalopatiju. U to je vrijeme u krvi iz prsta nađena izrazita hipoglikemija (1,2 mmol/l), visok inzulin (200 mmol/l) i C-peptid (6,63 μmol/l). Ultrasonografija i MEI potvrdile su inzulinom u glavi pankreasa. Prema našim saznanjima ovo je prvi slučaj ataka inzulinom tijekom spavanja.

Liječ Vjesn 2008;130:104–105

Case report

A 64-year-old woman was referred for polysomnography to get a diagnosis of night paroxysmal episodes. She had a hypernefroma of the right kidney with nefrectomy performed a year ago and was treated for hyperlipidemia and hypertension. Her symptoms began 9 months ago with attacks of bizarre movement, which were always present in the second part of the night. She had no attacks during the daytime. Her husband reported confusion and disorientation followed by long periods of unresponsiveness (1–2 hours). Twice during the attack she fell from bed and sustained light injuries. Neurological, general examination and daytime EEG were normal. MRI brain scan demonstrated a few small ischemic lesions in periventricular regions on both sides. During the last 9 months a psychiatrist and a neurologist diagnosed her with parasomnia? late onset epilepsy? Diazepam in the evenings was prescribed by the psychiatrist.

The patient underwent a polysomnography recording. In the first part of the night her sleep was normal. Some obstructive apnoeas (AHI = 8) were found. Around 4 o'clock in the morning bizarre movements with stereotypic behaviour appeared. She was rolling her head from side to side, moaning, and stretching her limbs. Tonic or clonic components were absent. No dystonic postures were observed. These periods first lasted for minutes, and were constantly repeated. During these attacks she was intermittently confused and disoriented. After 6 o'clock she became cold, sweaty and confused. EEG showed bilateral slow theta and delta activity, accentuated in the centro-temporal regions bilaterally. There was no epileptiform activity. EEG findings suggested metabolic encephalopathy. At that time finger prick test revealed a profound hypoglycaemia (1.2 mmol/l). High insulin (200 pmol/l) and C-peptide (6.63 nmol/l) secretion were found. After administration of sweet tea and 5% glucose infusion she become oriented and well. Polysomnography showed wake state with alpha activity. The patient was transferred to the Endocrine Unit. Ultrasonography of abdomen was negative, while on the endoscopic ultrasonography hypoechogenic lesion- insulinoma- (2.9×1.5 cm) was found in the head of the pancreas. MRI (T2 FAT SAT) confirmed hyper intensive round formation (2.2×1.8 cm) in the head of the pancreas. While waiting for surgery the patient was given a late dinner and a small snack during the night and was completely free of night attacks. Two months later surgery of the pancreas was performed, but insulinoma was not found. Currently she is under endocrine and neurological observation as an outpatient and is practicing the same eating regime. Measurements of glucose are in normal range. She has been completely free of above mentioned nighttime events for a year.

* **Institute of Clinical Neurophysiology, University Medical Centre, Ljubljana, Slovenia** (dr. sc. Leja Dolenc Grošelj, dr. med.; dr. sc. Dušan Butinar, dr. med.)

Adresa za dopisivanje: Dr. L. Dolenc Grošelj, Institute of Clinical Neurophysiology, University Medical Centre, Ljubljana, Slovenia, e-mail: leja.dolenc@kclj.si

Primljeno 7. veljače 2008., prihvaćeno 22. travnja 2008.

Conclusion

Insulinoma is pancreatic endocrine tumour characterized by hyperinsulinemic hypoglycaemia, which can mimic various neurological pictures (acute disorder of cognition, consciousness, epilepsy, transient ischaemia, psychosis or chronic disorders of dementia and neuropathy.[1–4] To our knowledge our case is the first patient with insulinoma attacks only during sleep time. Therefore we suggest, that when bizarre paroxysmal movement is present only during nighttime, metabolic disorders are considered in the differential diagnosis.

REFERENCES

1. *Graves TD, Gandhi S, Smith SJM, Sisodiya SM, Conway GS.* Misdiagnosis of seizures: insulinoma presenting as adult-onset seizure disorder. J Neurol Neurosurg Psychiatry 2004;75:1091–1092.
2. *Shaw C, Haas L, Miller D, Delahunt J.* A case report of paroxysmal dystonic choreathetosis due to hypoglycemia induced by an insulinoma. J Neurol Neurosurg Psychiatry 1996;61(2):194–5.
3. *Striano P, Striano S.* Insulinoma presenting as refractory late-onset epilepsy. Epilepsia 2006;47(2):452.
4. *Dion MH. Cossette P, St-Hilaire JM i sur.* Insulinoma misdiagnosed as intractable epilepsy. Neurology 2004;62(8):1443–5.

* * *

Vijesti
News

European Society of Pathology
Academy of Medical Sciences of Croatia
Institute for Clinical Medical Research of Sestre milosrdnice University Hospital Zagreb
Veterinary Faculty Zagreb

19th Ljudevit Jurak International Symposium on Comparative Pathology



SECOND ANNOUNCEMENT AND CALL FOR PAPERS

June 6–7, 2008
Multimedia center, Sestre milosrdnice University Hospital,
Vinogradska cesta 29, Zagreb, Croatia

(http://www.kbsm.hr/Jurak/symposium.htm)

The main symposium topics are Pediatric Pathology and Advances in Pathomorphology Techniques.

PRVI HRVATSKI KONGRES DILEMA U NEUROLOGIJI
s međunarodnim sudjelovanjem

8.—12. listopada 2008.
Opatija, Grand Hotel Adriatic, Hrvatska

Organizatori kongresa:

Klinika za neurologiju Medicinskog fakulteta i Kliničkog bolničkog centra Zagreb
Hrvatsko društvo za EEG i kliničku neurofiziologiju, HLZ
Hrvatsko neurobiološko društvo, HLZ
Hrvatsko društvo za neuromuskularne bolesti i EMNG, HLZ
Sekcija za intenzivnu neurologiju Hrvatskog neurološkog društva, HLZ
Referentni centar za epilepsije Ministarstva zdravstva i socijalne skrbi RH
Referentni centar za neuromuskularne bolesti Ministarstva zdravstva i socijalne skrbi RH
Referentni centar za demijelinizacijske bolesti središnjeg živčanog sustava Ministarstva zdravstva i socijalne skrbi RH

Kontakt osoba: gđa Aleksandra Novosel, Klinika za neurologiju, Klinički bolnički centar Zagreb
Kišpatićeva 12, 10 000 Zagreb; tel. +385 1 2388 310, fax: +385 1 2421 846
e-mail: dileme2008@net.hr

Registracija i prijava sažetaka: www.dileme2008.com

Kotizacija:

Do 1. srpnja 2008.	1500 Kn
Nakon 1. srpnja 2008.	2000 Kn
Za specijalizante	1000 Kn
Za osobe u pratnji	1000 kn
Za jedan dan kongresa	1000 Kn

Rok za slanje sažetaka: 1. srpnja 2008.